Guideline Supplementary Paper

New Zealand Autism Spectrum Disorder Guideline’s supplementary paper on the effectiveness of physical activity interventions for young people on the autism spectrum



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

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#

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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 Updates to the guideline are required when the guideline’s recommendations are no longer valid in view of research that has emerged after the guideline’s literature searches were undertaken.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 guideline recommendations and good practice points. These topic updates which supplement the guideline, known as Supplementary Papers, have been produced annually since 2009 [2-8]. A second edition of the guideline was published in 2016 [9], 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 physical activity 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 [9] with evidence published from 2004 onwards relating to physical activity interventions. 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 considers the effectiveness of physical activity interventions which are aimed at improving social, cognitive and/or behavioural functioning for people on the autism spectrum.

Physical activity interventions are activities that require physical movement and effort that can be conducted in groups or individually across a wide range of domains. Different modalities include sports-based and aerobic activities, recreational activities, martial arts, relaxation training, creative therapies, exergaming, and horse riding. Animal assisted therapy including equine therapy was only included when there was a strong physical component (such as horse riding).

Interventions targeting physical fitness as a contributing determinant of general health outcomes have also been investigated. However studies evaluating physical fitness outcomes were excluded where they lacked at least one primary outcome for the current review (ie, social functioning, behavioural functioning, or cognitive functioning).

This document should be read in the context of the guideline’s 2nd edition [9] and the guideline’s Supplementary Papers [2-8, 10-12].

## Definitions

The current report relates to physical activity. Physical activity interventions involve repeated gross-motor movements requiring physical exertion. In the current review, they are defined as “planned, structured, repetitive, and purposeful” (p. 128) [13].

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 [14], four of the pervasive developmental disorder subcategories specified in