Guideline Supplementary Paper

New Zealand Autism Spectrum Disorder Guideline’s supplementary paper on the effectiveness of music therapy interventions for children and young people on the autism spectrum



With the support of the New Zealand Autism Spectrum Disorder  
Guideline’s Living Guideline Group

As at 9 April 2021

# 

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The work was researched and written by INSIGHT Research Ltd employees or contractors. Appraisal of the evidence, formulation of recommendations and reporting are independent of the Ministries of Health and Education.

**Statement of intent**

INSIGHT Research produces evidence-based best practice guidelines, health technology assessments and literature reviews to help health care practitioners, educators, policy-makers and consumers make decisions about practices in specific circumstances. The evidence is developed from systematic reviews of international literature and placed within the New Zealand context.

Guidelines, including supplementary papers, are not intended to replace a health practitioner’s judgement in each individual case.

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# About the evidence review

## Purpose

The first edition of the New Zealand Autism Spectrum Disorder Guideline (referred to henceforth as “the guideline”) was published in April 2008 [1]. As part of their commitment to the implementation of the guideline, New Zealand’s Ministry of Health and Ministry of Education agreed to establish a “Living Guideline process” in 2009. This process ensures that the guideline is regularly updated and refined to reflect new evidence and changing user needs.

A multidisciplinary advisory panel called the Living Guideline Group (LGG) are responsible for prioritising what topics should be updated. Updates to the guideline are required when the guideline’s recommendations are no longer valid in view of research that has emerged after the guideline’s literature searches were undertaken. For each topic, a systematic literature review is undertaken by INSIGHT Research which includes a critical synthesis of research published since 2004 (when the guideline’s original searches were conducted). The LGG consider the completed systematic review, and report on any implications for revising existing relevant guideline recommendations and good practice points as well as the potential for developing new ones. These topic updates which supplement the guideline, known as supplementary papers, have been produced annually since 2009 [2-11]. A second edition of the guideline was published in 2016 [12], incorporating revised and new recommendations and good practice points from the first seven supplementary papers.

The current supplementary paper updates the guideline with respect to the effectiveness of music therapy for people on the autism spectrum.

This supplementary paper and the entire living guideline process is co-funded by New Zealand’s Ministry of Health and Ministry of Education.

## Scope of the evidence review

The current review aims to update the guideline [12] with evidence relating to music therapy published since January 2004. The LGG identified this topic as an area worthy of updating and one which could lead to revised or additional recommendations for the guideline.

The review considered the effectiveness of music therapy interventions for people on the autism spectrum. Music therapy is defined as the planned use of musical experiences and the relationships that develop through them for therapeutic goals delivered through regular sessions by a music therapist. Included was music therapy when offered in multiple regular sessions to individuals or group, and included sub‑types such as family-centred music therapy and improvisation music therapy. Whilst no age restriction was applied, the results principally related to children and young people on the autism spectrum. Randomised controlled trials, cross-over studies (where condition order was randomised), and pseudo-randomised controlled studies were included, where more than 5 people were allocated to the music therapy condition.

This document should be read in the context of the guideline’s 2nd edition [12] and the guideline’s Supplementary Papers [2-11].

## Autism terminology

Autism Spectrum Disorder (ASD) is a condition that affects communication, social interaction and adaptive behaviour functioning. In the current edition of the diagnostic manual of mental disorders, the DSM-5 [13], four of the pervasive developmental disorder subcategories specified in the manual’s predecessor, the DSM-IV [14], are subsumed into one broad category of autism spectrum disorder. These subtypes are autistic disorder, Asperger’s disorder (Asperger syndrome), childhood disintegrative disorder (CDD), and pervasive developmental disorder not otherwise specified (PDD‑NOS). The name pervasive developmental disorder (PDD) was changed to Autism Spectrum Disorder (ASD).[[1]](#footnote-1) The diverse range of disability, learning needs and functioning expressed by people across the autism spectrum requires that a wide range of services and supports be employed to reflect the heterogeneity of the condition.

The guideline’s first edition [1] was prescient in recognising the movement toward considering autism as a spectrum condition. The term ASD is still used widely internationally, particularly by clinicians and researchers. However, many in the Autism Community are uncomfortable with the term ASD because the word “disorder” conveys a sense of autism as a medical problem rather than a reflection of neurological differences in the brain (or ‘neurodiversity’) [15]. Whilst an autistic person has support needs, autism itself “does not equal disability, disability is what someone experiences when they interact with a society that cannot reciprocate or accommodate them” [16].

To address these concerns, the acronym ASD has sometimes been defined as autism spectrum *difference* rather than *disorder*, or replaced by the term Autism Spectrum Condition (ASC). And some people have described themselves as ‘having autism’ or ‘having Asperger’s’. However there has been a shift from person-first to identify-first language in recent years such that people formally diagnosed with ASD or self‑diagnosed refer to themselves as being ‘autistic’, ‘autists’ or ‘Aspies’ rather than a ‘person with autism’. The deliberate choice of the inclusive term Autistic by the Autism Self-Advocacy Network recognises autism as a central part of their identity – of who they are, rather than as something separate to themselves, that can be cured or be put aside.

These changing preferences are reflected in a recent Australian study of 198 adults which found the terms ‘autistic’, ‘person on the autism spectrum’, and ‘autistic person’ rated as most preferred and least offensive, with ‘person on the autism spectrum’ ranked the most preferred term overall [17]. In New Zealand, this preference is also reflected in the use of the Māori term for autism, Takiwātanga. Takiwātanga is a derivation of the phrase “tōku/tōna anō takiwā” meaning “my/their own time and space”. Opai’s research (2020) suggests that the term tends to be used to refer to one’s identity – “I am takiwātanga” – rather than as a separate condition [18].

The living guideline process provides the opportunity for each supplementary report to incorporate changes in language that best reflect current community preferences. In this report, the terms ‘person on the autism spectrum’ or ‘autistic person” are used to refer to someone understood to have met criteria for the diagnosis of ASD. The acronym ASD is only used when referring to a person’s formal diagnosis, such as when used as a selection criteria in cited research studies.

It is understood that the term “high functioning” to describe more cognitively and verbally able groups of people on the autism spectrum is considered unhelpful and divisive by many on the autism spectrum. In this report, the term “high functioning” is only used when quoting specific inclusion criteria for appraised studies. In such studies, the term refers to people with higher cognitive ability either as established either by cognitive assessment (generally indicated by full-scale IQ scores of 70 or above), or through the diagnosis of “high-functioning autism,” or Asperger syndrome (under DSM‑IV criteria) [13]. It is acknowledged that these distinctions may no longer be used clinically in light of the removal of Asperger syndrome as a separate diagnostic classification in DSM-5 [6]. It is noted that DSM-5 utilises “specifiers” including whether or not the ASD is accompanied by intellectual impairment [6].

## Target audience

The systematic review that forms the bulk of this report aims primarily to provide an updated synthesis of research evidence on a specific topic for consideration by the Living Guideline Group. As such it is written in an academic style and is not intended for the general reader.

The systematic review informs the Living Guideline Group in revising and developing new recommendations and good practice points to update the New Zealand Autism Spectrum Disorder Guideline [12]. These outputs (detailed in **Part 3** of this paper) are intended for a broader audience, including the providers of professional health, education and support services for New Zealanders on the autism spectrum, as well for people on the autism spectrum themselves, their families, and whānau.

## Treaty of Waitangi

INSIGHT Research acknowledges the importance of the Treaty of Waitangi to Aotearoa/New Zealand, and considers the Treaty principles of partnership, participation and protection as central to improving Māori health and education.

INSIGHT Research’s commitment to improving Māori health outcomes means we attempt to identify points in the guideline or evidence review process where Māori health must be considered and addressed. In addition, it is expected that Māori health is considered at all points in the guideline or evidence review in a less explicit manner.

## Recommendation development process

The research topic was identified and prioritised by the LGG. A systematic review updating the published evidence was conducted by Marita Broadstock (INSIGHT Research) (**Part 1** and **Part 2**) and disseminated to the LGG as pre-reading for a one day, face-to-face meeting on 25 November 2020. At the meeting, the currency of the guideline was discussed in view of the updated evidence and specifically its implications for revising existing relevant guideline recommendations and good practice points and the potential for developing new ones. These are described, accompanied by the LGG’s rationale and additional notes, in **Part 3** of this paper.

INSIGHT Research follows specific structured processes for evidence synthesis. Full methodological details and a list of Living Guideline Group members is provided in **Appendix 1**. **Appendix 2** presents a [Glossary](#Glossary) of key epidemiological and topic-specific terms, abbreviations and acronyms. **Appendix 3** presents evidence tables of included studies for the current review update.

# Executive Summary

This reports supplements the New Zealand Autism Spectrum Disorder Guideline [1, 12] (‘the guideline’) by providing an update on the effectiveness of music therapy for children and young people on the autism spectrum.

## Scope

Music therapy is the planned use of music, musical elements and musical experiences and the relationships that develop through these to support the health or wellbeing of people who have social, communicative, emotional, intellectual, physical and spiritual needs.

The review considered music therapy offered in in multiple regular sessions to individuals or in groups, using a range of approaches. There was no restriction on type of music therapy or age of client. Study designs included randomised controlled trials, cross-over studies (where condition order was randomised), and pseudo-randomised controlled studies, where more than 5 people were allocated to the music therapy condition.

## Results

A broad search strategy of database and citation searching was undertaken of peer reviewed studies published between July 1, 2013 and 15 September 2020. These dates updated the search employed for a Cochrane Collaboration review [23] on the topic.

In the current review, 11 studies met selection criteria and were critically appraised using formal checklists. These included 4 systematic reviews and 7 primary studies (reporting on 6 trials) include data on a total of 537 participants on the autism spectrum, mostly primary school aged children.

From their meta-analysis [23] of 10 randomised controlled trials and cross-over trials, the Cochrane reviewers concluded that music therapy has moderate to large effects in improvements to social interaction within and beyond therapy, and in initiating behaviour within therapy, low to moderate improvements in social adaptation, and possible improvements in social-emotional reciprocity. Three subsequent systematic reviews similarly found promising evidence for music evidence improving social communication and social interaction in autistic children.

The primary studies identified in the current review add to the growing body of evidence that music therapy interventions can provide benefits across a number of domains for children on the autism spectrum, particularly with respect to social communication.

Heterogeneity in the research precluded conclusions about the most successful and necessary components, delivery, and duration of music therapy interventions.

Limitations of the research, gaps in understanding, and recommendations for future research are provided.

## Revisions to the Guideline based on the review

The Living Guideline Group (LGG), an expert advisory panel, presented their decisions on the implications of the updated body of evidence for the guideline. Their new recommendation supplements the guideline on this topic:

* **Recommendation 4.5.3:** “Music therapy can enhance social communication skills and should be considered for children and young people on the autism spectrum.” This recommendation is graded B (see **Summary Table**).

Summary Table: New recommendation relevant to music therapy and autism

|  |  |  |
| --- | --- | --- |
| **Reference** | **Revised recommendation** | **Grade** |
| 4.5.3 | Music therapy can enhance social communication skills and should be considered for children and young people on the autism spectrum | B |

**Note**: Grades indicate the strength of the supporting evidence rather than the importance of the evidence. Grade A indicates good evidence, B is fair evidence, C is international expert consensus, and I is insufficient, poor quality, or conflicting evidence. See **Table A1.2** in **Appendix 1** for details.

The LGG provided detailed **Rationale** for this recommendation in relation to the considered body of evidence and its reflection in the assigned grade of B (‘fair evidence’).

The LGG specified that the new Recommendation be referenced within Part 4 (Treatment and Management of ASD) of the guideline, within Section 4.5 relating to ‘Other interventions’. The LGG further advised that a cross-reference be provided in Part 3 (under *3.2.c Sensori-motor development)*, and in Appendix 8, where music therapy is discussed within a broader overview of educational interventions.

# Part 1: Introduction

## 1.1 Background

### Music therapy

Music therapists use music or musical elements to support the health or wellbeing of people who have social, communicative, emotional, intellectual, physical and spiritual needs [19-21]. A generally accepted definition is that therapeutic goals are sought “through musical experiences and the relationships that develop through them as dynamic forces for change” [22]. In sessions, musical activities are undertaken by the therapist and the client. These may variously include the use of percussion or tuned instruments, live music making, pre-recorded music, vocalisations, singing, composition, and/or improvisation. A client does not need to have any previous experience or skills in music. Techniques of music therapy may include [23-25]:

* *free improvisation* (where music is created without any stated boundaries)
* *structured improvisation* (where music is created within established musical parameters)
* *active music therapy* (music making where precomposed songs, music or activities performed or re-created)
* *music composition* (where songs and instrumental music are created)
* *receptive music therapy* (where the client engages in listening experiences).

There is a range of theories about how music therapy works, including behavioural, cognitive, phenomenological, humanistic and psychoanalytic schools [20]. The therapist is said to attune to the client both musically and emotionally so as to create periods of synchronisation, and this allows the client to understand and experience the reciprocal nature of communication [23]. The relationship developed between the therapist and client is therefore core to the approach.

Music therapists sometimes use ‘interventions’ (more typically termed ‘methods’ or ‘strategies’ in New Zealand practice) which require them to follow protocols, principles and procedures. The methods and/or strategies may be manualised in a systematic treatment plan, while still allowing therapists to respond flexibly to the client, adapting to their needs and the therapeutic situation as it presents itself in each session [23]. Less structured, relational approaches using improvisation are particularly client-led and client-centred. Therapists may use a non-directive approach, drawing on the musical interests of the client, as well as their immediate expression and behaviour, to enhance social engagement and the expression of emotions.

Therapy is usually given individually but can be group-based, peer-mediated, or family centred where family members are included in the therapy sessions [26]. It can occur in a range of settings (including homes, classrooms, music rooms, community settings, hospitals), and usually occurs on a regular and ongoing basis, for a year or more. It is sometimes used alongside other therapies, for example dance therapy, and in the creation of musical social stories.

Internationally, qualified music therapists hold a clinical training qualification at an undergraduate or graduate level in music therapy. Many are also required to register with an appropriate authority in order to practice. In New Zealand, Registered Music Therapists (NZ RMTh) must have approved masters’ level qualification in music therapy from a New Zealand university, or qualifications and experience assessed by the Registration Board as equivalent [19].

### Music therapy for people on the autism spectrum

Social communication is a core area affected in people on the autism spectrum. Music therapy seeks to exploit the potential for music to act as a medium for social communication [20].

Research with infants with developmental disabilities suggests that children can be sensitive to the melodic, rhythmic, harmonic, and dynamic dimensions of maternal speech and its emotional tone such that they are “born ready” for the “communicative musicality” of conversation [27]. By reflecting the back and forth nature of a conversation, music can engage the attention and involvement of a client, whether or not they are verbally communicative. And when applied systematically and skilfully by a trained music therapist, music can be used as a medium for engaging in non-verbal social exchange with people on the autism spectrum. This interaction between therapist and client becomes a form of communication at an emotional, relationship-oriented level, through music [28].

In relationship-based music therapy approaches, improvisation can be used to encourage skills common to music making and communication, including joint attention, affect sharing, eye contact, and turn-taking [23, 29, 30]. Following a contingency plan, a therapist can create flexible opportunities for reciprocal interactions through music making, attempting to attune or relate to the behaviours and responses of the client. A therapist can attempt to match the client’s mood or behaviour, follow their lead, and work to develop musical and emotional synchronicity [31].

Music therapists can use musical prompts to teach social and communicative norms as well as encourage them to be practised. Rhythmic and structural components of musical stimuli can provide external cues or anchors that support autistic individuals in organising, predicting, planning and responding to others around them [32]. For example, rhythmic cues can be used to aid in the initiation of a behaviour, lyrical cues can provide directives, and the form and structure of the music can provide anticipation for a response. Musical structure and repetition can therefore be used to both contain and facilitate emotional expression, and to support the development of emotional regulation, turn-taking, and social skills [33].

Given music’s universal appeal and intrinsic reward value, music therapy has been proposed as a strength-based approach for autistic people that draws on their particular skills, interests, preferences, and motivations [23, 34]. Many people on the autism spectrum, including both verbal and non-verbal communicators, have a preference for music and may have a particular affinity for it [34]. There may be also be a neurological basis to this. A study of autistic people identified stronger activations of their brains’ cortical speech and auditory areas when exposed to song compared with neurotypical children, suggesting that musical stimuli may more effectively engage people on the autism spectrum [35].

Music therapy has been used in clinical and research settings for supporting autistic people, particularly children, to develop social and emotional interactions, communication and expression, and to facilitate positive changes in behaviour and emotional wellbeing. In classroom and group settings, it has been used to support the development of relationships with peers, and in engagement in school routines and tasks of daily living [33]. Music therapy, particularly when involving family members, may also improve the quality of the parent-child relationship [36]. In 2006, a Cochrane Collaboration systematic review investigated the effectiveness of music therapy for children on the autism spectrum [37]. From three included studies, the review concluded that music therapy may help such children improve their communicative skills, and called for further research that investigated the approach in ways more typical of usual clinical practice, and for longer periods.

In New Zealand, music therapists are recognised as professional providers of services to children with ongoing difficulties via the Ministry of Education’s Ongoing Resourcing Scheme (ORS), either individually or through a school’s flexible use of funding. They may also be employed privately by a family. Whilst there is the potential to support adults, services are easier to access for younger children on the autism spectrum [33]. Music Therapy NZ is an organisational member of Allied Health Aotearoa New Zealand (AHANZ) along with other professions that are not regulated under the Health Practitioners Competence Assurance Act 2003.

## 1.2 Music therapy in the existing autism guideline

Music therapy is largely absent from the current New Zealand Autism Spectrum Disorder Guideline. There are currently no relevant recommendations or Good Practice Points related to the therapy. Music therapy is mentioned in the ASD Guideline’s [12] Appendix 8 under educational interventions (p. 300):

“Music therapy uses music in a planned and creative manner to promote good health and to address physiological, emotion, cognitive and social needs through the development of a therapeutic relationship [38]. Music therapy has also been promoted as an effective treatment in facilitating communication by offering a means by which alternative communication can be established to help achieve engagement, interaction and relationships [39].

At present, the evidence for the effectiveness of music therapy is unclear [39]. Standardised models of assessment in music therapy should be considered for future development. No large scale randomised control trial involving young children with autism has been conducted. With this level of evidence, broad claims about the universal effectiveness of music therapy for all children with autism must be met with caution and tested through studies with appropriate design and methodological rigour [38].”

## 1.3 The current review update

### Review objectives

The objectives of the current review update were to:

* Systematically identify, select, and narratively synthesise research studies published since January 2004 which evaluate the best evidence relating to the effectiveness of music therapy interventions for individuals on the autism spectrum.
* Consider this evidence as it supplements the guideline [12] in order to inform the LGG’s revision of any existing relevant recommendations/good practice points and/or the development of new ones.

# Part 2: Systematic review of effectiveness of music therapy for autistic children and young people

This chapter describes the findings of a systematic review relating to music therapy interventions for children and young people on the autism spectrum.

## 2.1 Scope and methods

Full details of review methods including search strategies, appraisal of study quality and data extraction are presented in **Appendix 1**.

### Research question

The review update’s primary research questions are:

1. What is the effectiveness of music therapy for individuals on the autism spectrum?
2. What are the features of an effective music therapy intervention for individuals on the autism spectrum?

### Identification and selection of studies for inclusion

The search strategy was initially limited to articles published from January 1, 2004, to ensure capture of articles published since the search was conducted for the original guideline [1]. A systematic search strategy used a combination of terms for autism and music therapy (see **Appendix 1** for details) to search titles, abstracts and subject fields of 11 bibliographic, health technology assessment, and guideline databases on 9–17 June 2020. Results were limited to those published in peer reviewed academic journals in the English language, and involving human participants. A total of 728 articles were originally identified.

From scoping, a high quality systematic review conducted for the Cochrane Collaboration [23] was identified as directly relevant and eligible for inclusion. This review updated an earlier Cochrane review cited in the Introduction (see **Part 1; 1.1**) [37]. In order to restrict the current review to consider the “best evidence” [40], the current review’s scope was therefore refined to update the literature search undertaken for the Cochrane review, thus limiting studies to those published from July 1, 2013. The search was updated to include articles published to 15 September, 2020. This process identified a set of 370 potentially relevant articles that were screened for eligibility. Inclusion and exclusion selection criteria (see **Table 2.1**) were applied to titles and abstracts to identify articles for retrieval, and then applied to retrieved full text, to identify studies for inclusion.

Table 2.1: Inclusion and exclusion criteria for selection of eligible studies

|  |  |
| --- | --- |
| **Characteristic** | **Inclusion criteria** |
| Publication type | Published in the English language in peer reviewed journals |
| Publication date | Published between 1 July, 2013 and 15 September, 2020 inclusive |
| Scope | Investigating the effectiveness of eligible music therapy interventions for people diagnosed with ASD |
| Participant characteristics | Participants diagnosed with ASD of any age in all comparator groups  Sample included at least 51% of participants diagnosed with ASD, or which had results synthesised and reported separately for a sub-group/s of participants diagnosed with ASD |
| Sample size | Sample included > 5 participants receiving music therapy |
| Intervention | Music therapy – the planned use of music to achieve therapeutic goals delivered by music therapist  Given in multiple, regular sessions individually or in groups |
| Comparator | Comparators were no intervention, placebo, or standard care  For studies comparing the relative effectiveness of components of music therapy programmes, comparators were alternative forms of music therapy |
| Outcomes | Primary outcome:   * Social communication * Social interaction * Initiating behaviour * Social-emotional reciprocity * Social adjustment * Communication skills (non-verbal and verbal)   Secondary outcome:   * Decreasing undesirable maladaptive behaviour, stereotypic behaviour * Social adaptation skills * Improving quality of life * Improving family relationships |
| Study design | Level 1 evidence: systematic reviews and/or meta-analyses that have a clear and relevant review question, use at least one electronic bibliographic database, report on eligible study population and intervention (solely or separately as a synthesised sub-group), and include at least one study meeting inclusion criteria for the current review (regardless of date of publication)  Level II evidence: randomised controlled trials; and controlled cross-over trials (including single case experimental design studies where condition order is randomised between individuals)  Level III.1 evidence: pseudo-randomised, controlled clinical trials |
| Analyses | Comparison of outcomes between subjects in comparison groups  Comparison of outcomes within subjects between comparison conditions.  Within a treatment group in an eligible study, analysis of the impact of potential moderator variables on effectiveness outcomes, including features of the intervention received, its dose, and characteristics of the participants |

**Table 2.1: Inclusion and exclusion criteria for selection of studies *(continued)***

|  |  |
| --- | --- |
| **Characteristic** | **Exclusion criteria** |
| Language | Non-English language articles |
| Publication type | The following were excluded   * correspondence, dissertations, book chapters, narrative/non-empirical articles, abstracts, unpublished data * single case reports, observational studies, uncontrolled before-and-after studies, case series, and cross-over studies where order of condition is not randomised |
| Scope | Studies which were not deemed relevant to the research question or nature of the review, including if they:   * were studies comparing autistic/neurotypical people with typically developing people * were animal studies, prenatal studies, genetic studies, brain studies, biomarker studies * evaluated interventions where music is a component but music therapy is not the core intervention * evaluated music education, where the prime goals are musical skills or improved musical performance * evaluated interventions where music is used incidentally as a tool for reinforcement or reward * evaluated interventions delivered solely via technology without the involvement of a music therapist * were studies describing the development of an intervention, outcome measure, scale or index |

Bibliographies of retrieved publications and recent narrative reviews were examined to identify any additional eligible studies. Narrative reviews retrieved for this purpose or to provide background material were not critically appraised or eligible for inclusion. Hand searching of journals and contacting of authors for unpublished research was not undertaken. Authors were contacted for clarification where needed.

### Publication type

Included were studies published in the English language in peer reviewed journals.

### Publication date

As referred to above, in order to update the search dates of the included Cochrane review identified in scoping [23] the publication dates of the search for the current review were limited to those published between 1 July 2013 and 15 September 2020.

### Scope

Included were studies where the key focus (ie, as a stated aim or in a significant representation of the results) was investigating the effectiveness of eligible music therapy interventions for people diagnosed with ASD.

### Participants

The study population were people of any age diagnosed with ASD. Whilst the guideline [12] defines autism spectrum disorder as classified by or consistent with DSM-IV [14] or DSM-5 [13] diagnostic criteria, studies were not limited by how the ASD diagnoses were identified. Studies of broader populations were included where results were reported separately for the eligible sub-group, or for samples of mixed disabilities, if at least 51% of its participants were reported as having been diagnosed with ASD.

### Sample size

The study included more than 5 participants receiving music therapy (ie, in the intervention group or in each comparator group for studies comparing music therapy programmes).

### Intervention

Included studies evaluated music therapy interventions. These are defined as involving the planned use of musical experiences and the relationships that develop through them for therapeutic goals, delivered through regular sessions by a music therapist. This definition is broadly consistent with those used in key systematic reviews on this topic [23, 24]. Interventions may be given individually, or in a group (eg, family, class, community).

### Comparator

The comparison/control condition could be no intervention, standard/usual care, or “placebo”. A placebo therapy attempts to control non-specific features of music therapy, such as the client’s motivation to participate or the attention of the therapist, without the musical element that is considered to be the active component of therapy. For example, a social story may be spoken instead of sung, or the same play activities may be used without music [23].

Comparators could also be alternative forms of music therapy that varied in a feature that is a critical component of treatment. This approach is relevant to answering the second research question and to determine the effectiveness of specific music therapy techniques and their contribution rather than music therapy *per se*.

### Outcomes

Included studies needed to report on at least one primary outcome which relates to social communication (including social interaction, joint attention, initiating behaviour, social-emotional reciprocity), as a core characteristic of autism, or communication.

Primary outcome:

* Social communication
* Social interaction
* Initiating behaviour
* Social-emotional reciprocity
* Social adjustment
* Communication skills (non-verbal and verbal)

Secondary outcome:

* Decreasing undesirable maladaptive behaviour, stereotypic behaviour
* Social adaptation skills
* Improving quality of life
* Improving family relationships

Data sources included published instruments and rating scales completed by participants, parent/care-giver, therapist, or researcher.

### Study designs

There was initially no restriction on study designs or sample size for primary studies, with the exception of the exclusion of single case reports.

However criteria relating to study designs was refined after the initial search was conducted in order to determine the level of evidence to be applied within the NHMRC hierarchy [40]. The goal was to identify “best evidence” representing the higher levels of the evidence hierarchy and only in their absence, include lower order evidence (see **Appendix A1.3** for further details).

Applying this approach, the selection criteria were refined to include:

* Level I evidence: secondary studies (systematic reviews and/or meta-analyses, including those informing clinical practice guidelines) where they had a clear and relevant review question, used at least one electronic bibliographic database, reported on the eligible study population and intervention (solely or separately as a synthesised sub-group), and included at least one primary study meeting other selection criteria for the current review (regardless of date of publication)
* Level II evidence: randomised controlled trials (RCT), and controlled cross-over trials including single case experimental design (SCED) studies where condition order is randomised
* Level III.1 evidence: pseudo-randomised, controlled clinical trials.

### Exclusions

Publications were **excluded** if they:

* were not published in the English language
* were correspondence, dissertations, book chapters, non-empirical articles (including editorials, commentaries, narrative reviews, expert opinion, book reviews), news reports, trade magazine articles, articles published only in abstract form, conference proceedings, poster presentations, unpublished data
* were case reports, observational studies, uncontrolled before-and-after studies, case series (including SCED studies where order of condition is not randomised between participants)
* were studies comparing autistic/neurotypical people with typically developing people (eg, assessing traits of autistic people using music therapy techniques)
* were animal studies, prenatal studies, genetic studies, brain studies, biomarker studies
* were studies describing the development of an intervention, outcome measure, scale or index
* evaluated interventions where music is a component but not the core intervention (including auditory integration therapy,[[2]](#footnote-2) sensory integration therapy, sound therapy, background music, dance therapy, movement based therapy, speech training using music, drama/theatre therapy, rhythmic therapy, Applied Behaviour Therapy, speech therapy)
* evaluated interventions where music is used incidentally (eg, as a tool for reinforcement or reward)
* evaluated interventions where music therapy cannot be isolated from other interventions (eg, musically accompanied dance therapy)
* evaluated music education, where the prime goals are musical knowledge, skills or improved musical performance
* evaluated interventions delivered solely via technology (eg, robot, cloud pillow, tablet/iPad, computer) without the involvement of a person trained in music therapy.

### Critical appraisal of included studies

Selection criteria were applied to titles and abstracts to identify articles for retrieval, and then to retrieved full text articles, to identify included studies.

Key characteristics and results of each study were entered into Evidence Tables (**Appendix 3**).

Study quality of included studies was formally appraised using the SIGN quality checklists from the Scottish Intercollegiate Guidelines Network [41] as appropriate to study design. The quality and resistance to risk of bias of an individual study was scored as either ++ (high quality), + (acceptable), or – (low quality).

Results are presented using numerical and thematic narrative syntheses.

Full details of review methods including search strategies, data extraction, and appraisal of study quality are presented in **Appendix 1**.

## 2.2 Body of evidence

### Overview

Following a comprehensive database search and citation searching of primary and secondary studies published between 1 July 2013 and 15 September 2020, 370 unique abstracts were identified. After applying inclusion and exclusion criteria, 11 studies were eligible for inclusion in the review: 4 secondary studies (systematic reviews and/or meta analyses), and 7 primary studies (reporting on data from 6 trials). The secondary studies include the Cochrane Collaboration review [23] which was used as a starting point for literature searching by the current review in order to update best available published evidence on the topic.

Detailed study attributes are presented in Evidence Tables (see [**Appendix 3**](#Appendix4)). These include: the country the study was conducted in, study design, evidence level and SIGN study quality rating (see **Appendix 1**), study aim, study setting, participant characteristics, selection criteria, procedure, outcome measures, results, authors’ conclusions, reviewer’s comments, and source of funding. For appraised systematic reviews and meta analyses, randomised controlled trials included therein that met selection criteria for the current review were are also identified to give an indication of the overlap of primary studies appraised in the secondary literature.

Summary characteristics for the included secondary studies are presented in **Table 2.2,** andincluded primary studies in **Table 2.3**, organised by year of publication (oldest first), and alphabetically by first author.

### Systematic reviews

Four secondary studies on the review topic were identified which included systematic reviews using selection criteria overlapping with the current review [23, 24, 42, 43].

Study characteristics are presented in **Table 2.2**.

#### Scope

Of the four secondary studies identified relevant to this topic published since July 2013, two were systematic reviews which included meta analyses [23, 43], and two were stand-alone systematic reviews [24, 42].

All but one review considered individuals of all ages on the autism spectrum in their searches; the exception focused on preschool-aged autistic children [43].

Two considered music therapies explicitly [23, 24]. One was a broad review of complementary and alternative medicine (CAM) interventions with a separate synthesis, table and discussion of studies relating to music therapies [42]. A fourth study was a meta-analysis of diverse interventions including cognitive, developmental, and behavioural approaches that included a narrative synthesis of music therapy studies.

#### Country

Review teams often included researchers from more than one country, but considering the leading author’s location, reviews were undertaken in Italy, Japan, New Zealand, and Norway.

#### Quality

Review quality was reasonably good, with three reviews rated (using the SIGN checklist) as being of acceptable quality (+), and the Cochrane review as high quality (++) [23].

Methodology strengths included broad search strategies, independent selection and extraction by multiple researchers, and detailed tabulated data of included studies. Limitations variously included a lack of appropriate appraisal checklists to assess methodological quality, lack of appropriate methodological critique, and limited descriptions of study design and results. Reviews were also impacted by the limitations of the evidence base including poorly controlled studies, small samples, and high rates of study attrition. Limitations of research in this area, including in the current review, are discussed further in **Section 2.4** of the current review.

#### Narrative summary of secondary studies

Studies are presented below in chronological order by date of publication, from oldest to youngest. Note that included systematic reviews had overlapping scope, search strategies, selection criteria, and therefore included primary studies. It is therefore important that review findings are not summated or given additional weight where the same studies are reported. Appraised trials relevant to the current review are cited in the evidence tables to illustrate this overlap.

##### Geretsegger et al (2014) for the Cochrane Collaboration

The systematic review and meta-analysis from the Cochrane Collaboration (2014) [23] (rated as ‘high quality’) had a scope closest to the current review with respect to interventions, study designs and outcomes. The 10 eligible studies included young children aged 2 to 9 years across 4 RCTs [26, 44-46], 5 cross-over trials where order of condition was randomised [47-51], and one pseudo-randomised trial described as “counter-balanced”, where the process for randomisation was unclear [52]. Sample sizes ranged from 4 to 50 participants, with a total of 165 participants represented across the 10 included studies. As the current review updates research published since the Cochrane review’s comprehensive search strategy, a summary of the 10 unique randomised and pseudo-randomised controlled trials identified are presented in **Table 2.3** and **Table 2.4** so that these can be considered in the current review’s body of primary evidence. Note that these studies have not been formally appraised in the current review.

The analysis suggested that there was *moderate to large* improvement in social interaction (measured within, and generalised beyond the therapy context) and initiating behaviour (within therapy), and *low to moderate* improvement in verbal communication (within therapy) and social adaptation. However, there was no significant improvement for those receiving music therapy compared to placebo or standard care for verbal communication outside of therapy or non-verbal communication (within, or beyond therapy). Whilst improvements were identified for social-emotional reciprocity (within therapy), joy, and the quality of parent-child relationships, these results should be taken with caution due to high rates of attrition and/or low sample sizes.

##### Brondino et al (2015)

More recently, a systematic review (rated as ‘acceptable quality’) from Italy by Brondino et al (2015) [42] considered music-associated approaches within a broader review of complementary and alternative medicine (CAM). The review identified 15 primary studies, including an RCT appraised in the current review [53]. Thematic analysis concluded that, as well as being safe, there is promising evidence that supports the use of music therapy in children on the autism spectrum with respect to improvements in communication, social reciprocity, and emotion.

##### James et al (2015)

In the same year a team led from New Zealand, James et al (2015) [24], conducted a systematic review (of ‘acceptable quality’) of music therapy for individuals on the autism spectrum. The review included group studies, single case experimental design studies, and before and after studies. Of 12 included studies, the authors suggested 7 provided conclusive evidence (based on methodological quality and findings) of positive outcomes for music therapy. Similar to Brondino et al (2015) [42], the reviewers concluded that music therapy is a promising practice for individuals on the autism spectrum and for specific purposes.

##### Su Maw and Haga (2018)

Most recently, Japanese researchers Su Maw and Haga (2018) [43] undertook a systematic review and meta-analysis (of ‘acceptable quality’) of randomised controlled trials investigating cognitive, developmental, and behavioural interventions for pre-school children on the autism spectrum. Of 14 identified trials, the three studies that evaluated music therapy were some of the most effective. It was noted that this was despite one of the studies investigating an intervention of the shortest duration and lowest intensity of all the appraised trials. That particular Australian trial evaluated family-centred music therapy intervention which was delivered in sessions of 30-minutes per week, over 4 months [31]. The three music therapy trials were all included in the Cochrane review [23]. The reviewers concluded that music therapy appears to be an effective tool for improving social interaction in pre-school aged children on the autism spectrum.

Table 2.2: Characteristics of secondary studies

|  |  |  |  |  |  |  |
| --- | --- | --- | --- | --- | --- | --- |
| **Author/s (year)** | **Country** | **Evidence level, SIGN rating** | **Population** | **Intervention** | **Search strategy and analysis** | **Results/Recommendations** |
| Geretsegger et al (2014) [23] | Norway | I  ++ (high quality) | Individuals diagnosed with ASD | Music Therapy | Comprehensive search strategy  Three researchers independently applied eligibility criteria, and two extracted data and appraised studies, and applied the risk of bias tool.  Meta-analysis | 10 studies: 4 RCTs, 5 randomised cross-over trials, and 1 pseudo-randomised controlled trial [31, 44-52]  Music therapy may improve autistic children’s skills in social interaction, verbal communication within therapy, initiating behaviour, social-emotional reciprocity, social adaptation.  Music Therapy may promote the quality of parent-child relationships.  No significant improvements to verbal or non-verbal communication beyond therapy. |
| Brondino et al (2015) [42] | Italy | I  + (acceptable quality) | Individuals diagnosed with ASD | Complementary and alternative medicine (CAM), including Music Therapy | Broad search strategy  Two researchers independently applied eligibility criteria.  Narrative analysis, no appraisal tool used. | 15 music therapy studies, including 10 appraised in Geretsegger et al (2014) [23], and 1 RCT [53] appraised in current review (4 non relevant).  Concluded that music therapy is a promising intervention, with evidence that use of music improves communication, social reciprocity, and emotion, and is extremely safe. |
| James et al (2015) [24] | New Zealand | I  + (acceptable quality) | Individuals diagnosed with ASD | Music Therapy | Broad search strategy  Two researchers independently applied eligibility criteria, two extracted data, and applied certainty of evidence criteria.  Narrative analysis, tables lacked some key data. | 12 studies, including 5 appraised in Geretsegger et al (2014) [23] (and 7 non relevant).  Concluded that music therapy is a promising practice for individuals on the autism spectrum and for specific purposes. |
| Su Maw & Haga (2018) [43] | Japan | I  + (acceptable quality) | Pre-schoolers aged 1-6 years diagnosed with ASD | Cognitive, developmental, behavioural approaches, including Music Therapy | Broad search strategy  Two researchers independently applied eligibility criteria. One extracted data and applied the risk of bias tool, confirmed by second reviewer.  Narrative synthesis. Table data lacked key information. Meta-analysis performed. No sensitivity analyses. | 3 music therapy, all appraised in Geretsegger et al (2014) [23]  Concluded that music therapy appears to be an effective tool for improving social interaction in pre-school children on the autism spectrum. |

**Key**: ASD=Autism Spectrum Disorder; CAM=complementary and Alternative Medicine; RCT=randomised controlled trial; SIGN=Scottish Intercollegiate Guidelines Network

Table 2.3: Characteristics of controlled trials in the appraised Cochrane review

|  |  |  |  |  |  |  |  |
| --- | --- | --- | --- | --- | --- | --- | --- |
| **Author/s (year)** | **Design** | **Country** | **Sample, gender** | **Mean age (range)** | **Intervention** | **Comparator** | **Duration of activity** |
| Arezina (2011) [47] | Randomised cross-over trial\*\* | US | N=6 83% male | 3–5 years | Interactive MT | Non interactive music play | Over 5 weeks: 6 sessions x 10 min (1 hour) |
| Brownell (2002) [52] | Pseudo-randomised cross-over trial\*\* | US | N=4 100% male | 6–9 years | Structured receptive MT (sung social stories) | Story therapy (read social stories) | Over 4 weeks: 5 sessions |
| Buday (1995) [48] | Randomised cross-over trial\*\* | US | N=10 80% male | 4–9 years | Structured receptive MT (songs to teach signs) | Rhythm therapy | Over 2 weeks: 5 sessions |
| Farmer (2003) [44] | RCT | US | N=10 90% male | 2–5 years | Active and receptive MT (guitar playing, songs) | Placebo | Over 1 week: 5 sessions x 20 min (1 hour 40 min) |
| Gattino (2011) [45] | RCT | Brazil | N=24 100% male | M=10 years (7–12 years) | Relational MT (improvised) | Standard care | Over 7 months: 12 sessions x 20 min (4 hours) |
| Kim et al (2008) [49] | Randomised cross-over trial\*\* | Korean | N=15 87% male | M=4 years (3–6 years) | Relational MT (improvised) | Play sessions with toys | Over 8 months: 12 sessions x 30 min (6 hours) |
| Lim (2010) [46] | RCT | US | N=50 88% male | M=5 years (3–5 years) | Music training (videotaped songs with target words) | Speech training | Over 5 days: 6 sessions |
| Lim & Draper (2011) [50] | Randomised cross-over trial\*\* | US | N=22 77% male | M=4 years (3–5 years) | Music training (songs with target words) | Speech training | Over 2 weeks: 6 sessions |
| Thomas & Hunter (2003) [51] | Randomised cross-over trial\*\* | US | N=6 83% male | 2–3 years | MT (songs, instruments, vocalisations, movement) | Play-time | Over 12 weeks: 12 sessions x 15 min (3 hours) |
| Thompson et al (2012, 2013) [26, 31] | RCT | Australia | N=23 82% male | 3–6 years | Family-centred MT (songs, improvisation) | Standard care | Over 16 weeks: 16 sessions |

**Key**: M=mean; MT=music therapy; N=sample size (in intervention & control groups); RCT=randomised controlled trial (with parallel groups); US=United States of America.

\* significant improvements in the group receiving the intervention compared with those in the control group

\*\* session order randomised

Table 2.4: Summary of findings for controlled trials in the Cochrane review

|  |  |  |  |
| --- | --- | --- | --- |
| **Outcomes (Measures)** | **Relative effect (95% CI)** | **Number of participants (studies)** | **Quality of the evidence (GRADE)** |
| Social interaction – Generalised (outside sessions, daily life)  (CARS, PDDBI, Vineland SEEC, SRS) | The mean social interaction – generalised (outside sessions, daily life) in the intervention groups was 0.71 standard deviations higher (0.18 to 1.25 higher) | 57 (3 studies) [25,43,47] | moderate |
| Communicative skills: non-verbal – Generalised (outside sessions, daily life) (CARS, ESCS, MBCDI-W&G) | The mean communicative skills: non-verbal – generalised (outside sessions, daily life) in the intervention groups was 0.48 standard deviations higher (0.02 lower to 0.98 higher) | 57 (3 studies) [42,46,47] | low |
| Communicative skills: verbal – Generalised (out- side sessions, daily life) (CARS, MBCDI-W&G) | The mean communicative skills: verbal – generalised (outside sessions, daily life) in the intervention groups was 0.30 standard deviations higher (0.28 lower to 0.89 higher) | 47 (2 studies) [25,43] | low |
| Initiating behaviour – Non-generalised (requesting – initiating joint attention; limitation of engagement frequency, requesting behaviour) | The mean initiating behaviour – non-generalised in the intervention groups was 0.73 standard deviations higher (0.36 to 1.11 higher) | 22 (3 studies) [45,47,49] | moderate |
| Social-emotional reciprocity – Non-generalised (emotional and musical synchronicity, frequency, and duration) | The mean social-emotional reciprocity – non- generalised in the intervention groups was 2.28 standard deviations higher (0.73 to 3.83 higher) | 10 (1 study) [47] | low |
| Social adaptation – Non-generalised (interaction – engaging in joint attention; compliant or non-compliant response frequency, no response frequency, on-task) | The mean social adaptation – non-generalised in the intervention groups was 1.15 standard deviations higher (0.69 to 1.61 higher) | 22 (3 studies) [45,47,49] | moderate |
| Quality of parent-child relationship (MPIP, PCRI) | The mean quality of parent-child relationship in the intervention groups was 0.82 standard deviations higher (0.13 to 1.52 higher) | 33 (2 studies) [25,47] | moderate |

Note: results are for comparisons between Music Therapy and placebo or standard care comparators [22]

**Key**: See **List of Acronyms** for Scale names

### Primary studies

Seven primary studies (reporting on six trials) were included in the current review, published since (and therefore excluded from) the search strategy completed for the appraised Cochrane review (ie, since July 1 2013). These are randomised and pseudo-randomised controlled trials evaluating music therapy interventions for individuals diagnosed with ASD [20, 32, 34, 53-56]. Study characteristics are presented in **Table 2.5** and **2.6** according to their relevance to research question 1 (effectiveness of music therapy) or research question 2 (effective features of music therapy) respectively.

#### Sample characteristics

There were 532 participants across the 6 trials. Sample sizes for the studies averaged 88.6 people, ranging from 17 to 364 individuals on the autism spectrum. The largest sample (n=364) came from the 10-site Time-A trial [23]. Additional outcomes and analyses were reported for 81 children at the UK site of the trial [55].

All of the 6 appraised primary trials related to school-aged children recruited from mainstream or special education schools, with ages ranging from 4–21 years, and mean age (where reported) ranging from 5–15 years. There were a majority of males, averaging at 78% (range: 44% – 97%), broadly reflecting the 4:1 male to female ratio that is commonly seen in studies of ASD prevalence [57].

The primary studies were conducted in a diverse range of countries, including the United States of America (n=2), United Kingdom (n=1), Canada (n=1) and France (n=1), whilst the multi-site Time-A trial was conducted across 9 countries (Australia, Austria, Brazil, Israel, Italy, Korea, Norway, UK, US).

#### Quality

Whilst inclusion criteria restricted studies to randomised controlled trials, the methodological quality varied, with one trial rated using the SIGN checklist as being of high quality [34], 4 trials (5 studies) as of acceptable quality [20, 32, 54-56], and one trial as being low quality [53].

#### Interventions

Music therapy was delivered individually in three trials/four studies [34, 54-56] and to groups of participants in three studies [20, 32, 53]. One study’s intervention involved the therapist singing a social story. Five studies’ interventions involved participants performing using instruments and/or voice/singing, including two which also involved movement. Of the five interventions involving participants in music performance, two explicitly referred to the use of cueing by the music therapist, and three referred to the use of improvisation.

The comparators for these trials included usual care [56], enhanced standard care [53, 54], and in four studies, active comparators. These were a read (rather than sung) social stories [53], a group exercise and game session without music [32], a play‑based intervention [33], and music listening [20].

Intervention programmes varied widely in duration and intensity, involving between 3 and 25 sessions, each of 30 to 50 minutes, one to three times per week, for between one week and 25 weeks (spread over 8 months). The total intensity/duration ranged from the shortest of about 1.5 hours (approximately, over 3 sessions on consecutive days) for the social story trial [53] and the greatest of 12 to 12.5 hours for both the Time-A multi-site trial [54, 55] and the French trial [20].

#### Outcomes

Assessments were made at baseline (pre-test), and shortly following the intervention/ control (post-test). There was longer follow-up reported for three studies, including after an additional 5 weeks [32], 13 weeks [56], and in the TIME‑A multi-site trial, 7 months [54].

Primary outcomes included parent-rated measures of social interaction in 3 trials, including through subscales of the Social Responsiveness Scale (SRS) in two [34, 54] and subscales of the Autism Social Skills Profile (ASSP) in another trial, including social reciprocity [53]. Joint attention and eye gaze were measured by clinician assessments in one study [32]. Other primary outcomes related to communication skills (non-verbal and verbal) measured in two studies using measures such as parental report of the Children’s Communication Checklist (CCC) [34], a clinical assessment of vocabulary (PPVT) [34], and the Social Skills Improvement System Rating Scales (SSIS) completed by young participants and their parents [56].

Global assessments of autistic functioning or severity of symptoms (autism characteristics) were used in 4 trials using scales completed by clinicians including the Autism Diagnostic Observation Schedule (ADOS-social affect) in the TIME‑A trial [54], the Childhood Autism Rating Scale (CARS) [20], the Clinical Global Improvement scale (CGI) [20] and the Autism Treatment Evaluation Checklist (ATEC) [32], or through parental assessments of the SRS’s total score [34].

Also reported were several secondary outcomes. Two trials assessed maladaptive behaviour according to the parental report on the Vineland Adaptive Behavior Scales (VABS-MB) [34], or clinician assessments of the Aberrant Behavior Checklist (ABC) [20]. Quality of life was assessed in two trials relating to parental assessments of family relationships using the Beach Family Quality of Life (FQOL) [34], and visual analogues scales to assess the parent’s perceived quality of their child’s and the own quality of life [54]. The UK site within the Time-A trial assessed parental stress (using the PSI) and mental wellbeing on the Warwick-Edinburgh Mental Well-Being Scale (WEMWBS) [55].

Other measures included magnetic resonance imaging (MRI) assessments of brain connectivity on the Resting state functional connectivity (RSFC) [34], and comprehension of social stories using Comprehension Checks (CC) [53].

Table 2.5: Characteristics of primary studies comparing music therapy to control/comparator (research question 1)

|  |  |  |  |  |  |  |  |  |  |  |
| --- | --- | --- | --- | --- | --- | --- | --- | --- | --- | --- |
| **Author/s (year)** | **Design** | **Country** | **SIGN rating** | **Sample, gender** | **Mean age (range)** | **Intervention** | **Comparator** | **Duration of activity** | **Improved outcomes (Measure) (*assessor*)** | **No difference (Measure) (*assessor*)** |
| LaGasse et al (2014) [32] | RCT | US | Acceptable quality ( + ) | N=17 76% male | M=8 years (6–9 years) | Group-based MT   * musical cueing of sensory and social experiences, playing instruments, movement | Exercise (games, ball passing, copying movements) without music | Over 5 weeks: 10 sessions x 50 minutes (8.3 hours) | * Social skills (SRS) *(Parent)* * Joint attention with peers (*Clinician*) * Eye gaze (*Clinician*) | * Joint attention with adults (*Clinician*) * Autism functioning (ATEC) (*Clinician*) |
| Porter et al (2017) [56] | Multi-centre RCT | UK | Acceptable quality ( + ) | N=34 44% male (Note 1) | M=NR (8–16 years) | Individual Alvin model   * improvisation, voice, playing instruments, movement | Usual care | Over 12 weeks: 12 sessions x 30 minutes (6 hours) |  | * Communicative and interactional skills (SSIS) *(Participant)* * Communicative and interactional skills (SSIS) *(Parent)* |
| TIME‑A Trial (2017) [54, 55] | Multi-centre RCT | Australia, Austria, Brazil, Israel, Italy, Korea, Norway, UK, US | Acceptable quality ( + ) | N=364 83% male | M=5 years (4–7 years) | Individual MT   * improvisation, voice, playing instruments | Enhanced standard care (with 3 parent information sessions) | Over 5 months: Median 24 sessions x 30 minutes (12 hours) (Note 2) | * Proportion responders in autism symptom severity (ADOS-social affect) *(Clinician)* * Social motivation (SRS subscale) *(Parent)* (Note 3) | * Autism symptom severity (ADOS-social affect) *(Clinician)* * Social awareness, social cognition, social communication, autistic mannerisms (4 x SRS subscales) *(Parent)* * Quality of life of participants and parents (visual analogue scale) *(Parent)* * Parental stress (PSI) *(parents)* (Note 4) * Parental wellbeing (WEMWBS) *(parents)* |

*Continued next page*

**Table 2.5: Characteristics of primary studies comparing music therapy to control/comparator (research question 1)** *(continued)*

|  |  |  |  |  |  |  |  |  |  |  |
| --- | --- | --- | --- | --- | --- | --- | --- | --- | --- | --- |
| **Author/s (year)** | **Design** | **Country** | **SIGN rating** | **Sample, gender** | **Mean age (range)** | **Intervention** | **Comparator** | **Duration of activity** | **Improved outcomes (Measure) (*assessor*)** | **No difference (Measure) (*assessor*)** |
| Sharda et al (2018) [34] | RCT | Canada | High quality ( ++ ) | N=51 48% male | M=10 years (6–12 years) | Individual MT   * playing instruments, singing, rhythm cues | Play-based intervention | Over 8–12 weeks: M=10 sessions x 45 minutes (4.8 hours) | * Communication (CCC) *(Parent)* * Family quality of life (FQoL) *(Parent)* * Brain connectivity changes (RSFC) *(MRI)* | * Autism symptom severity (SRS) *(Parent)* * Receptive vocabulary (PPVT) *(Clinician)* * Maladaptive behaviour (VABS-MB) *(Parent)* |
| Rabeyron et al (2018) [20] | RCT | France | Acceptable quality ( + ) | N=36 88% male | M=5 years (4–7 years) | Group-based MT   * playing instruments and vocal improvisation | Music listening | Over 8 months: M=25 sessions x 30 minutes (12.5 hours) | * Autism symptom severity (CGI) *(Clinician)* * Non-adaptive behaviour (ABC-total score) *(Clinician)* * Lethargy, and stereotypy (2 x ABCsubscales) *(Clinician)* | * Autism symptom severity (CARS, and 15 subscales) *(Clinician)* * Irritability, inappropriate speech, and hyperactivity (3 x ABCsubscales) *(Clinician)* |

**Note 1**: estimated from larger study sample; **Note 2:** Treatment group combined high intensity (3 per week, median 34 sessions, 17 hours) and low intensity groups (1 per week, median 15 sessions, 7.5 hours) (See **Table 2.6** for analyses between active treatment groups); **Note 3**: When assessed at 5 month post baseline (immediate post-test). **Note 4:** In the UK site only, PSI and WEMWBS assessed at 5 month post-baseline.

**Key**: CG=control group; M=mean; MRI=magnetic resonance imaging; MT=music therapy; N=total sample (randomised participants in intervention and control groups); SIGN=Scottish Intercollegiate Guidelines Network; UK=United Kingdom; US=United States of America; See text description, and List of Acronyms, for Scale names.

#### Narrative summary of primary studies

The six clinical trials (reported across 7 primary studies) are narratively summarised below. **Tables 2.4** and **2.5** presents key characteristics for each trial, including outcomes where there are significant differences for people receiving music therapy compared with the comparator, and non-significant differences.

##### Schwartzberg et al (2013)

A US based study [53] provided music therapy sessions on three consecutive days to 30 youth on the autism spectrum attending a summer camp. The study was assessed as being ‘low quality’ using the SIGN checklist. A feature of MT was varied between the treatment and control conditions: whether a social story was sung or read, therefore making the study relevant to the second research question investigating effective features of music therapy (see **Table 2.6**). In this pseudo-randomised study, participants were randomised as clusters into six music therapy groups to receive one of three different versions of social stories (targeting different aspects of social skills) in either sung (intervention) or read (control) conditions. From 107 youth initially recruited, only 30 completing pre-test and post-test assessments were included (aged 9-21, M=16 years). Baseline assessments were not compared between groups, or between completers and those dropping out, making it difficult to rule out allocation and participation biases.

The study found no significant difference in assessed (unblinded to condition) social skills for the *sung* social story groups compared with the *read* story groups. Comprehension of stories increased for both groups over time, without any benefit for the musically delivered social story condition. It appears that whether the feature of social stories was musically delivered or not has not made a significant difference to outcomes in this study. However impact may have been limited by the brevity of the intervention, and the lack of an inactive control group without music therapy.

##### LaGasse et al (2014)

Another study from the US in the same year [32] compared music therapy groups with social skills groups. The small-sampled randomised controlled study (rated as ‘acceptable quality’) included 17 children (9 in the MT group), aged 6 to 9 years (M=8 years). Ten 50-minute sessions were given over 5 weeks with follow-up assessment  weeks post-intervention. The approach followed a manualised intervention based on a Transformational Design Model to create music experiences that were functionally similar to non-musical experiences. For the music group, music and rhythmic structure were used as anticipatory cues to facilitate desired social skills, and engagement with music (playing an instrument, moving to music) was used to practice these skills. By contrast, in the Social Skills group the social skills were encouraged through non-musical tasks such as playing card games, passing a ball, and movement without music.

The Music Therapy group were rated by parents (unblinded to condition) as having improved social behaviours after the programme compared with before, unlike the Social Skills group. A measure of autism functioning improved similarly over time regardless of group allocation or whether assessed by parent or therapist (blinded to condition). Video analysis of rated behaviours, blinded to stage of assessment but not condition, suggested that music therapy may improve joint attention to peers, and eye gaze. There were no clear group differences for communication, joint attention to adults, or withdrawal behaviours, although there was high variability between individuals. Intention to treat analyses were not conducted, excluding all data for  children (18%) dropping out or not attending at least 9 of the 10 sessions. The small sample may have been under-powered to pick up group differences, and any differences may have been masked by the social skills group itself being an effective approach.

##### Porter et al (2017)

A multi-centre randomised controlled trial [56] (rated as ‘acceptable quality’) in Northern Ireland evaluated the effectiveness of music therapy for young people aged 8 to 16 years recruited from six Child and Adolescent Mental Health Service (CAMHS) community care facilities. Within this broader study of children with social, emotional, behavioural and developmental difficulties, analyses were reported on a sub-group of young people on the autism spectrum. In this sample, 18 participants received 12 sessions of one-to-one music therapy and usual care, and 16 received usual care alone (counselling and/or medication). The manualised Alvin Model of improvised music therapy was followed where clients are encouraged to create music through their voices, movement and instruments to express their feelings and responses, with tailored support from the Music Therapist.

The primary outcome was a measure of communicative and interactional skills, assessed in reports by the participants, and their parents, and recorded by a researcher blinded to condition. Intention to treat analyses identified no differences between groups. The researchers argue that the lack of a significant effect of MT in this trial for autistic young people may be due to the study being underpowered. There were significant drop-outs at post-study assessment, particularly for the treatment group (33% attrition), despite reportedly high participation in the therapy programme itself. The study authors’ argument that some young people who benefited from therapy may have been less likely to be motivated to complete post-test assessments, thus biasing results against finding a treatment effect, seems unconvincing.

##### TIME‑A Trial (2017)

The TIME‑A Trial [54, 55] was a major international randomised clinical trial conducted across 10 centres and 9 countries. It included 364 autistic children aged 4 to 7 years randomly allocated to either a treatment group receiving individual improvisational music therapy (IMT) for 5 months, or a control condition. Those allocated to the music therapy arm were further randomised to receive either high-intensity (three 30-minute sessions per week, median 34 sessions) or low-intensity (weekly sessions, median 15 sessions) IMT. This aspect allowed the second research question of the current review to be explored in relation to assessing whether treatment intensity was a feature of MT that impacted on effectiveness. Both groups received enhanced standard care which consisted of three “counselling sessions” for parents where concerns around autism were discussed and information provided. This was in addition to the child’s usual concomitant therapies (eg, speech and language therapy, sensory-motor therapy, etc).

The primary outcome of the study was assessor-blinded autism symptom severity (and specifically, social affect) on the Autism Diagnostic Observation Schedule (ADOS). In intention-to-treat analyses in this pragmatic trial, there was no difference in this outcome between the intervention and control groups at initial follow-up (5 months post baseline, which was immediately post treatment). This result relates to the review’s first research question investigating effectiveness of music therapy (see **Table 2.5**).

Exploratory additional analyses of parent-rated, secondary outcomes found no significant group effects immediately post intervention for 4 of 5 subscales relating to social communication (SRS), or for two parent-rated visual analogue scales reporting on quality of life of participant, and of family (see **Table 2.5**). A subset of 81 children from two UK-based centres [55] received two parent-report measures assessing parental stress and parental mental wellbeing with 49% response rate (see **Table 2.5**) with no significant differences at 5 months post baseline.

With respect to the second research question investigating effective features of music therapy (see **Table 2.6**), nor were there dose effects, with no difference for either high‑intensity, or low-intensity IMT treatment groups, when compared with controls on this outcome. In post-hoc analyses, there was a higher proportion of ‘responders’ (moving one point on the symptom subscale) in the IMT group than the control group, particularly for those receiving at least 15 sessions. In exploratory analyses, there were no significant differences between high-intensity, low-intensity and control groups immediately post intervention for any of the SRS social communication subscales, quality of life of family, or (in the UK site) parental wellbeing or parental stress. There was a small improvement in parent-rated quality of life of participant for the high‑intensity group compared with control group. However, there were different rates of attrition between groups which may have biased results, and the authors advise treating this finding with caution.

Together the few significant effects for unblinded outcomes were not adjusted for multiple comparisons, and so may be chance effects, or artefacts of reporting and response biases. The researchers of this trial (rated as ‘acceptable quality’) concluded that the findings do not support the use of improvisational music therapy for symptom reduction in children on the autism spectrum.

Rather than the music therapy not being successful, an alternative interpretation suggested by the American Music Therapy Association is that the music intervention did as well as the enhanced care arm of the trial, which represented treatment options including speech and language therapy, communication training, and parent counselling [58]. This raised the bar in terms of symptom improvement for the enhanced standard care group, whose participants received more hours of these usual therapies compared with the MT group. It is possible that the effort and travel involved in attending regular music therapy sessions, particularly for the high intensity group, hampered access to and benefit from the enhanced standard care, leading to comparable improvement between groups.

It is also possible that music therapy led to benefits that the measures employed were not sensitive to picking up. Qualitative research from this trial [59] suggested that the improvisational music therapy was well received, enjoyed and perceived as positive. Designed as a diagnostic tool, the focus on a global measure of autism symptom severity (the ADOS) as a primary outcome measure has been questioned. As the researchers themselves noted, it may be too generalised, distal and blunt to detect changes from an intervention targeting specific behaviours including affect sharing and joint attention. The secondary outcome was also a measure of autism symptom severity, the Social Responsiveness Scale (SRS). Such measures may not capture changes in other clinical, developmental, functional and family domains that can be targeted by music therapy services, and which may be of greater value for children on the spectrum and their parents [58]. It has been argued that outcomes relating to wellbeing and adaptive functioning may matter more to people on the autism spectrum than symptom severity [15]. Limitations around outcome measures are further discussed under Review Limitations (see **Section 2.4**).

##### Sharda et al (2018)

A Canadian randomised controlled trial [34] (rated as ‘high quality’) compared the effects of 8 to 12 weeks of weekly child-centred music therapy (involving songs, instruments and rhythmic cues) with a play-based control condition. Participants were 51 primary school aged children aged 6 to 12 years, half of whom had some language impairment.

Compared with the play-based group, the music therapy group demonstrated improved social communication (including in speech, semantics, inappropriate initiations, and social relations), and family-related quality of life (particularly in terms of family interaction, cohesion, coping, and disability-related supports) in assessments made by parents blinded to condition (though 60% of them had become aware of group allocation). There was no longer term follow-up to see if these effects were maintained over time. There were no differences between groups for symptom severity, vocabulary, or maladaptive behaviour.

In addition to behavioural measures, this study also assessed brain connectivity through MRI scans, finding changes in the music therapy group such that there was greater connectivity between regions usually associated with either under-connectivity in people on the autism spectrum (ie, between auditory and motor regions) and reduced connectivity in areas usually associated with over-activity in autistic people (between auditory and visual association regions). These changes were associated with behavioural improvements in social communication. The researchers suggest that music therapy may improve social functioning through changes to brain networks.

##### Rabeyron et al (2018)

Most recently, a French RCT [19] investigated the impact of group-based improvisational music therapy compared to music listening for 37 young children aged 4 to 7 years of low cognitive ability (IQ range 49-62, M=52). There appeared to be one music therapy group, and one active control music listening group. The therapy was offered in 25 half-hour sessions over an 8 month study period. Fidelity was not reported but therapists reported finding it difficult to remain neutral during the music listening group, and so the possibility of performance bias occurring cannot be excluded, which would reduce the chance of group effects. The groups were similar at baseline with the exception that the music therapy group had more girls than the control group (4 c.f. 1). All analyses were controlled for gender.

In repeated measures regression analyses, the music therapy group showed greater improvements at post-treatment assessment for autism symptom severity measured on the Clinical Global Impression–Improvement scale (CGI-I), with a large effect size (*d*=0.80). Nearly two-thirds (63%) of children receiving music therapy improved by 2 points on the CGI-I compared with 29% of those in the music listening control group. However, no differences were found in “autistic symptoms” on the Childhood Autism Rating Scale (CARS) total score or any of its 15 subscales. After controlling for gender, those receiving music therapy had greater reductions in non-adaptive behaviour on the Aberrant Behavior Checklist (ABC) total score at post-test than the music listening control group in both ABC total score, and in two ABC subscales (stereotypy, and lethargy), but not for the subscales of irritability, inappropriate speech, or hyperactivity.

The researchers suggest that the lack of an effect of music therapy on the CARS is similar to the lack of a difference found for autistic symptoms on the ADOS in the TIME‑A multi-site trial, although the CGI-I is also a generalised symptom scale. They conclude that music therapy is ‘efficient’ and should be considered as an add-on for other interventions for young children on the autism spectrum. Unfortunately, the trial (rated as ‘acceptable quality’) included no measures of social and communication outcomes, and there was no longer term follow-up to see whether any effects were sustained. Finally there were many tests conducted, including *ad hoc* analyses on subscales, with the possibility of chance effects and clinically insignificant findings (for example, on the ABC’s *lethargy* subscale).

For a more general account of the limitations of appraised studies, see the discussion in **Section 2.4**.

Table 2.6: Characteristics of primary studies comparing features of music therapy (research question 2)

|  |  |  |  |  |  |  |  |
| --- | --- | --- | --- | --- | --- | --- | --- |
| **Author/s (year)** | **Country** | **Design SIGN rating** | **Sample size, gender** | **Age (Mean, range)** | **Intervention Comparison groups** | **Significant outcomes (Measure) (*informant/assessor*)** | **No difference in outcomes (Measure) (*informant/assessor*)** |
| Schwartzberg et al (2013) [53] | US | Pseudo-randomised trial low quality ( – ) | N=30 97% male | M=15 years (9–21 years) | Group-based MT over 3 days: 3 sessions x 50 minutes (1.5 hours)  Sung social story (within music therapy session)  Read social story (within music therapy session) | For sung social story cf read social story:   * comprehension of social story (CC) (*Clinician* | For sung social story cf read social story:   * social reciprocity (ASSP) *(Parent)* * social participation (ASSP) *(Parent)* * detrimental social behaviours (ASSP) *(Parent)* |
| TIME‑A Trial (2017) [54, 55] | Australia, Austria, Brazil, Israel, Italy, Korea, Norway, UK, US | RCT acceptable quality ( + ) | N=364 83% male | M=5 years (4–7 years) | Individual MT   * High intensity music therapy (MT-HI): 30 min x 1/wk x 5 month; Median=34 sessions (17 hours) * Low intensity music therapy (MT-LI): 30 min x 3/wk x 5 month; Median=15 sessions (7.5 hours) * Control group | For MT-HI group cf CG:   * greater improvement in autistic mannerisms (SRS subscale) *(Parent)* * quality of life of participants (analogue scale) *(Parent)*   **(Note 1*)*** | For MT-HI cf MT-LI groups:   * autism symptom severity (ADOS – social affect) *(Clinician)* * social communication (awareness, cognition or motivation (3 SRS subscales) *(Parent)* * quality of life of family (analogue) *(Parent)* * parental stress (PSI) *(parents)* * parental wellbeing (WEMWBS) *(parents)*   **(Note 2)** |

**Note 1:** All reported 3-group analyses relate to 5 month post-baseline (immediate post-test). **Note 2:** PSI and WEMWBS for UK site only.

**Key**: CG=control group; MT=music therapy; M=mean; N=total sample (randomised participants in intervention and control groups); min=minutes; SIGN=Scottish Intercollegiate Guidelines Network; UK=United Kingdom; US=United States; /wk=per week; See **List of Acronyms** for Scale names.

## 2.3 Synthesis of results

### Systematic reviews

The Cochrane Collaboration systematic review [23], rated as being of high quality, reported on meta-analyses of 10 trials including 4 RCTs, 5 randomised cross-over trials, and one pseudo-randomised cross-over trial. Sample sizes were small, totalling 165 (Range=4-50, M=16.5). The authors concluded that music therapy may lead to moderate to large improvement in autistic children’s social interaction and initiating behaviour, low to moderate improvement in verbal communication within therapy and social adaptation, and possible improvement in social-emotional reciprocity and the quality of parent-child relationships. However it did not identify significant improvements to verbal or non-verbal communication that generalised to situations beyond therapy.

Since the Cochrane review, three other secondary reviews relevant to music therapy have been identified. All of these have been similarly positive about the effectiveness of this intervention for children on the spectrum. In 2015, two reviews [24, 42] including mostly studies already appraised in the Cochrane review reported “promising evidence” of music therapy. Music therapy was found to be a safe intervention with evidence for the use of music in autistic children with respect to improvements in communication, social reciprocity, and emotion [42]. Most recently, a meta-analysis of 14 RCTs [43] considered a diverse range of interventions for pre-school children on the autism spectrum. Three music therapy trials (all included in the Cochrane review) were identified as being particularly effective. The reviewers concluded that music therapy, despite being of relatively short duration and intensity in appraised studies, was an effective tool for improving social interaction in pre-school aged autistic children.

### Primary studies

The current review identified seven primary studies reporting on data from six clinical trial published since July 1, 2013. A Canadian trial assessing individual MT among primary-school aged autistic children compared with “enhanced care” was rated as being high quality using the SIGN appraisal checklist [34]. Four of the trials were rated as being of acceptable quality, including the multi-site TIME‑A trial [54, 55], and a pseudo-randomised trial of the briefest intervention (1.5 hours over 3 consecutive sessions) was rated as being of low quality [53].

The evidence base overall has grown significantly with the addition of the 6 trials over the last 7 years. These trials represent 537 participants in intervention and control arms (Range=17-364, M=89). When combining data from with the 10 studies identified in the Cochrane review [23], the evidence base now consists of 702 participants across 16 trials, with study samples ranging from 4 to 364 people on the autism spectrum (M=44). Ages in the six appraised primary studies ranged from 4 to 21 years, and when including the Cochrane review’s trials, between 2 and 21 years across all 16 trials published. Whilst the US summer camp-based trial included children up to 21 years [53], 4 studies related to primary school aged children aged, and the UK included children aged 8 to 16 years [56]. Four of the six trials appraised in the current review, and all 10 in the Cochrane review, included 76% or more males in their samples, reflecting the 4:1 ratio of males to females typically seen in prevalence studies [57].

The primary research question (research question 1) of this review relates to the effectiveness of music therapy compared to a placebo, usual care, or active control intervention.

As a core characteristic of autism, social communication and social interaction-related measures were key primary outcomes and were reflected in a range of indices completed by a range of informants across three trials, all rated as being of acceptable quality using the SIGN checklist. In a multi-centre UK trial [56] involving older children aged 8 to 16 years, there was no significant improvement for the individual music therapy group for communicative and interactional skills (SSIS) rated by parents and participants. A small US trial [32] for younger children reported improved parent-rated social skills (SRS), and clinician-rated joint attention with peers, and eye gaze, for the group-based music therapy group compared to exercise, but no difference in joint attention with adults. Finally, the large multi-site TIME‑A trial [54] for young children aged 4-7 years found no difference in the social affect subscale of the clinician-rated ADOS measuring autism severity, or in 4 subscales of the parent-rated SRS relating to social communication. In exploratory, post hoc analyses, there was a significant improvement in the social motivation subscale (SRS), and a greater proportion of responders in terms of improved scores on the ADOS social affect subscale, for the MT group compared with the enhanced standard care group.

Global measures of autism symptom severity (ie, total scores relating to core characteristics of autism relating to both domains of social communication and social interaction, and restricted and repetitive behaviours and interests) were assessed in three studies, with mixed results. One acceptable quality study of younger children [20] found reduced clinician-rated autism severity on the CGI but not on the CARS. Whereas a good quality study of primary-school aged children [34] found no improvement on the parent-rated SRS, and an acceptable quality study found no effect of music therapy on the clinician ATEC checklist of autism functioning [32].

Communication skills were improved in one good quality study of pre-school children receiving individual music therapy compared to play therapy [34].

There was mixed results for maladaptive behaviour from two studies. In one acceptable quality study of younger pre-school children receiving group-based music therapy [20] there were improvements on total scores of clinician-rated scale of aberrant behaviour, and specifically subscales relating to reduced lethargy and stereotypy. By contrast, the good quality trial of primary-school children receiving individual music therapy found no significant change in maladaptive behaviour.

There was also inconsistent evidence of improved quality of life from two studies, with one good quality study reporting improved family relationship quality as rated by parents [34] for primary-school aged children receiving individual music therapy. However the Time-A multi-site trial finding no significant impact on parent-rated quality of life of themselves or of their young children who received individual music therapy, or in the UK site, of the parents’ self-rated stress, or wellbeing [54, 55].

There was no significant improvement in receptive vocabulary from one good quality study of primary school aged children [34].

Finally, changes in brain connectivity measured using MRI scans indicated changes in the music therapy group that were consistent with reducing both over-activity and under-activity in autistic people that were correlated with improvements in social communication [33]. This finding adds some support to the researchers’ suggestion that music therapy may improve social functioning through changes to brain networks.

Two studies reported results relating to the second research question of the review concerning the effectiveness of features of music therapy. In the Time-A trial [54, 55] there was no evidence of a dose effect for high-intensity music therapy (M=17 hours) compared with lower-intensity music therapy (M=7.5 hours) in the social affect subscale of the ADOS.

There was also no significant effects on social communication across 3 parent-rated subscales for the inclusion of a sung social story compared to a read social story within otherwise identical music therapy sessions in the low quality, cluster-randomised summer-camp trial evaluating a brief intervention over three days [53]. Comprehension of the social story was greater in the sung condition compared with the read condition which may relate to music being more engaging for participants’ attention.

## 2.4 Review limitations

The current study used a structured approach to review the literature. However, there are some inherent limitations with this approach. The review is limited by the review’s methodology and the quality of the studies included in the review.

### Limitations of review methodology

The current review was restricted to English language studies. Restriction by language may result in study bias, but the direction of this bias cannot be determined. However, it is arguable that developed English-speaking countries most comparable to New Zealand’s health system are more likely to publish in English-language Journals.

The review was limited to the published academic literature, and has not appraised unpublished work. Such restriction is likely to lead to publication biases since studies that show an absence of effect are less likely to be published. However it was reassuring that the two meta analyses appraised in the current review investigated publication through funnel plot reviews and found it to be absent [23, 43].

The review had a broad scope reflected in using inclusive search terms and being unrestricted with respect to study design and sample size in the search strategy. Studies were initially selected for appraisal by examining the articles’ abstracts. Therefore, it is possible that some studies were inappropriately excluded prior to examination of the full text article. To minimise this possibility, where detail was lacking or ambiguous, papers were retrieved as full text. Supplemental searching, including considering the reference list of all retrieved studies, and narrative reviews retrieved as background material, extended the search catchment, increasing the likelihood of inclusion of eligible primary studies.

Geographically, most of the studies tended to be conducted by researchers in industrialised, developed countries. The 6 trials appraised in the current review included two studies from the US, one each from Northern Ireland, Canada, and France, and the multi-centre Time-A trial [54] included 10 sites across 9 countries (Australia, Austria, Brazil, Israel, Italy, Korea, Norway, UK, US). This diversifies further the evidence provided by the 10 trials included in the Cochrane review [23] which included 7 US-based trials, and single trials in Australia, Brazil and Korea.

It is noted that all primary studies included in this review were conducted outside New Zealand, and therefore their generalisability to the New Zealand population, ethnic culture and autism service context may be limited. Such factors must be considered in implementing the research findings (and this guideline update) locally. This is particularly needed to honour the Crown’s obligations to Te Tiriti o Waitangi with respect to considering what approaches and resources are needed to achieve equitable health outcomes for Māori.[[3]](#footnote-3)

Whilst no New Zealand research study met inclusion criteria for the review, sectoral consultation considering a late draft of this Supplementary Paper identified consideration of the applicability of music therapy research and practice to Māori. Specifically, the Standards of Practice [60] require that Registered Music Therapists in New Zealand be informed about the meaning and implications of the Treaty of Waitangi for their practice; and actively incorporate the principles of protection, participation, and partnership in their work with Māori, and other groups. Further, Music Therapy New Zealand, and training institution Te Herenga Waka – Victoria University of Wellington, are working with Māori to increase their participation in music therapy practice and research, to determine and maximise the relevance of western music therapy approaches for Māori (Daphne Rickman, *personal communication*, 17 March 2021).

The search, data extraction, synthesis and report preparation was performed by a single reviewer over a limited timeframe (July to November 2020). For a detailed description of interventions, methods and results of the studies appraised, the reader is referred to the original papers cited.

### Limitations of appraised studies

The review’s conclusions are limited by the methodological quality of included studies.

#### Study quality

For the current review, included studies were systematic reviews and more recently published randomised controlled trials and pseudo-randomised controlled trials. Study quality was rated using the SIGN critical appraisal checklist [41]).

The four appraised systematic reviews included the Cochrane review, which the current review updates, rated as being of “high quality” [23]. The other three reviews were rated as “acceptable quality” [24, 42, 43], limited in isolated areas such as lacking a formal appraisal tool, reporting of limited data, and a lack of sensitivity analyses.

The Cochrane review [23] included 10 trials: four RCTs, five randomised cross-over trials, and one pseudo-randomised cross-over trial. Cross-over trials randomise the session order for receiving music therapy or the control/comparator condition. This approach is only appropriate for chronic conditions and for interventions with short-acting effects, and are best used in early trials where resources for larger samples are limited [23]. Only some of the outcome measures were published measurement tools, and some relied on parental reports that are unblinded to group allocation, and may be biased towards the intervention. Evidence quality across the outcomes considered was judged to be low or moderate.

Of the six trials published since the Cochrane review, and using the SIGN quality appraisal checklist, one trial [34] was rated as ‘high quality’, four were rated as ‘acceptable quality’, and a pseudo-randomised trial of the briefest intervention (1.5 hours over 3 consecutive sessions) was rated as ‘low quality’ [53].

Key methodological limitations of these seven included primary studies (representing six trials) are discussed below.

#### Sampling and recruitment

When studies have small samples, limited recruitment, and poor response and study completion rates, sampling, detection, and attrition biases can lead to unpredictable effects. They reduce the likelihood that the results represent, and are generalisable to, the population the intervention is targeted towards.

As presented in the synthesis above, the evidence base has been greatly enhanced since the Cochrane review of 10 trials by an additional six trials identified by the current review. Across the 16 trials, over 700 participants represented ranging in age from two to twenty-one years, over three-quarters of whom were male, reflecting the higher prevalence for ASD among males typically seen [57]. Notably two included studies had more female than male participants, representing 56% of the sample in the UK trial [56] and 52% in the Canadian RCT [34].

Levels of cognitive functioning were not always reported in the appraised studies, making it difficult to determine their generalisability to the wider autism population. In three trials however, there was some evidence that samples were inclusive in this respect. The high quality Canadian trial [34] included a sample where over half of participants had language impairment. The TIME‑A trial [54] included 45% of participants with low cognitive functioning (IQ<70), and children in the French trial [20] had a reported average IQ of 52. Given that music therapy has been promoted as being a way into communication, it is crucial that its effectiveness across a broad range of children with differing cognitive and communication needs is demonstrated.

Information on recruitment was generally limited. Two studies recruited participants from health services, including a psychiatric day-care facility [20] and a mental health community health service [56]. One study was very targeted in involving autistic participants in a summer camp where music therapy was provided [53]. Others were more general, referred to using word of mouth and flyers for recruitment [32], and another sample were simply screened “in the community” [34]. The multi-site Time-A trial used diverse recruitment methods across the 10 centres involving clinics, kindergartens, and other forms of community outreach [54, 55].

All but one of the six trials appraised for the current review were fully and robustly randomised with the exception of US cluster-randomised trial [53] where randomisation method was not reported. This opens up the possibility of biases in the allocation to group process which may make the groups different at baseline. Two-thirds of the sample allocated to groups and completing their baseline assessment did not return their follow-up assessments, and were then excluded from analyses without ‘intention to treat’ management of missing data. These methodological flaws contributed to the SIGN rating of this study as being of ‘low quality’. As there were no analyses reported at baseline between conditions, or between those lost to follow-up and those retained, it is not known whether there were biases introduced by these issues. However that no group differences were reported by this study for the primary outcome of social skills suggests that any biases did not inflate findings in favour of music therapy.

Considering the other 5 primary studies appraised, attrition rates were very low for two trials [20, 34], but were moderate for three, ranging from 14-28%. All found higher drop-out rates for the control groups [32, 54, 56]. If dropping out is potentially related to dissatisfaction with the control group comparators, this could introduce attrition bias that would artificially inflate positive outcomes for the control group and therefore inhibit the study’s ability of detecting significant treatment effects for the music therapy group.

#### Interventions and comparators

One of the challenges in synthesising results across this intervention is that the form and delivery of music therapy offered varied widely. In half of the trials, music therapy was delivered individually [34, 54, 56] whereas it occurred in groups in the other half [20, 32, 53]. One study’s intervention involved the therapist singing a social story. Five interventions involved the participants performing by playing instruments and/or singing, including two interventions involving movement. Of the five interventions involving performance, two explicitly referred to the use of cueing by the music therapist, and three referred to the use of improvisation in performance. Across these various approaches, it is not always clear what the underlying mechanism is that is understood to lead to improvements in music therapy, and therefore what strategies and components are considered necessary. There is the possibility that in music therapy, the care and attention of the therapist itself, regardless of musical content, provides a beneficial element. However, it has been argued that the therapeutic relationship itself cannot be disentangled from music therapy [56].

These complexities make it difficult to generalise across music therapy interventions that have distinctly different features. Programme fidelity (the delivery of an intervention as intended) was not formally investigated or reported in four of the six included studies [20, 32, 53, 56]. Notably the Time-A multi-site trial’s [54, 55] authors observed that fidelity was ‘overall adequate’ but admitted that it was difficult to ensure consistency across 10 trial sites and 30 music therapists in nine countries. Whilst variation in practice may be considered a methodological limitation, it also reflects the real world situation of variation on practice across different musical therapists.

The use of active comparators that are structurally matched to the intervention aim to control for non-specific factors that may be provided by music therapy but are not *core* to it. These features can include positive expectancies (ie, placebo effects), general support, therapist attention and emotional engagement [34]. In the current review, four of the six trials included some form of active comparator. These included a read social story [53], a group exercise and game session without music [32], a play-based intervention [33], and listening to music [20]. Whilst use of active comparators strengthen study design by isolating effects to unique music therapy components, they also make it harder to detect benefits of an intervention over and above a comparator which itself may offer benefit. One trial offered enhanced standard care including support and parents’ information sessions as a means of ethical equivalence, rather than as an active control [34]. However in this trial, participants in the control group received more established concomitant therapies (such as speech and language therapy, communication training, occupational therapy, and physical therapy) than the music therapy group as part of usual care, arguably because they had more time to schedule them.

In routine practice, music therapy can occur weekly for months or years. The music therapy interventions appraised in the current review were generally much briefer, and may not reflect the impact of usual longer-term practice. The delivery of the therapy itself varied widely in terms of time involved, the frequency and number of sessions, and the period over which these were held. And so, the briefest intervention was for 1.5 hours over three consecutive days [53] whereas the most intense was thrice weekly sessions over 12 weeks [54]. The longest study period was 25 weekly sessions of group therapy offered over a period of 8 months [20].

Such variability in intervention intensity and duration makes it difficult to determine the optimal frequency and dosage for greatest improvement, including whether there is a ceiling effect after which benefits diminish. Whilst the multi-site Time-A trial [54] did attempt to explore whether there was a dose effect by including a thrice-weekly high-intensity group, none was found in the measures employed. Notably, drop-out rates and missed sessions were high and the researchers reported that many parents had difficulty scheduling in that frequency of therapy. It is important in intervention research that the music therapy offered within the research setting approximates its use in routine practice, is practical and realistic. Whilst most appraised trials provided manuals outlining their approach, they usually permitted flexibility within the general framework, allowing the therapist to tailor their approach to the participant as they would in the ‘real world’.

#### Assessment

The wide range of outcome domains and related assessment tools used in the trials conducted on the use of music therapy for autistic people makes synthesising the evidence challenging, particularly as there has been little overlap between tools employed. Outcome domains such as social interaction and communication include specific behaviours (eg, eye gaze), subscale areas (eg, social awareness, social cognition, social motivation) and global autism symptom severity measures. Measures can apply to the participant’s behaviour observed during the therapy itself, shortly after, or in generalised behaviour in other settings. Which of these are used and reported is key in interpreting whether music therapy has short-acting impacts on specific behaviours, or whether any improvements generalise to other situations and contexts outside of therapy, which is the ultimate goal. For this reason, longer term follow-up to assess maintenance of any improvements is very important. In the trials appraised for the current review, longer term follow-up post conclusion of the treatment period was evident for only three studies, including after 5 weeks [32], 13 weeks [56], and 7 months [54]. Such longer-term follow-up is crucial to determine whether any improvements to outcomes are maintained after the intervention ends.

Statistically different scores may also not translate to clinically significant changes, or indeed to outcomes that are valued by therapy participants, such as quality of life. Reduction of “undesirable or challenging behaviours” is subjective and some of those specified in tools, such as aberrant vocalisations, rewinding/fast forwarding video tapes, and rummaging in the kitchen, may not be a problem for the autistic person or those around them [24]. The degree to which some behaviours may be undesirable may therefore vary in terms of context and the perceptions of others, and are socially constructed. Choice of outcomes that are important to people in the autism spectrum is key.

A limitation that is particularly important to consider for this review relates to blinding. That is, assessors or informants knowing whether the participant received the intervention or usual care/control condition. Participants were not able to be blinded to condition but could be blinded to the study design or hypotheses, thus reducing the risk of performance bias [23]. Therapists are unable to be blinded to study allocation and therefore the risk of performance bias introduced by therapists administering the intervention is unknown. However assessments by researchers or clinicians were blinded in all but one of the appraised studies [53]. In the Time-A trial, clinicians were asked whether the blinding had been broken and revealed that 5% (20/364) of participants had inadvertently revealed their allocated group to them during the course of the assessment interview. This suggests that blinding was generally successful. By contrast, parents were rarely blinded to their child’s group allocation, occurring in only one study, the high quality Canadian trial [34]. Even then, the researchers determined that 61% of parents had inadvertently become aware of treatment condition during the course of the trial (eg, from comments by their children).

Unblinded studies cannot control for reporting biases of observers in seeing an improvement, which may particularly apply to parents with expectations about the value of the programme, and/or a desire to assist the researchers for the time and effort invested in offering the music therapy programme. Reporting biases from lack of blinding may artificially inflate ratings of the effectiveness of an intervention. In **Table 2.5**, of the 13 significant analyses reported, four outcomes were unblinded, including three from parent-completed outcomes [32, 54] and one clinician completed outcome [53]. Two outcomes were nominally blinded parent-outcomes (albeit with 61% of parents being unofficially aware of condition) [34]. The remaining seven significant outcomes in three studies were clinician blinded [20, 32, 34], though three of these were total and subscale scores of the same instrument [19]. There was also a lack of reporting of interobserver agreement ratings to determine accuracy of coding of observed behaviours.

## 2.5 Future research

Addressing the limitations of the current evidence base will inform future research into music therapy interventions for individuals on the autism spectrum.

### Sampling and recruitment

It is positive to see that larger studies have emerged since the Cochrane review, particularly the ambitious multi-centre Time-A trial [54]. Larger sampled, well-powered studies permit sub-group analysis that may identify individuals for whom music therapy is most effective. From their systematic review, James et al (2015) [24] recommended studies investigate the effectiveness of different music therapy approaches among people of different ages, functioning, and language impairment.

With respect to age, the focus of much existing research has been on young, pre-school aged children and those attending primary school. This is consistent with an increased emphasis on early intervention research in autism, particularly around communication. Research is needed in the potential benefits of music therapy for older children, adolescents, and adults on the autism spectrum [24]. Whilst not every intervention is suited to every individual, it is important that studies include a diverse range of participants representative of varying communication needs, cognitive functioning, motor skills, socioeconomic status, and cultural/ethnic backgrounds. Half of the appraised studies included children with language or cognitive impairment, whereas the others did not report this information. More diverse samples generally will permit the systematic investigation of whether music therapy is more or less appealing, effective, and appropriate for people across varying characteristics or backgrounds.

### Interventions and comparators

Researchers have investigated the impact of music therapy on a range of outcomes without necessarily establishing the underlying mechanism considered to be at work. Future studies should use sound theory to describe and measure the expected active components of music therapy and assess how these impact on autistic participants.

Some components of music therapy may be more effective, and more necessary, than others. Understanding the mechanisms by which music therapy impacts on outcomes is likely to lead to improvements to how the interventions are designed, what key elements are included, and what may be left out, making programmes more effective and efficient in achieving their goals. Rigorously conducted research is needed to systematically vary and assess the contribution of different elements of music therapy programmes [24]. Moderators and mediators of effectiveness may include features such as activity type, choice of music, choice of instrument, overall duration, intensity, delivery, experience of the therapist, and location. For example, the large sampled Time‑A trial included two active music therapy groups that varied by intensity (frequency and dose of sessions) of music therapy. Unfortunately, interpreting the lack of difference found is hampered by higher dropout rates for the high-intensity condition which may have related to practical difficulties of attendance [54].

In their systematic review James et al (2015) [24] found that most evaluated music therapy interventions used songs with lyrics related to target skills, and improvisation. They suggest that future research should evaluate other used music therapy approaches including structured music activities, song-writing, and composition. Interventions should also be pragmatically delivered; that is, conducted under similar conditions and to a level of quality to that of usual practice. Inclusion of more than one therapist can also increase generalisability. Pragmatic trials are inevitably harder to maintain consistency in implementation [54] but they better reflect the nature of many music therapy techniques which tend to be flexible and adjusted for the individual client [23].

Delivery of music therapy, whether individually or in groups, should also be robustly investigated. Participation of parents and other family members is a promising development that may impact on effectiveness. Notably, the Australian trial [26, 31] included in the Cochrane review [23] found evidence of increased social interaction from this therapy approach.

### Assessment

In the current review, there was a broad range of outcome domains, and assessment tools employed to measure these, that varied significantly across the studies. This makes synthesis challenging. The consistent use of validated, standardised instruments would permit more meaningful study comparisons, data synthesis and meta-analysis.

The inclusion of general measures of autism’s core characteristics has been questioned. The large multi-site TIME‑A trial included the ADOS as its primary outcome, a scale originally developed as a diagnostic tool to assess unusual impairments or behaviours that may indicate autism [54]. Measures of autism severity may not be appropriate tools to assess intervention effectiveness. Designed to assess clinical differences, they may be less sensitive to changes in the degree and type of benefits from music therapy that are valued by people on the autism spectrum and their families. The Brief Observation of Social Communication Change has been suggested as a better tool to assess changes in social communication skills in children on the autism spectrum and may be more sensitive to change than generalised symptom scales [55].

It should be acknowledged that choice of outcomes is a value-based decision. There has been a call for a shift away from outcomes relating to a reduction of symptoms, impairments and autistic traits, terms associated with a medical model of disability as disease as opposed to a social model of disability which sees people as being disabled by barriers in society, not by their impairment or difference [15]. It is important for researchers to embrace outcomes that matter to autistic people. It has been argued that the focus should therefore extend beyond reduction of autism characteristics, to quality of life, wellbeing and adaptive functioning, including being able to participate in learning, school, and work and to have meaningful relationships with others [34, 54]. Measuring the impact of music therapy on mental health challenges which are also commonly present in people on the autism spectrum such as anxiety is also desirable [55].

Assessment of outcomes that can generalise to contexts outside the therapy situation and with unfamiliar and familiar assessors is also important. Measures rated by parents and teachers can assess behaviours across settings, circumstances and time periods. This can be particularly important for children with complex needs who may be non‑compliant in formal test situations [31]. Where possible, outcomes should also be completed by independent, blinded-to-condition assessors. Parents can be kept blind to group allocation though it is understood that children may inadvertently make a comment that will reveal which condition they were in. However the success of blinding can be gauged by asking assessors or informants if and how they may have discovered the condition a participant has been allocated to, as employed in the TIME‑A trial [54].

With respect to study design, the Cochrane review [23] included randomised cross‑over trials using single case experimental designs (SCED) whilst no additional SCED studies were identified in the current review. SCED studies have the advantages of being easier and less expensive to conduct, requiring fewer participants, and controlling for individual differences by permitted participants to act as their own control. However, they are open to maturation and learning effects, and are less adequate than group RCTs for assessing the impact of longer term interventions and sustained effects [23]. Whilst both study designs are useful, larger, parallel RCTs are therefore likely to provide the strongest evidence where possible of effectiveness.

More trials employing longer follow-up assessment are also needed to see whether benefits are maintained over time or cease when music therapy ends. The TIME‑A trial [54] is the first trial to examine whether music therapy for participants with autism is associated with benefits that persist several months beyond the period of treatment.

It should be noted that the guideline update process is primarily concerned with whether a supportive approach or intervention can be recommended or not, and this is based on whether it is effective in leading to sustained benefits for the autistic individual. Consistent with the evidence-based practice model, this question is most robustly answered using study designs that appear higher in the evidence hierarchy which have a lower risk of bias (see **Table A1.1**), such as well conducted, controlled experimental studies. However this focus does not mean that such research designs are valued more highly *per se*. Well-conducted observational and qualitative studies are core to any research programme. They offer unique insights that inform the development of interventions; guide their improvement, delivery and implementation; and explore individual responses, and their cultural and contextual influences. Mixed methods research which attempts to integrate both quantitative and qualitative elements are able to provide complementary evidence about whether an intervention or support works, as well as how it works and for whom it works. It can also help to understand why an intervention is not effective and how it may be improved or better implemented.

## 2.6 Systematic review summary and conclusions

### Overview

This systematic review updates evidence for the New Zealand Autism Spectrum Disorder Guideline [12] with respect to the effectiveness of music therapy interventions for people on the autism spectrum. Following a comprehensive database search and citation searching of primary and secondary studies published between 1 July 2013 and 15 September 2020, 370 unique abstracts were identified. After applying inclusion and exclusion criteria, 11 studies were included for critical appraisal: 4 systematic reviews, and 7 primary studies (reporting on 6 trials) collecting original data.

### Evidence for effectiveness of music therapy

The four systematic reviews all supported the potential for music therapy to have a positive impact on the lives of people on the autism spectrum across several domains. The Cochrane Collaboration’s [23] meta-analyses of 10 randomised controlled trials and cross-over trials provided low or moderate evidence relating to music therapy’s effectiveness for children on the autistic spectrum. The authors argued that this permitted “fairly robust conclusions” (p. 20) that music therapy may make moderate to large improvements to autistic children’s skills in social interaction and initiating behaviour, and low to moderate improvements to social adaptation, and verbal communication within therapy, and possibly social-emotional reciprocity and the quality of parent-child relationships. However it did not identify significant improvements to verbal or non-verbal communication that generalised to situations beyond therapy. The review’s positive assessment was supported by findings of three more recently published secondary reviews, describing evidence for effectiveness of music therapy for children on the spectrum as ‘promising’ [24, 42], ‘safe’ [42], and ‘effective’ [43] for improving social communication and social interaction in autistic children.

The addition of 6 trials appraised in the current review extend the evidence base significantly from the 10 trials included in the Cochrane review [23] with respect to sample size, gender, age, and geographical location, reporting on 537 participants on the autism spectrum, mostly primary school aged children.

Five fully randomised controlled studies investigated research question one relating to whether music therapy interventions can provide benefits for children on the autism spectrum.

In the core domain of social communication, there was moderate quality but somewhat inconsistent evidence from three trials relating to various outcomes. One multi-centre UK trial [56] found no significant improvement in communicative and interactional skills. A small US trial [32] reported improved parent-rated social skills (SRS), and clinician-rated eye gaze and peer joint attention, but not joint attention with adults [32]. And the multi-site TIME‑A trial [54] found no difference in the social affect subscale of the clinician-rated ADOS measuring autism severity (social affect subscale), or in 4 subscales of the parent-rated SRS relating to social communication. However there was a significant improvement in the social motivation subscale (SRS), and a greater proportion of responders in terms of improved scores on the ADOS social affect subscale for the music therapy group compared with the enhanced care control group (albeit in post-hoc analyses).

There was inconsistent evidence with respect to whether music therapy improves global autism severity symptoms and functioning when measured using global diagnostic scales [20, 32, 34].

There was moderate to good quality evidence of improvement in pragmatic communication skills [34]. However no apparent benefit with respect to receptive language [34].

There was good quality evidence from one trial that music therapy may improve family relationship quality [34]. However the multicentre TIME‑A trial found no improvement to quality of life of participants or their parents [54], or at one site, to parental stress or wellbeing [55].

There were mixed findings relating to whether music therapy reduces maladaptive behaviours [20, 34].

These studies need to be considered alongside the trials included in the Cochrane review [23] which found moderate to large effects in improvements to social interaction within and beyond therapy, and in initiating behaviour within therapy, low to moderate improvements in social adaptation, and possible improvements in social-emotional reciprocity. It is possible that the assessment tools employed by recent trials to measure the impact on social communication appraised in the current may too broad and crude to pick up improvements to specific areas within the broad domain of social communication and interaction.

### Evidence for effective features

With respect to the second research question of effective features of music therapy, two trials were designed to consider specific variations [53-55].

Regarding dose of intervention, the large multi-site TIME‑A trial [54, 55] did not demonstrate a consistent benefit in reduced autism severity for the high intensity group (17 hours) compared with the low-intensity group (7.5 hours). However, those receiving more than 15 hours were more likely to have had a significant response on an autism severity scale score for social communication compared with control group in exploratory analyses. Parent-rated quality of life was improved for participants (but not their families) receiving high intensity music therapy [53].

Another low quality trial found no impact on social communication of varying one component of music therapy such that a social story was sung rather than read [53], except finding that comprehension was enhanced for the sung story group.

### Conclusions

There is a growing body of evidence that music therapy interventions can provide benefits across a number of domains for children on the autism spectrum, particularly with respect to social communication.

Whilst some systematic reviews have attempted to conduct meta-analyses, synthesising the current evidence base is challenging given its heterogeneity. It is therefore not currently possible to offer specific recommendations about the most successful and necessary components, delivery, and duration of music therapy interventions. It is also not possible to determine whether there was greater effectiveness of music therapy evident for sessions delivered to groups versus individuals, or for different music therapy approaches. Possible explanations for discrepancies between studies include variation in music therapy interventions and duration, participant characteristics, outcome domains, measures employed, informants involved, and the quality of studies themselves (as discussed under the limitations of the review, earlier).

There is currently a research gap in understanding whether music therapy may be effective for young people or adults on the autism spectrum, with most research relating to young children. It is also not clear whether benefits found in some studies are likely to apply to participants with different levels of cognitive ability or severity of autistic characteristics. The area would benefit from a greater explication of underlying mechanisms involved, how they are operationalised in therapy, and their association with outcomes. A better understanding of the underlying active features of therapy would inform the selection of relevant process and outcome measures.

The Cochrane review authors [23] made an interesting observation that music therapy should be backed by research into related interventions which are also aimed at enhancing “cooperation with others involved in treatment and care of clients, active engagement of clients, and establishing structure, predictability, and routines” (p. 19). These very features which may contribute to making music therapy effective and appealing to some people on the autism spectrum may also be worth pursuing in other approaches. That is, centring an intervention around a special interest or skill that an autistic person is passionate about, and encouraging the development of a strong therapeutic relationship in supporting this passion.

# Part 3: Recommendation development

The Living Guideline Group (LGG) was tasked with considering the systematically reviewed evidence reported in **Part 2** above in addition to the evidence already appraised and included in the guideline and it’s Supplementary Papers [2-12]. Specifically, the LGG considered whether the updated body of evidence required revisions of any existing relevant recommendations and good practice points (GPP) as well as the development of new ones.

The text and graded ‘strength of evidence’ of any potential new recommendations (see **Appendix 1**, **Table A1.2**) were considered at an all-day face-to-face meeting. The LGG’s decisions are presented below, and summarised in **Summary Table 1.** Where considered helpful, these decisions are accompanied by additional explanatory text, and/or with a brief rationale which highlights any particular issues that the LGG took into account in their deliberations.

## Decisions of the Living Guideline Group

### Preamble

There is a growing body of evidence that music therapy interventions can provide benefits across a number of domains for children on the autism spectrum, particularly with respect to social communication.

In this review, music therapy is defined as the planned use of musical experiences and the relationships that develop through them for therapeutic goals. Therapy is delivered in regular sessions to individuals or groups by a music therapist (or person trained in music therapy techniques by a music therapist). Whilst no age restriction was applied, the results principally related to children and young people on the autism spectrum.

It should be noted that this review and the LGG’s recommendation do not relate to the value of music *per se*, or to interventions where music is a component but where music therapy is not the core intervention.

**New Recommendation 4.5.3:** “Music therapy can enhance social communication skills and should be considered for children and young people on the autism spectrum.” Grade B

**Rationale:** The LGG considered the body of evidence relating to the effectiveness of music therapy as measured in 6 trials appraised in the current review, in addition to 10 trials considered in the Cochrane systematic review and meta-analysis [22].

The original Guideline [11] noted that “no large scale randomised control trial involving young children with autism has been conducted” (p. 315). Since then there has been a significant number of trials, including the large Time-A multi-centre trial [54, 55]. The authors of this significant trial, conducted across 9 countries and involving 365 young autistic children, reported that it “did not support the use of improvisational music therapy for symptom reduction in children on the autism spectrum” (p. 525). This conclusion was based on a lack of a significant effect on its primary outcome measure which was a diagnostic instrument (ADOS). The LGG considered that this measure may not be sensitive to change or reflect outcomes that are important to people on the autism spectrum. The trial was also limited in that children in the comparative “enhanced usual care” arm tended to receive more hours of other established concomitant therapies (such as speech and language therapy, communication training, occupational therapy, and physical therapy) than the music therapy group.

Considering the evidence base as a whole, particularly its limitations with respect to choice of outcome measures and heterogeneity of samples and interventions, the LGG considered that there was sufficient evidence to recommend music therapy for children and young people on the autism spectrum. However the mixed findings across trials led this recommendation being graded as a B rather than an A.

In terms of placement of the recommendation within the guideline, music therapy was originally referenced briefly in the Guideline [11] in Part 3 (Education for learners with ASD), under Section 3.2.c relating to ‘Sensori-motor development.’ However the LGG decided that the new Recommendation was more appropriately referenced within Part 4 (Treatment and Management of ASD), within Section 4.5 relating to ‘Other interventions’.

**Additional text:** A cross-reference to this Recommendation is also provided in Part 3 (under *3.2.c Sensori-motor development)*, and in Appendix 8, where music therapy is discussed within a broader overview of educational interventions.

**Summary Table I** (p. x) of the Executive Summary presents the new recommendation.

# Appendix 1: Methods

## A1.1 Acknowledgements

This project is funded by the New Zealand Ministries of Health and Education. Marita Broadstock prepared the systematic review and delivers the living guideline process. The Living Guideline Group consider implications for the Guideline as detailed in Part 3.

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### Declarations of competing interest

None

## A1.2 Search strategy

Search strategies were limited to publications from 1 January 2004 onwards. Database searches were conducted on 9–17 June, 2020, and updated on 15 September 2020. Full search strategies are available upon request. Bibliographic, health technology assessment and guideline databases were included in the search strategy, listed below.

* Medline (Ebsco-host)
* Cinahl (Ebsco-host)
* Embase (Ovid)
* PsychInfo (Ebsco-host)
* PsycARTICLES (Ebsco-host)
* ERIC (Ebsco-host)
* Cochrane Library (Ovid)
* Cochrane Database of Systematic Reviews (CDSR)
* Central Register of Controlled Trials (CRCT)
* Database of Abstracts of Reviews of Effects (DARE)
* Health Technology Assessment Database (HTA Database)

A combination of search terms were used and adapted for different databases. The following illustrative search was used for databases accessed through OVID:

1. (music or sing or SINGING or song#).ti. or (music or sing or SINGING or song#).ab
2. (music or singing or music therapy).sh
3. (autis\* or asperger\* or pervasive developmental disorder).sh. or (autis\* or asperger\* or pervasive developmental disorder?).ti. or (autis\* or asperger\* or pervasive developmental disorder?).ab
4. 1 or 2
5. 3 and 4
6. limit 5 to (human and english language and yr=“2004 -Current”).

Searches were limited to English language publications involving human participants.

Following removal of duplicates, 740 potentially relevant articles were identified published since 2004. Following the principal of appraising “best evidence”, the search for the current review was further refined to publications from 1 July 2013, to update the search completed by a high quality systematic review undertaken for the Cochrane Collaboration in the same month [23]. This led to a reduced set of n=338, with an additional 32when the search was re-ran in 15 September 2020, summing to 370 potentially relevant abstracts scanned for eligibility.

Table A1.1: Hierarchy of evidence

|  |  |
| --- | --- |
| **Level** | **Intervention** |
| I | A systematic review of level II studies |
| II | A randomised controlled trial |
| III-1 | A pseudo-randomised controlled trial (ie, alternate allocation or some other method) |
| III-2 | A comparative study with concurrent controls:   * Non-randomised experimental trial * Cohort study * Case-control study * Interrupted time series with a control group |
| III-3 | A comparative study without concurrent controls:   * Historical control study * Two or more single arm study * Interrupted time series without a parallel control group |
| IV | Case series with either post-test or pre-test/post-test outcomes. |

**Source**: NHMRC [40]

Studies already appraised for a relevant research question in the guideline’s first or second edition [1, 12] were excluded from the current review regardless of date of publication. A single researcher performed study selection, data extraction, critical appraisal, and synthesis.

## A1.3 Levels of evidence

Selection criteria relating to study design was determined after considering the results of the search strategy so as to identify the “best evidence” to inform recommendation development.

Research study designs are broadly associated with particular methodological strengths and limitations in terms of how bias is minimised. This allows studies to be assigned a “level of evidence” within an evidence hierarchy [40], so as to rank them in terms of quality from most robust (level I) to least (level IV) (see **Table A1.1**). Level I evidence includes systematic reviews and associated meta analyses which include at least one level II study, a randomised controlled trial. Systematic reviews of lower order evidence rank at the same level as that order of evidence.

In the current review, there was sufficient higher level relevant evidence to permit research designs to be restricted to level I, II and III.1 evidence published from July 2013, the date of the search strategy employed by a high quality Cochrane review [23].

## A1.4 Data extraction

Study characteristics were extracted for each of the appraised studies and entered into evidence tables (see **Appendix 3**). Key features recorded for primary studies included:

* Authors, country, study design, level of evidence, SIGN checklist quality rating
* Sample characteristics, recruitment, allocation to groups, drop-outs, method of randomisation
* Selection criteria, timing of assessments, study fidelity, blinding of assessment, reliability of coding, analyses
* Intervention and comparators, duration, dose
* Outcomes assessed, assessor/informant
* Results including comparisons at baseline
* Authors’ conclusions
* Key methodological strengths and weaknesses
* Funding source of study

Evidence tables for secondary studies included the review search strategy and methodology. Tables for secondary reviews also listed primary studies that met selection criteria for the current review, and those which were also included in the Cochrane review [23].

## A1.5 Critical appraisal

In addition to the level of evidence associated an intended study design, the quality of how a study design is actually conducted can be assessed using critical appraisal tools. In the current review, included studies were formally appraised using the relevant SIGN quality checklists from the Scottish Intercollegiate Guidelines Network [41].

The quality and resistance to risk of bias of an individual study was rated as follows:

* **High quality** (++): Majority of criteria met. Little or no risk of bias
* **Acceptable quality** (+): Most criteria met. Some flaws in the study with an associated risk of bias
* **Low quality** (-): Either most criteria not met, or significant flaws relating to key aspects of study design
* **Reject** (0): Poor quality study with significant flaws. Wrong study type. Not relevant to guideline

## A1.6 Preparing recommendations

A one-day face-to-face meeting was held on 25 November 2020 where the LGG considered the findings of the current systematic review. Using their collective professional judgement and experience, the LGG discussed the body of evidence with respect to the research question and the applicability of the evidence within New Zealand. They considered any existing affected recommendations (and good practice points) from the guideline [12] and the development of new ones.

Developing recommendations involves consideration of the whole evidence base for the research question. The quality and consistency of the evidence and the clinical implications of the evidence within a New Zealand context is weighed up by all the LGG members. The grades of recommendations used by the LGG, and also used in the guideline [12], are presented in **Table A1.2**.

Each recommendation is assigned a grade to indicate the overall ‘strength of the evidence’ upon which it is based. Strength of the body of evidence is determined across three domains [40]:

* quality (the extent to which bias was minimised as determined by study design and the conduct of the study)
* quantity (magnitude of effect, numbers of studies, sample size or power)
* consistency (the extent to which similar findings are reported.

The wording of recommendations and GPP, and the evidence grade, is determined by the LGG through discussion and group consensus during the meeting.

It should be noted that systematic reviews and meta analyses (secondary studies) considered which draw on publications over an overlapping timeframe could report on (some of) the same primary studies appraised. For this reason it is important to be aware that the results from secondary studies should not be summated as independent sources of evidence as this would misrepresent the quantity of studies and give shared primary studies undue weight.

Table A1.2: Guide to grading recommendations

|  |  |
| --- | --- |
| **Recommendations** | **Grade** |
| The recommendation is supported by good evidence (based on a number of studies that are valid, consistent, applicable and clinically relevant) | **A** |
| The recommendation is supported by fair evidence (based on studies that are valid, but there are some concerns about the volume, consistency, applicability and clinical relevance of the evidence that may cause some uncertainty but are not likely to be overturned by other evidence) | **B** |
| The recommendation is supported by international expert opinion | **C** |
| The evidence is insufficient, evidence is lacking, of poor quality or opinions conflicting, the balance of benefits and harms cannot be determined | **I** |
| **Good practice point** | **Grade** |
| Where a recommendation is based on the clinical and educational experiences of members of the Living Guideline Group, or feedback from consultation within New Zealand, it is a Good Practice Point. | **✓** |

**Note:** Grades indicate the strength of the supporting evidence rather than the importance of the evidence [40].

## A1.7 Consultation

Seeking comments from stakeholders is vital for peer-review and quality assurance processes in developing the report. In a focused consultation eight key stakeholder organisations/individuals were approached for feedback on a late draft of the report. Particular attention was sought regarding the relevance of the report to New Zealand’s services and needs, clarity and ease of use of the report, and implementability of the revised or new recommendations and GPP.

Detailed responses were received from 6 organisations/individuals representing a 75% response rate. The lead researcher (INSIGHT Research) collated feedback and drafted revisions for the LGG to consider. Suggestions identified in the consultation led to several improvements to the final report. INSIGHT Research and the LGG are grateful to those individuals and organisations who participated in the consultation process.

# Appendix 2: Abbreviations and glossary

## A2.1 Abbreviations and acronyms

### Miscellaneous terms

AAC alternative and augmentative communication

ABA Applied Behaviour Analysis

ANOVA analysis of variance

ANCOVA analysis of covariance

AS Asperger’s syndrome

ASD Autism Spectrum Disorder

CAM complementary and alternative medicine

cf compared with

CAMHS Child and Adolescent Mental Health Service

EBP Evidence-based Practice

FCMT family-centred music therapy

GPP Good Practice Points

GRADE Grading of Recommendations: Assessment, Development, Evaluation

IQ intelligence quotient

INSIGHT Research Independent Specialist in Guidelines & Health Technology Research

k number of studies

LGG Living Guideline Group

LMEM linear mixed-effects models

MT music therapy

mth month

M mean

ns not significant

N (or n) number (usually, sample size)

NHMRC National Health and Medical Research Council (Australia)

NR not reported

OR Odds Ratio

PDD Pervasive Developmental Disorder

PDD-NOS Pervasive Developmental Disorder – Not Otherwise Specified

PECS Picture Exchange Classification System

PRISMA Preferred Reporting Items for Systematic Reviews and Meta-Analyses

QoL quality of life

RCT randomised controlled trial

SCED single case experimental design

SES social-economic status

SIGN Scottish Intercollegiate Guidelines Network

SLT speech and language therapy

SMD standardised mean differences

UK United Kingdom

US United States of America

/wk per week

### Tests, scales and measures

ABC Aberrant Behavior Checklist

ADOS Autism Diagnostic Observation Schedule

ASSP Autism Social Skills Profile

ATEC Autism Treatment Evaluation Checklist

CARS Childhood Autism Rating Scale

CCC Children’s Communication Checklist

CC comprehension checks

CGI Clinical Global Improvement Scale

DSM-IV-TR Diagnostic and Statistical Manual of Mental Disorders – IV (text revision)

DSM5 Diagnostic and Statistical Manual of Mental Disorders – 5th edition

ESCS Early Social Communication Scales

FQoL Beach Family Quality of Life

ICD-10 International Statistical Classification of Diseases and Related Health Problems

MBCDI-W&G MacArthur-Bates Communicative Development Inventories – Words and Gestures

MPIP Mother Play Intervention Profile

PCRI Parent-Child Relationship Inventory

PDDBI Pervasive Developmental Disorder Behavior Inventory

PSI Parental Stress Index

RSFC Resting state functional connectivity

SRS Social Responsiveness Scale

SSIS Social Skills Improvement System Rating Scales

WEMWBS Warwick-Edinburgh Mental Well-Being Scale

VABS Vineland Adaptive Behavior Scales

Vineland SEEC Vineland Social-Emotional Early Childhood Scales

### Databases

CDSR Cochrane Database of Systematic Reviews

CENTRAL Cochrane Central Register of Controlled Trial

CINAHL Cumulative Index to Nursing and Allied Health Literature

DARE Database of Abstracts of Reviews of Effects

EMBASE Excepta Medica Database

ERIC Education Resources Information Centre

HTA database Health Technology Assessment Database

Medline Medical Literature Analysis and Retrieval System Online

PsycINFO Psychology Information Database

## A2.2 Glossary

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| **Bias** | Bias is a systematic deviation of a measurement from the ‘true’ value leading to either an over- or under-estimation of the treatment effect. Bias can originate from many different sources, such as allocation of patients, measurement, interpretation, publication and review of data. |
| **Blinding** | The concealment of group assignment – to either the treatment or control group – from the knowledge of study participants, assessors, and/or investigators in a clinical trial of whether an intervention/treatment being administered is a placebo treatment (ie, the control group) or the intervention/treatment being investigated. |
| **Case series** | Case series are collections of individual case reports, which may occur within a fairly short period of time. Cases consist of either only the exposed people with the outcomes, or people with the outcome regardless of the exposure. In neither of these examples can the risk for the outcome be determined. |
| **Controlled clinical trial** | Controlled trials where, unlike RCTs, allocation to condition has not been explicitly stated as fully randomised. Also applies to pseudo-randomised controlled trials where allocation method is known but not strictly random (see under pseudo-randomised controlled trial). |
| **Cross-over randomised trial** | An experiment in which people receive one intervention (or control) and then cross-over to receive the alternate comparison condition at a point in time. The order that individuals receive the intervention and comparison condition is randomised per session for each individual. |
| **Cross-sectional study** | A study that examines the relationship between exposures (eg, risk factor) and outcomes (eg, disease), as they exist in a defined population, at a particular time. A group of people are assessed at a particular point (or cross-section) in time and the data collected on outcomes relate to that point in time; ie, proportion of people with asthma in October 2014. This type of study is useful for hypothesis-generation, to identify whether a risk factor is associated with a certain type of outcome, but more often than not (except when the exposure and outcome are stable; eg, genetic mutation and certain clinical symptoms) the causal link cannot be proven unless a time dimension is included. |
| **Detection bias** | Detection bias refers to systematic differences between groups in how outcomes are determined. Awareness by outcome assessors/respondents of whether an intervention was received or not (ie, they are not blind to allocated condition) may increase the risk of their measurements/ratings/ reports being affected by detection bias. |
| **Effect size** | A quantitative measure of the strength of a phenomenon, a standardized measure of the size of the difference between two groups. The effect sizes can be interpreted in accordance with common guidelines for interventions in the behavioural sciences 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 [61]. |
| **Effectiveness** | A measure of the extent to which a specific intervention, procedure, regimen, or service, when deployed in the field in routine circumstances, does what it is intended to do for a specified population. |
| **Generalisability** | Applicability of the results to other populations. |
| **High functioning** | Whilst it is acknowledged that the term “high functioning” is not universally favoured, in this report, the term “high functioning” is used to refer to people with higher cognitive functioning either as established by intelligence tests (generally indicated by full IQ scores of 70 or above), or through the diagnosis of “high-functioning autism” or Asperger syndrome (under DSM‑IV criteria). In light of the removal of Asperger syndrome as a separate diagnostic classification in [DSM-5](http://en.wikipedia.org/wiki/DSM-5), these distinctions may no longer be used clinically. |
| **Level of evidence** | Levels within a hierarchy of study evidence that indicates the degree to which bias has been eliminated in the study design. For example, see **Appendix 1, Table A1.1**. |
| **Mean** | Calculated by adding all the individual values in the group and dividing by the number of values in the group. |
| **Neurodiversity** | An approach to learning and disability which suggests that diverse neurological conditions appear as a result of normal variation in the human genome. This term was coined in the late 1990s as a challenge to prevailing views of neurological diversity as inherently pathological, and it asserts that neurological differences should be recognized and respected as a social category on a par with gender, ethnicity, sexual orientation, or disability status. |
| **Neurotypical** | An abbreviation of neurologically typical, a term coined in the autism community as a label for people who are not on the autism spectrum, otherwise referred to as typically developing people. |
| **Observational studies** | Also known as epidemiological studies. These are usually undertaken by investigators who are not involved in the clinical care of the patients being studied, and who are not using the technology under investigation. Distinct from experimental studies. |
| **Performance bias** | Performance bias refers to systematic differences between groups in the care that is provided, or in exposure to factors other than the interventions of interest. After enrolment into the study, blinding (or masking) of study participants and personnel may reduce the risk that knowledge of which intervention was received, rather than the intervention itself, affects outcomes. Effective blinding can also ensure that the compared groups receive a similar amount of attention, ancillary treatment and diagnostic investigations. Blinding is not always possible, however. |
| **Post-test** | Case series where only outcomes after the intervention (factor under study) are recorded in the series of people, so no comparisons can be made. |
| **Pre-test/ post‑test** | Case series where measures on an outcome are taken before and after the intervention is introduced to a series of people and are then compared (also known as a ‘before- and-after study’). |
| **Power** | The probability that a statistical test or study will detect a defined pattern in data and declare the extent of the pattern as showing statistical significance. |
| **Prevalence** | A measure of the proportion of people in a population who have some attribute or disease at a given point in time or during some time period. |
| **Pseudo-randomised controlled trial** | As for a randomised controlled trial except that a pseudo-random method (such as alternate allocation, days of the week, date of birth, or odd-even medical record numbers) is described for allocating individuals into treatment or control group conditions. The outcomes from each group are compared. Sometimes known as quasi-randomised controlled trials. Can be regarded as clinical controlled trials (CCT). |
| **Quality of evidence** | Degree to which bias has been prevented through the design and conduct of research from which evidence is derived. |
| **Randomised controlled trial (RCT)** | An experiment in which a unit (eg, people, or a cluster of people) are allocated using a fully random mechanism (such as a coin toss, random number table, computer-generated random numbers) into either the intervention condition (eg, preventive or therapeutic procedure, manoeuvre, or treatment) or a control comparison condition (eg, placebo, usual care, alternative treatment). The outcomes from each group are compared. Conditions are run in parallel. See also cross-over randomised controlled trial and pseudo-randomised trial. |
| **Secondary study** | An analysis or synthesis of research data reported elsewhere, including systematic reviews, meta analyses and guidelines. |
| **Selection bias** | Error due to systematic differences in characteristics between those who are selected for inclusion in a study and those who are not (or between those compared within a study and those who are not). |
| **Strength of evidence** | The strength of evidence for an intervention effect includes the level (type of studies), quality (how well the studies were designed and performed to eliminate bias) and statistical precision (P-value and confidence interval). |
| **Systematic review (SR)** | A literature review reporting a systematic method to search for, identify and appraise a number of independent studies. |
| **Treatment effect** | An effect attributed to a treatment (intervention), which in a clinical trial is based on a comparison between active treatment and a placebo control, or two or more treatment regimens. |
| **Whānau** | Extended family |

# Appendix 3: Evidence Tables of included studies

Tables are ordered by study type (primary then secondary studies), and then within each table, according to the following hierarchy: year of publication (oldest first), and alphabetically (by first author’s surname).

Table A3.1: Evidence Tables for included primary studies

| **Schwartzberg et al (2013) [53]** | | | | | |
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| **Country, study, aims** | **Participants** | **Selection criteria** | **Intervention, control, outcomes** | **Results** | **Conclusions, quality issues** |
| **Country:** US  **Study type:** pseudo-randomised controlled trial (RCT)  **Evidence level:** III.1  **Study Quality** (SIGN checklist): – (low quality)  **Aims:** to examine the effects of music-based social stories on social skills in children on the autism spectrum  Note: both control and treatment group received music therapy, with the varied condition being one component (sung or read social story). | **Setting:** Recruited 2 weeks prior to one-week summer camp for young people on the autism spectrum  **Participants:** 30 (of 87 recruited and randomised) children and young people diagnosed with ASD; aged 9–21 years (M=15.8 years); 29 (97%) male. Participants cluster randomised into one of 6 groups: 3 treatment and 3 control groups.  3 Treatment groups (TG, each with one of 3 *sung* social stories): N=16  3 Control groups (CG, each with one of 3 *read* social stories): N=14  Dropout: 57/87 (65%) who were allocated at pre-test did not return their post-test results and were therefore excluded.  **Randomisation by cluster.** Method not presented. | **Inclusion:** diagnosed with ASD; attending summer camp; gave consent and returned completed pre- and post-test measures.  **Exclusion:** non-completion of post-test assessments.  **Assessment intervals:** pre‑test 0-2 weeks before camp, every day x 3 of music therapy, and post-test one-week after camp  **Fidelity:** Social stories (both conditions) delivered by a Board certified, experienced Music Therapist who also trained camp counsellors. Observed no guidance of responses by counsellors.  **Analysis:** repeated measures ANOVAs. Intention to treat not used such that those not completing all assessments were excluded. | **Treatment (TG):** social story sung together (novel composition), integrating PECS (3 versions: targeting social reciprocity, social participation, and detrimental social behaviours) within 50 minute music therapy sessions (also including songs, movement and music, instrument playing, relaxation and music) x 3 (consecutive days) in groups of 6 campers and 8 staff.  **Control (CG):** social story read together within therapy session  **Outcomes**  Autism Social Skills Profile (ASSP): generalisation of target social skills with 3 sub-categories: social reciprocity (SR); social participation (SP); detrimental social behaviours (DSB) *(completed by legal guardians)*  Comprehension Checks (CC): measured comprehension of each social story *(administered by camp counsellors to participants)*  **Blinding:** Assessments were not blind to condition. | At baseline, no analyses reported of whether there were differences between groups. Also no analyses of whether those who dropped out at post-test differed from participants. Therefore unclear on whether randomisation was effective or drop-outs were different.  **Key findings**  4-way repeated measures ANOVA for ASSP found no main effects for condition (treatment versus control).  3-way repeated measures ANOVA for CC only found a main effect for time, F(4,96)=2.84, p=0.028, partial =0.106 such that comprehension of social stories (as expected) improved between pre- and post-test. | **Author conclusions:** Lack of significant main effects for social skills was likely a function of minimal treatment dose. Use of active control (read social story) may have prevented between group differences, and dose may not have been sufficient (3 sessions). Clinicians might usefully pair social stories with music to facilitate comprehension, generalisation and on-task behaviours conducive to learning social information.  **Reviewer’s comments:** verification of ASD diagnoses not mentioned. Moderate sample size with only one girl. Very high dropout of 65% who were excluded from the study without any analyses at pre-test, and no intention to treat analyses. Lack of no music therapy control group. Assessment was not blind to condition. Small samples for each treatment subgroup reducing power to identify real effects. Social skills behaviour not independently measured. Maintenance not assessed.  As treatment and control conditions received music therapy, additional feature of music-based social story may not have provided additional benefit.  **Source of funding:** none reported. |

| **LaGasse et al (2014) [32]** | | | | | |
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| **Country, study, aims** | **Participants** | **Selection criteria** | **Intervention, control, outcomes** | **Results** | **Conclusions, quality issues** |
| **Country:** US  **Study type:** parallel-group randomised controlled trial (RCT)  **Evidence level:** II  **Study Quality** (SIGN checklist): + (acceptable quality)  **Aims:** to examine the effects of a music-therapy (MT) group intervention on children on the autism spectrum | **Setting:** Recruited by word of mouth and flyers in one large metropolitan area.  **Participants:** 17 (of 25 recruited and randomised) children diagnosed with ASD; aged 6–9 years (M=7.6 years); 13 (76%) male.  Treatment group (TG, music therapy group): N=9.  Control group (CG, social skills group): N=8.  **Dropout:** 5/22 (23%) including 3 withdrawals and 2 excluded due to non-attendance at two or more sessions (higher for CG). Three week follow-up assessments not completed by 4 parents (2 in each group).  Randomisation by computerised random numbers table. | **Inclusion:** formal diagnosis with ASD; primary language of English.  **Exclusion:** dual diagnosis, having had music therapy within previous 2 years, had 2 or more absences from sessions.  **Assessment intervals:** SRS *(parent):* pre-test before sessions, and post-test 3 days after 5 week programme. ATEC *(parent):* pre-test, at session 2, 4, 8, 1 week post, and 3 weeks post. ATEC *(therapist*): after sessions 2, 4, 8, 10.  **Fidelity:** MT delivered by Board certified Music Therapist. Manual defined exercises, support level, fidelity measures.  Inter-rater reliability for coding of behaviours in videos between 0.82 and 0.94.  **Analysis:** Used mixed and repeated ANOVAs for between group analyses. ANCOVA for video sessions with session 3 as covariate, session 10 as dependent variable. | **Treatment (TG):** 50-minute music therapy session x 5 weeks x 2/wk (10 sessions) in groups of 3–4 children and music therapist (and 2 helpers). Manualised sessions included songs, and exercises offering sensory and social experiences, including playing instruments moving to musical cues.  **Control (CG):** group exercises (games, ball passing, copying movements) without music led by educator (and 2 helpers).  **Outcomes**  Social Responsiveness Scale (SRS): level of impairment of social skills *(completed by parent/caregiver)*  Autism Treatment Evaluation Checklist (ATEC): functioning in speech and communication, sociability, sensory/cognitive awareness, physical wellbeing *(completed by parent, therapist)*  Video analysis of sessions 3rd and 10th to rate random 5-minute clips for eye gaze, joint attention, initiating and responding to communication (by gesture, AAC, speech), withdrawal behaviours *(2 music therapy researchers)*  **Blinding:** Parents, children, teachers not blind to group. Assessment blind for therapist on ATEC, videos not blind to condition, but blind to session number. | At baseline, no group difference in attendance, age, level of functioning (CARS). No difference at pre-test between drop-outs and completers.  **Key findings:**  SRS: Two-way mixed ANOVA found significant Time x Group interaction: F(1.14)=5.646, *p*=0.032, partial =0.287; paired samples *t*-test indicated improvement in SRS scores post-test cf pre-test for TG (*p*=.018), but not for CG.  Parent-rated ATEC: Repeated measures ANOVA found main effect for Time but not Group, and Time x Group interaction not significant: (F(5,68)=2.29, *p*=0.0549. Given trend and low sample sizes, Tukey post hoc analyses run between intervals. Music group improved in ATEC at 3 week post-test follow cf baseline (t(68)=3.88, *p*=0.0031).  Therapist-rated ATEC: Repeated measures ANOVA found main effect for Time but not Group, and the Time x Group interaction did not reach significance (*p*=.157).  Video analysis: ANCOVA found between-group difference for eye gaze F(1.14)=6.669, *p*=0.022, partial =.323, joint attention with peers F(1.14)=5.735, *p*=0.031, partial =0.291; TG better than CG.  No group differences in initiation or response to communication, joint attention with adults, or withdrawal behaviour. High variability between individuals. | **Author conclusions:** There are indications that music therapy improved parent-reported social behaviours. Music therapy may improve joint attention and eye gaze towards peers.  **Reviewer’s comments:** ASD diagnoses were verified. Small sample size. Severity of disability and language ability not reported. Groups did not differ at baseline, and dropouts (n=5) not different to completers at baseline. Assessment was not blind to condition for parent measures, was blinded for therapist-assessed ATEC, and blind to session number for video analysis. Good inter-rater reliability for assessments of behaviour from videos. Maintenance at 3‑weeks post-test assessed for ATEC only.  Intention to treat analyses not conducted and those not attending 2 or more sessions were excluded. Small samples may be under-powered to identify real effects, particularly at follow-up. The effectiveness of the social skills group itself may have masked benefits of MT such that both were equally effective (significant effects for Time but not Group) for ATEC. Potential for bias in parental assessment which was unblinded.  Source of funding: Arthur Flagler Fultz Research Fund of the American Music Therapy Association. |

| **Porter et al (2017) [56]** | | | | | |
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| **Country, study, aims** | **Participants** | **Selection criteria** | **Intervention, control, outcomes** | **Results** | **Conclusions, quality issues** |
| **Country:** UK  **Study type:** multicentre parallel-group randomised controlled trial (RCT)  **Evidence level:** II  Study Quality (SIGN checklist): + (acceptable quality)  **Aims:** to examine the efficacy of music therapy (MT) in clinical practice for young people with social, emotional, behavioural, and developmental problems. As part of this larger trial, only data relating to the sub-analysis for autistic children are reported here. | **Setting:** Recruited from 6 Child and Adolescent Mental Health Service (CAMHS) community care facilities in Northern Ireland. Therapy took place in private room at CAMHS.  **Participants:** Whole sample N=181 (of 251 recruited and randomised) child and parent dyads; children with social, emotional, behavioural and developmental difficulties aged 8–16 years (59% over 13 years); (44%) male; 97% white, 31% from most deprived and 26% from least deprived areas.  Of these, a subgroup of 34 (of 47 randomised) had diagnosis of ASD:  Treatment group (TG): N=18 (of 76 in total sample).  Control group (CG): N=16 (of 105).  **Dropout:** 13/34 (18%) including 5/23 (22%) of TG and 8/24 (33%) of CG at 1 week post-test.  Randomisation by computer generated sequence stratified by recruiting centre, with concealed allocation. | **Inclusion:** registered with CAMHS; social, emotional, behavioural and developmental difficulties, aged 8–16 years.  **Exclusion:** substance abuse, psychosis, suicidal behaviour, inability to complete measures or attend MT.  **Assessment intervals:** baseline, 1 week after MT (13 weeks post baseline) and 13 weeks follow-up (26 weeks post-baseline).  **Fidelity:** one accredited Music Therapist with ongoing supervision and monitoring of videos. Manualised MT. Attendance ≥ 10 of 12 sessions (for whole sample).  **Analysis:** Intention to treat analysis regardless of sessions completed. Effect sizes calculated difference in between group means, adjusted for corresponding baseline score using ANCOVA. | **Treatment (TG):** 30 minute individual music therapy session x 12 weeks x 1/wk with music therapist. Received Alvin model of free improvisation encouraging client to create music through voice, instrument or movement with tailored support from therapist. Sessions led by the participant encouraged to choose an instrument to express their feelings.  Also received usual care.  **Control (CG):** usual care of psychiatric counselling, and/or medication.  **Outcome**  Social Skills Improvement System Rating Scales (SSIS): communicative and interactional skills *(completed by young person, and by parent)*  Note: other outcomes assessed but analyses not reported for subgroup analyses of autistic participants.  **Blinding:** Researchers blind to group when recording assessments. Therapist, parents and children not blind to group. | At baseline for whole sample analyses, characteristics were similar between groups, and similar between participants and those lost to follow-up. Analyses not reported.  **Key findings**  No significant group differences in the communicative and interactional skills (SSIS) completed by young person, or by parent. (Also, no difference in autism subgroup and those without autism for SSIS completed by young person or parent). | **Author conclusions:** (for the whole sample) “While the findings provide some evidence for the integration of music therapy into clinical practice, differences relating to subgroups and secondary outcomes indicate the need for further study”. With respect to the autism sub-group, the study does not add significant evidence because the number of participants was “not great enough to enable sufficiently powered statistical analysis”.  **Reviewer’s comments:** Small sample size for autism sub-group within a larger study. High dropout rates for completing assessments. Whilst some descriptive data and analyses were only available for the whole group, these suggested groups did not differ at baseline, or status of retention, though more people dropped out of the CG than the TG in the autism sub-group. Assessment was single blind to condition for researcher interviewing respondents but the reports of the participants and parents were unblinded. Processes evident to support programme fidelity.  Other outcome measures and longer follow up to assess maintenance not reported for subgroup analysis of autistic participants.  Small samples may be under-powered to identify real effects. Potential for bias in participant and parental reports (not blind to condition).  **Source of funding:** the Big Lottery Fund. |

| **TIME‑A Trial (2017) [54, 55]** | | | | | |
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| **Country, study, aims** | **Participants** | **Selection criteria** | **Intervention, control, outcomes** | **Results** | **Conclusions, quality issues** |
| **Country:** Multi-site across 9 countries (Australia, Austria, Brazil, Israel, Italy, Korea, ,Norway, UK, US).  **Study type:** multi-site randomised controlled trial (RCT).  **Evidence level:** II  **Study Quality** (SIGN checklist): + (adequate quality).  **Aims:** to evaluate effects of improvisational music therapy on generalised social communication skills of children on the autism spectrum. | **Setting:** outpatient settings (clinics, kindergartens, family homes).  **Participants:** N=364 (of 570 eligible recruited) children aged 4–7 years diagnosed with ASD (M=5.4 years); 83% male; 45% with low cognitive levels (IQ<70) impairment.  Treatment group – Low Intensity (TG-LI): N=92.  Treatment group – High Intensity (TG-HI): N=90.  Control group (CG): N=182.  **Dropout:** 50 (14%) lost to follow up at 5 months (immediately post-test), more in CG (n=33, 18%) than TG (n=17, 9%).  Computer generated randomisation 1:1:2 ratio stratified by site and randomly varying block sizes of 4 and 8.  UK sites: 2 of the sites (both in the UK) also reported on two additional measures of parental stress and wellbeing [55].  ... *continued* | **Inclusion:** meeting ICD‑10 criteria for ASD, aged 4–7 years.  **Exclusion:** serious sensory issues (blindness, deafness), having received MT within previous 12 months, inability to provide consent.  **Assessment intervals:** baseline, 2 months and 5 months post baseline (at end of treatment), and 12 months (7 months follow‑up).  **Fidelity:** 30 qualified Music Therapists at least to degree level. Sessions video-taped and random segments assessed by 2 independent raters who rated 93% of sessions as conducted adequately. Manual used. As a pragmatic trial, 93% of TG actually received MT.  87% parents received parent advice and support sessions. Most common concomitant other therapies were SLT (58%); and sensory-motor therapy or physiotherapy (41%).  ... *continued* | **Treatment (TG):** 30-minute individual (possibly with family) improvisational music therapy sessions over 5 months:  TG-LI: 1 weekly session (median=15 sessions).  TG-HI: 3 weekly sessions (median=34 sessions).  Music therapist sang or played instruments with child, using improvisational techniques of mirroring, synchronisation, or grounding.  Also received “enhanced standard care” including clinician-led advice and support sessions for parents (up to 3 x 60 minute sessions) and usual therapy (median=36 sessions, 31 for high intensity group and 40 for low intensity.)  **Control (CG):** enhanced standard care of 3 parent sessions, (median of 45 therapy sessions).  **Outcomes**  *Primary outcome:*  Autism Diagnostic Observation Schedule (ADOS), social affect: autism symptom severity *(Clinician)*  Social Responsiveness Scale (SRS): autism symptom severity total scale 5 subscales (social awareness, social cognition, social communication, social motivation, and autistic mannerisms) *(Parent)*  ... *continued* | At baseline, groups were described as well-balanced between groups (tests not reported) in sex, age, diagnosis, IQ, language at home, parent’s education and employment, family size, child care. No group differences in treatment fidelity. No differences in baseline characteristics between those dropping out and staying.  **Key findings**  ADOS social affect (at 5 months):  LMEM found no significant Group x Time interaction in social affect between baseline and 5 mths post-test for TG (14.08 to 13.23) cf CG (13.49 to 12.58): mean difference=0.06, 95% CI[-0.70-0.81]; *p=0.88)*. No difference at 12 follow up.  No significant differences between high- or low- intensity MT and CG.  No group effects for age, autism subtype, music therapist.  Proportion of children responded in ADOS Social Affect Post hoc analyses found more responders in TG (52%) than CG (42%) at 5 mths (RR=1.25 [95% CI: 1.0‑1.56; Risk difference=0.10 [95% CI: 0.00-0.21] *p=0.047).*  More responders for those receiving ≥15 sessions of MT (58%) cf CG (42%): RR=1.39, 95% CI[1.11-1.74; Risk difference=0.16, 95% CI[0.05-0.27] *p=0.004).*  ... *continued* | **Author conclusions:** Improvisational MT compared with enhanced standard care resulted in no significant difference in autism symptom severity over 5 months (of treatment) based on the ADOS social affect domain. The few significant outcomes were small and unlikely to be clinically important. These findings do not support the use of Improvisational MT for symptom reduction in children on the autism spectrum.  **Reviewer’s comments:** Good sample size. High number of eligible people declining (n=206), mostly due to logistical reasons around attendance. Diagnoses confirmed by researchers. Programme fidelity was adequate. Extended post treatment follow up (7 months post) assessed.  Groups did not differ at baseline in except for higher drop-out rate in CG. No dose effects for intensity. A higher proportion of responders in the TG needs to be treated with caution given differential group attrition. MT groups received fewer other therapies than CG so possibility of uncontrolled effect.  Symptom severity (ADOS) primary outcome may not be sensitive to changes. Qualitative work [59] found that the MT was well received, enjoyed and perceived as positive.  The few significant differences found for secondary outcomes were small, exploratory, non-blinded, and were not adjusted for the multiplicity of tests.  ... *continued* |

| **TIME‑A Trial (2017) [54, 55] *continued ...*** | | | | | |
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| **Country, study, aims** | **Participants** | **Selection criteria** | **Intervention, control, outcomes** | **Results** | **Conclusions, quality issues** |
|  | ... *continued*  This sub-sample included N=81 children randomised to TG-LI (n=20), TG-HI (n=21), and CG (n=40). | ... *continued*  **Analysis:** Intention to treat analysis. Linear mixed-effects models (LMEM). Sensitivity analyses tested influence of missing data, therapist, site, and planned subgroup analyses for age and ASD subtype. | ... *continued*  Quality of Life 100mm visual analogue scales: QoL of participant, and QoL of family *(completed by parent)*  UK sites only: Parenting Stress Index (PSI) (parental stress), and Warwick-Edinburgh Mental Well-Being Scale (WEMWBS) (parental wellbeing) *(both completed by parents)*  Blinding: Assessors blind to group, but broken unintentionally 15 times for TG (8%) and 5 (3%) times for CG. Therapist, children and families not blinded. | ... *continued*  SRS: Small group effect for SRS mannerisms at 5 mths favouring TG cf CG. No difference for other 4 SRS subscales.  For 3-group tests considering intensity:  Small improvement found for SRS Autistic mannerisms for TG-HI cf CG at 5 months.  Quality of Life: post hoc comparisons found higher participant QOL (rated by parents) for TG-HI cf CG at 5 months. No difference for QoL of family. No serious adverse events reported.  UK sites only: No group differences at 5 months for parental stress, or at 5 or 12 months for parental wellbeing. Trend for lower parental stress at 12 months (p=0.05). However, response rates were 49% at 5 months and 53% at 12 months. | .. *continued*  UK site comparisons open to reporting and attrition biases due to poor response at follow‑up.  **Source of funding:** Primarily by the Research Council of Norway, with additional funding in each participating country from academic and government health research bodies with no role in study design, analysis or publication decisions. |

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| **Sharda et al (2018) [34]** | | | | | |
| **Country, study, aims** | **Participants** | **Selection criteria** | **Intervention, control, outcomes** | **Results** | **Conclusions, quality issues** |
| **Country**: Canada  **Study type**: parallel-group randomised controlled trial (RCT)  **Evidence level**: II  **Study Quality** (SIGN checklist): ++ (high quality)  **Aims:** to examine the effect of music intervention on social communication and brain connectivity in school-aged children with autism. | **Setting**: screened “in the community”. Appears to be University-based study.  **Participants**: Whole sample N=51 (of 57 eligible recruited) children aged 6-12 years diagnosed with ASD (M=10.3 years); 84% male; 53% with language impairment; non-verbal IQ M=106; full-scale IQ=98.  Treatment group (TG): N=26.  Control group (CG): N=25.  **Dropout**: 1/26 (4%) of TG only dropped out before post-test assessment. N=6 excluded from brain imaging due to motion or missing data.  Randomisation by coin toss or stochastic covariate adaptive minimisation algorithm. | **Inclusion:** meeting DSM-IV criteria for ASD, aged 6–12 years.  **Exclusion**: receiving individual music therapy within last 6 months; private music lessons up to 1 year; group music therapy at school; <35 weeks’ gestation; hearing disorders; neurological disease.  **Assessment intervals**: baseline, after treatment (8–12 weeks post baseline).  **Fidelity**: one accredited Music Therapist. Sessions video-taped and analysis by 2 independent coders of 20% demonstrated high content fidelity (80-100%) and content fidelity (>75%). Treatment protocols. Attendance M=10.3 sessions.  **Analysis**: Intention to treat analysis. Linear mixed-effects models (LMEM), independent samples t-tests, ANCOVA, linear regression models. Bonferroni adjusted alpha levels. | **Treatment (TG)**: 45 minute individual music therapy session x 8–12 weeks x 1/wk with music therapist. Child-centred approach using instruments, songs and rhythmic cues.  **Control (CG)**: Structurally matched active control play-based intervention.  **Outcomes**  *Primary outcomes:*  Social Responsiveness Scale (SRS-II): autism symptom severity *(completed by Parent)*  Children’s Communication Checklist (CCC-2): pragmatic communication *(Parent)*  Peabody Picture Vocabulary Test (PPVT-4): receptive vocabulary *(direct assessment)*  *Secondary outcomes:*  Vineland Adaptive Behavior Scales – maladaptive behaviour (VABS-MB): maladaptive behaviour *(Parent)*  Beach Family Quality of Life Scale (FQoL): family QoL *(Parent)*  MRI scan assessing Resting state functional connectivity (RSFC): functional brain connectivity of fronto-temporal brain networks  **Blinding:** Assessors blind to group. Therapist and children not blinded. Parents not told of group allocation but 61% were aware. | At baseline, groups did not differ on sex, age, language, motor skills, IQ, SES or musical ability. No group differences in treatment fidelity.  **Key findings**  CCC-2: LMEM found Group x Time interaction such that TG showed greater improvements than CG; (*ß=-1.35, p=0.01;* post Tukey test *t*=1.43, *P*=0.024) with 15/26 in TG showing improvement cf 5/24 in CG. This represents a medium-sized positive effect (d=0.34). Significant differences in subtests for Speech (*P*=0.01), Semantics (*P*=0.046), Pragmatics, Inappropriate Initiations (*P*=0.006), Social Relations (*P*=0.048), and Interests (*P*=0.02).  FQoL: There was a significant Group x Time difference *(ß=-1.9, p=0.01)* favouring the TG, particularly in terms of family interaction, cohesion and coping and benefits on disability-related supports.  RSFC: In ANCOVA models, greater RSFC (brain connectivity) post intervention in TG cf CG between auditory, and striatal and motor regions; and reduced RSFV (brain connectivity) in TG cf CG between auditory and visual regions.  In linear regression models, these effects were related with greater improvement in CCC-2 (communication) scores.  ... *continued* | **Author conclusions**: Study provides evidence that 8–12 weeks of a music intervention can improve social communication and functional brain connectivity, in ways specific to music therapy. There appear to be limited effects on reducing autism symptom severity or improving receptive vocabulary.  For the music therapy group, there was increased brain connectivity between bilateral primary auditory cortex and subcortical and motor regions, and reduced over-connectivity between auditory and visual-association areas, which were related to behavioural improvements in communication skills. This supports the bottom-up integration of sensorimotor brain networks leading to social functioning rather than top-down music-based reward.  **Reviewer’s comments**: Moderate sample size. High participation and very low dropout rates. Diagnoses confirmed by researchers. Groups did not differ at baseline in characteristics, assessments, or drop-out rate. High programme fidelity. Assessment was blind to condition for researcher but for parents, whilst blinded, over 60% aware of group allocation.  Longer follow-up not reported.  Potential for bias in 60% of parental reports.  **Source of funding**: none reported. Researchers based at University of Montreal. |

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| **Sharda et al (2018) [34] *continued*** | | | | | |
| **Country, study, aims** | **Participants** | **Selection criteria** | **Intervention, control, outcomes** | **Results** | **Conclusions, quality issues** |
|  |  |  |  | ... *continued*  No significant Group x Time differences in SRS-II, or PPVT-4. Both groups showed reduced maladaptive behaviours on the VABS (*ß=0.22, p=0.01)*, but there was no significant group interaction. |  |

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| **Rabeyron et al (2018) [20]** | | | | | |
| **Country, study, aims** | **Participants** | **Selection criteria** | **Intervention, control, outcomes** | **Results** | **Conclusions, quality issues** |
| **Country:** France  **Study type:** parallel-group randomised controlled trial (RCT)  **Evidence level:** II  **Study Quality** (SIGN checklist): + (acceptable quality)  **Aims:** to examine if music therapy is more effective than simply listening to music for children on the autism spectrum. | **Setting:** recruited from 5 psychiatric day-care facilities.  **Participants:** Whole sample N=37 (of 40 eligible recruited) children aged 4–7 diagnosed years with ASD (M=5.1 years); 88% male; IQ (M=52.2, SD=3.2, range 49–62).  Treatment group (TG): N=19.  Control group (CG): N=18.  **Dropout:** 1/37 (3%) of CG only dropped out before post-test assessment.  Randomisation by generated randomised list. | **Inclusion:** validated diagnoses of ASD based on the CARS, aged 4–7 years.  **Exclusion:** auditory impairment, neurological diseases (excepting stabilised epilepsy), using psychotropic medication, refusal.  **Assessment intervals:** baseline, after treatment (8 months post baseline).  **Fidelity:** one trained graduated Music Therapist. Fidelity and attendance not reported. No recording of sessions. Not clear how many groups there were, possibly only two.  **Analysis:** Repeated measures regression analyses. | **Treatment (TG):** 30-minute group-based music therapy x 1/week (except school holidays) over 8 months sessions with music therapist (attended by co‑therapist, trainee clinical psychologist). Included instrumental and vocal improvisation.  **Control (CG):** 25 x 30‑minute group-based over 8 months active control, music-listening intervention (same music).  **Outcomes**  *Primary outcomes:*  Clinical Global Impression – Improvement (CGI-I): autism symptom severity *(completed by Clinician)*  *Secondary outcomes:*  Childhood Autism Rating Scale (CARS): autistic symptoms *(completed by Clinician)*  Aberrant Behavior Checklist (ABC): non-adaptive behaviour *(completed by Clinician)*  **Blinding:** Assessors blind to group, not participants or parents. | At baseline, groups did not differ on sex, age, or IQ. More females in CG (n=1, 5%) cf TG (n=4, 23%) and so analyses were adjusted for gender. Outcomes measured at baseline did not differ significantly between groups for CGI, or CARS. The TG had higher baseline scores/ less adaptive behaviour (M=58.7) cf CG (M=53.1).  **Key findings**  CGI: significant Group x Time interaction such that TG showed greater improvements than CG; (OR*=*0.44*,* 95% CI [0.200–0.93]; *p=0.017), d*=0.80 (large).  CGI improvement: proportion of children showing improvement of 1 point in CGI was higher in TG (90%) cf CG (82%) (OR=5.56, CI 95% [0.68–33.76]. Sensitivity analyses: stronger effect when those improving by 2 points on the CGI were compared: TG (63%) cf CG (29%) (OR=7.36, CI 95% [1.76–36.60]. No change when controlling for gender.  CARS: Both groups improved similarly at follow-up with no Group x Time interaction for total score, and all 15 subscales, when controlling for gender.  ABC: Higher baseline scores for MT cf CG no longer evident when controlling for gender (p=0.91). Time by group interaction (p=0.03) such than TG improved > CG at follow-up. And Group x Time interactions: MT improved cf CG for ABC sub-scores of *lethargy*, and *stereotypy*, controlling for gender. No differences for *irritability, inappropriate speech, hyperactivity*. | **Author conclusions:** Music therapy is more “efficient” than music listening for children on the autism spectrum. MT should be considered as an add-on to relevant healthcare programmes.  Greater improvement in symptoms in music therapy group compared with control music listening group. Clinical improvement was associated with the improvement of autistic symptoms on lethargy and stereotypy subscales.  **Reviewer’s comments:** Moderate sample size, included people with low cognitive ability. Good participation and low dropout rates. Long intervention period. Groups differed at baseline. CG included more females, and showed less adaptive behaviour (on ABC) at baseline, than MT group. ABC difference disappeared when gender controlled for in analyses.  Programme fidelity not reported and sessions not videotaped. Therapists in CG found it difficult to remain neutral. Some bleeding of treatment may bias toward reduced group effects.  Lack of specific social and communication outcomes. No accounting for missing data. Sub-scale analyses post hoc. No accounting for multiple tests and chance effects. Effects on lethargy (ABC subscale) extremely small (*d*=0.02). No direct observation during sessions. Blinded assessment. Longer follow up not undertaken.  **Source of funding:** Entrependre pour Aider, and Academie Francaise. |

**Key:** AAC=alternative and augmentative communication; ABC=Aberrant Behavior Checklist; ADOS=Autism Diagnostic Observation Schedule; ANOVA=analysis of variance; ANCOVA=analysis of covariance; ASD=Autism Spectrum Disorder; ASSP=Autism Social Skills Profile; ATEC=Autism Treatment Evaluation Checklist; cf=compared with; CARS=Childhood Autism Rating Scale; CCC=Children’s Communication Checklist; CC=comprehension checks; CGI=Clinical Global Improvement Scale; CHMAS= Child and Adolescent Mental Health Service; DSM-IV=Diagnostic and Statistical Manual of Mental Disorders, 4th edition; FQoL=Beach Family Quality of Life; ICD-10=International Statistical Classification of Diseases and Related Health Problems; IQ=intelligence quotient; LMEM=linear mixed-effects models; M=mean; mth=month; ns=not significant; OR=Odds Ratio; PECS=Picture Exchange Classification System; PSI=Parental Stress Index; /wk=per week; QoL=quality of life; RCT=randomised controlled trial; RSFC=Resting state functional connectivity; SES=social-economic status; SIGN=Scottish Intercollegiate Guidelines Network; SLT=speech and language therapy; SRS=Social Responsiveness Scale; SSIS= Social Skills Improvement System Rating Scales; UK=United Kingdom; US=United States of America; WEMWBS=Warwick-Edinburgh Mental Well-Being Scale; VABS=Vineland Adaptive Behavior Scales; /wk=per week.

Table A3.2: Evidence Tables for included secondary studies

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| **Geretsegger et al (2014) [23]** | | | | |
| **Country, study type, aim** | **Search strategy** | **Appraisal methods** | **Results** | **Conclusions** |
| **Country**: Norway  **Study type**: systematic review and meta-analysis  **Evidence level**: I  **Study Quality** (SIGN checklist): ++ (high quality)  **Aims:** to assess the effects of music therapy for individuals on the autism spectrum | **Databases**: CENTRAL, Ovid MEDLINE, EMBASE, LILACS, PsycINFO, CINAHL, ERIC, ASSIA, Sociological Abstracts, and Dissertation Abstracts International, Music Therapy Research, Music Therapy World.  **Search**: Searched from 2004–29 July 2013, updating a Cochrane review from 2006 [37]. Transparent selection criteria. Range of keywords relating to music therapy and autism. Searching of reference lists, contacted authors, music therapy conference proceedings.  **Selection criteria**: randomised controlled trials, pseudo-randomised trials, or randomised cross-over trials (including single case experimental studies where order of condition is randomised between participants) comparing music therapy with placebo, no treatment, or standard care for individuals on the autism spectrum of any age. Music therapy defined as regular sessions delivered by a music therapist where social and emotional interaction is promoted through musical experiences and the relationships developed through them.  Excluded: single case experimental studies without randomised order of condition. | **Method:** Three authors independently selected studies, and two authors extracted data from included studies, and appraised studies (using the Cochrane Risk of Bias tool).  Effect size was indicated by the pooled standardised mean difference (SMD) and corresponding 95%CI. Heterogeneity assessed using the I2 statistic. Age, intensity of therapy, and treatment approach investigated as possible moderators.  Within-sessions changes (*non-generalised* outcomes) reported separately from those that occur outside sessions and in daily life (*generalised* outcomes).  A meta-analysis was conducted. Note that data from washout periods in the randomised cross-over trials were excluded (ie, for the group randomised to receive the intervention first, data from the second placebo/usual treatment phase was not analysed; rather, data was compared between those receiving intervention first and those receiving the comparator first). This essentially treated group crossover trials as RCTs. | **Included**: 10 studies included (n=165, *n* range 4–50; *age* range 2–9 years) with outcomes measured at 1 week to 7 months follow up. 4 randomised controlled trial studies, 5 randomised cross-over trials, and 1 pseudo-randomised cross-over trial (counter-balanced allocation).  **Key findings:**  Significant improvement in primary outcomes:   * social interaction *non-generalised*; k=1 study (n=10), SMD=1.06, CI(0.02–2.10) * social interaction *generalised*; k=3 studies (n=57), SMD=0.71, CI(0.18–1.35) * non-verbal communicative skills *non-generalised*; *k*=3 studies (n-57), *SMD*=0.57, CI(‑0.02–0.98); ns * non-verbal communicative skills *generalised*; *k*=3 studies (n=57), *SMD*=0.48, CI(-0.02–0.98); ns * verbal communicative skill *non-generalised*; *k*=2 studies (n=47), *SMD*=0.33, CI(0.16–0.50) * verbal communicative skill *generalised*; *k*=2 studies (n=47), *SMD*=0.30, CI(-0.28–0.89); ns * initiating behaviour *non-generalised*; *k*=3 studies (n=22), *SMD*=0.73, CI(0.36–1.11) * social-emotional reciprocity *non-generalised*; *k*=1 study (n=10), *SMD*=2.28, CI(0.73–3.83)   Significant improvement in secondary outcomes:   * social adaptation; *k*=4 studies (n=26), *SMD*=0.41, CI(0.21–0.69) * joy; k=1 study (n=10), SMD=0.96, CI(0.04–1.88) * quality of parent-child relationships; *k*=2 studies (n=33), *SMD*=0.82, CI(0.13–1.52).   There were no adverse events, and no studies reported on hyperacusis or cognitive ability. | **Author conclusions**: music therapy may help children on the autism spectrum to improve their skills in social interaction, verbal communication, initiating behaviour, and social-emotional reciprocity. Music therapy may contribute to enhancing non-verbal communication within therapy, increasing social adaptation skills, and promoting the quality of parent-child relationships.  Caution should be taken in interpreting the improvements in non-generalised social-emotional reciprocity and family relationships due to attrition in studies.  More research using larger samples and generalised outcome measures needed to corroborate findings and to examine whether the effects are enduring.  When applying the review to practice, music therapy requires specialised academic and clinical training.  **Reviewer’s comments**: Comprehensive search; identified, extracted and appraised by two researchers using checklists; appropriate data analysis.  **Source of funding**: none reported.  **Included relevant trials**:   * Arezina (2011) [47] * Brownell (2002) [52] * Buday (1995) [48] * Farmer (2003) [48] * Gattino (2011) [45] * Kim (2008) [45] * Lim (2010) [46] * Lim (2011) [50] * Thomas (2003) [51] * Thompson (2012) [51] |

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| **Brondino et al (2015) [42]** | | | | |
| **Country, study type, aim** | **Search strategy** | **Appraisal methods** | **Results** | **Conclusions** |
| **Country**: Italy  **Study type**: systematic review  **Evidence level**: I  **Study Quality** (SIGN checklist): + (acceptable quality)  **Aims:** to assess the effectiveness of complementary and alternative medicine (CAM) (including music therapy) for individuals on the autism spectrum. | **Databases**: MEDLINE, EMBASE, LILACS, Cochrane database of Systematic Reviews, CINAHL, Psychology and Behavioural Sciences Collection, Agricola, Food Science Source.  **Search**: Searched in October 2014. Transparent selection criteria. Range of keywords relating to autism, forms of CAM, music, and music therapy, in English. Searching of reference lists, contacted authors for unpublished data.  **Selection criteria**: randomised controlled trials and open label trials yielding primary results on the effects of CAM administration in the core symptoms of ASD. Music related therapies “which involved music in some way as an active part of the intervention”.  **Excluded:** case reports and case series. | **Method:** Two authors independently selected abstracts. No appraisal tools mentioned.  Included studies relating to music therapy reported separately in detailed tables of study characteristics, results and critique of methodological strengths and limitations, as well as in narrative synthesis. | **Included**: 15 studies included in the review.  6 randomised controlled studies, 4 randomised cross-over trials, and 1 pseudo-randomised cross-over trial, 2 case series studies, and 2 studies of music related therapy (not MT).  **Key findings:**  Noted conclusions of the Cochrane review [23] and other included papers.  Discussed common limitations of evidence base including small sample sizes, high drop-out rates, and definitional problems describing the standard methodology of MT that permits replicability. | **Author conclusions**: noted that MT is usually considered a behavioural intervention rather than a CAM treatment. Concluded that there is promising evidence that supports the use of music in children on the autism spectrum which seems to impact several symptom domains such as communication, social reciprocity, and emotion. The authors also concluded that the approach is extremely safe without side effects.  Suggested that music therapy could be added to conventional treatment as an augmentation to standard therapy, and could potentially reduce anxiety and enhance the positive response to behavioural and educational treatments.  Called for large randomised controlled trials with a better characterisation of patients into the efficacy of CAM.  **Reviewer’s comments**: Comprehensive search; identified by two researchers, no reference to use of methodological checklists. Detailed tables. Brief narrative description of selected studies.  **Source of funding**: none reported.  **Included relevant** **trials**   * RCT [53] * 10 trials included in the Cochrane review [23]   4 studies ineligible for the current review, including trials of non-music therapy interventions, and case series studies. |

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| **James et al (2015) [24]** | | | | |
| **Country, study type, aim** | **Search strategy** | **Appraisal methods** | **Results** | **Conclusions** |
| **Country**: New Zealand  **Study type**: systematic review  **Evidence level**: I  **Study Quality** (SIGN checklist): + (acceptable quality)  **Aims:** to assess the use of music therapy for individuals on the autism spectrum. | **Databases**: PsycINFO, MEDLINE, CINAHL, ERIC.  **Search**: Searched from 2004-September 2013, updating a Cochrane review from 2006 [37] in English language journals. Transparent selection criteria. Range of keywords relating to music therapy and autism. Searching of reference lists, and handsearching of key journals and Music Therapy World.  **Selection criteria**: experimental studies including randomised controlled trials (RCT), and single case experimental design (SCED) multiple baseline studies; and quasi-experimental studies (including ‘before and after’ group studies and SCED A-B studies) investigating music therapy on at least one individual on the autism spectrum. Music therapy defined as where music is used as a tool to address non-musical goals. Excluded: descriptive studies, assessment of skills, reviews, theoretical papers. | **Method:** Two authors independently selected studies (84% agreement), and two authors extracted data from included studies (the second checking the original papers for accuracy with agreement on 98% of data).  Outcomes variables coded as: decreasing undesirable behaviour, promoting social interaction/ communication, improving independent functioning, enhancing understanding of emotions, increasing verbal communication.  Study results rated as positive, negative or mixed based on the degree to which all or some dependent variables showed a statistically significant improvement in the treatment group cf control group (group studies), or showed improvement based on visual analysis of data (SCED studies).  The certainty of evidence was rated as insufficient, preponderant, or conclusive. *Conclusive* studies had an experimental design, inter-observer agreement of 80% or higher collected in at least 20% of sessions, operationally defined dependent variables, sufficient methodological detail to replicate the study. *Preponderant* studies had confounders relating to attrition, carry-over effects, or blinding. All quasi-experimental studies were rated as *insufficient*. | **Included**: 12 studies met criteria for inclusion in the review (n=147, *n* range 1–50; *age* range 3–38 years, 91% male).  10 were experimental (including 2 RCTs, 4 crossover trials, 4 single case experimental design studies) and 2 were quasi-experimental A–B designs.  Only one study, a SCED study, included follow-up, of 2 weeks, and no studies reported measures of generalisation.  **Key findings:**  7 (58%) of the studies reported positive outcomes (involving 147 participants), and 5 (42%) mixed outcomes.  7 (58%) of the studies provided “conclusive” evidence (mostly studies indicating positive outcomes), 3 provided “preponderant” evidence, and 2 “insufficient” evidence (the two quasi-experimental studies). | **Author conclusions**: Music therapy is a promising practice for individuals on the autism spectrum and for specific purposes. Additional research is warranted to further establish its generality; the mechanisms responsible for behaviour change; the effectiveness of a broader range of MT approaches in different age ranges and with differing severity of autism characteristics.  **Reviewer’s comments**: Broad search; identified and extracted by two researchers, appraisal adjusted for SCED and group studies. Certainty of evidence criteria aligned with SCED studies (eg, there was no reference to method of randomisation), detailed tables lacked clarity (eg, procedures reported study design and intervention together, no reporting of statistical tests for group studies).  **Source of funding**: none reported  **Included trials:**   * 5 trials included in the Cochrane review [23]   Other studies ineligible for the current review, including trials of non-music therapy interventions, case series studies, case studies and/or studies with small samples (N<5). |

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| **Su Maw & Haga (2018) [43]** | | | | |
| **Country, study type, aim** | **Search strategy** | **Appraisal methods** | **Results** | **Conclusions** |
| **Country**: Japan  **Study type**: systematic review and meta-analysis  **Evidence level**: I  **Study Quality** (SIGN checklist): + (acceptable quality)  **Aims:** to identify the most effective intervention for preschool aged children on the autism spectrum based on the cognitive, developmental, and behavioural approaches (including music therapy) and the factors that impact on the effectiveness of the intervention. | **Databases**: MEDLINE, PsycINFO, CINAHL, ERIC, Web of Science.  **Search**: Searched from 2001–2015 inclusive. Transparent selection criteria, using no language restrictions, restricted to human subjects. Range of keywords relating to autism, children, and intervention, treatment, or education. Handsearching of related articles and checking of reference lists of retrieved articles undertaken. Authors of included trials contacted for complete data for meta-analysis.  **Selection criteria**: randomised controlled trials comparing cognitive, developmental, and behavioural approaches with a control group in children aged 1–6 years on the autism spectrum. Interventions had to evaluate outcomes related to the core characteristics of ASD, such as communication and social interaction deficits, restrictive and repetitive behaviour, and language skill deficits as continuous outcomes.  Excluded: studies focusing on evaluating parental or caregiver outcomes, strictly medical problems, medication. | **Method:** First author selected studies, confirmed by second author (kappa=0.83). First author extracted data and appraised studies (using the Cochrane Risk of Bias tool), verified by second author.  Followed PRISMA reporting guidelines. A meta-analysis was conducted. Random-effects model was used to calculate the pooled estimate effect size, calculated as the pooled standardised mean difference (SMD; Cohen’s *d*) and corresponding 95%CI. Between study heterogeneity was also assessed using the I2 statistic. | **Included**: 14 RCTs (746 children, 72% boys). Of these, 3 studies evaluated music therapy interventions (all including children aged 3–5 years). These included:   * a developmental approach of in-home family-centred music therapy (FCMT) by Thompson et al (2013) [31] * behavioural approach of music-incorporated ABA by Lim et al (2011) [50] * an improvisational MT approach using the inherent communicability of children by Kim et al (2008) [49].   **Key findings:**  In broader review of 14 trials:   * pooled estimate of effect was 0.23, 95% CI[0.08–0.37]. No evidence of between-study heterogeneity. Observed that delivering interventions by trained therapists produced larger effect sizes. Not all high-intensity interventions could develop effective results.   For 3 trials relating to Music Therapy:   * SMDs for music therapies ranged from 0.40 to 0.60, 95% CI [0.22–1.85]. Two of the three trials had a high risk of bias. * Improvements found in parent-child relationship [31], speech & language production ) [50]; joint attention behaviours, non-verbal social communication [49]. * In-home FCMT [31] provided one the largest effect sizes (SM=-0.62, 95% CI [-0.22–1.54]) despite having the lowest intensity (at 30-minutes per week) of all interventions, and shorter duration (4 months). * music-incorporated ABA led to a higher effect size (SMD=0.60, 95% CI [0.09–1.85] than speech-incorporated ABA (SMD=0.49, 95% CI [0.28–2.09]) [50] * Improvisational MT also produced high ES (SMD=0.40, 95% CI [0–0.78] [49]. | **Author conclusions**: music therapy appears to be an effective tool for improving social interaction in pre-school aged children on the autism spectrum.  Concluded from a review of wider interventions that as a group, the music therapy studies were the most effective despite including one with the shortest duration and lowest intensity at 30‑minutes p/wk over 4 months [31]. Findings were consistent with the Cochrane review’s [23] appraised in the current review.  **Reviewer’s comments**: Comprehensive search; explicit search strategy; articles selected by 2 researchers with high inter-rater agreement, appraised and confirmed by two researchers (agreement not reported) using checklists based on Cochrane risk of Bias tool.  No formal sensitivity analyses of effects of approach, training of provider, duration or intensity (narratively applied).  Three music trials grouped together despite using different approaches, and methodological quality.  **Source of funding**: Japan Society for the Promotion of Science  **Included relevant trials**:   * Kim (2008) [49] * Lim (2011) [50] * Thompson (2013) [31] |

**Key:** ABA=Applied Behaviour Analysis; ASD=Autism Spectrum Disorder; CAM=complementary and Alternative medicine; CENTRAL=Cochrane Central Register of Controlled Trials; CINAHL=Cumulative Index to Nursing and Allied Health Literature; ERIC=Education Resources Information Centre; FCMT=family-centred music therapy; MEDLINE=Medical Literature Analysis and Retrieval System Online; MT=music therapy; ns=not significant; PRISMA=Preferred Reporting Items for Systematic Reviews and Meta-Analyses; RCT=randomised controlled trial; PsycINFO=Psychological Information Database; SCED=single case experimental design; SIGN=Scottish Intercollegiate Guidelines Network; SMD=standardised mean differences.

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1. Note that whilst Rett Syndrome was considered a PDD subtype under the DSM-IV, in DSM-5 Rett Syndrome is considered a separate diagnosis to ASD. [↑](#footnote-ref-1)
2. Auditory Integration Therapy (AIT) is a form of sound or listening therapy hypothesised to act physiologically and is therefore distinct from music therapy [23]. [↑](#footnote-ref-2)
3. See <https://www.health.govt.nz/system/files/documents/publications/achieving-equity-in-health-outcomes-summary-of-a-discovery-process-30jul2019.pdf> [↑](#footnote-ref-3)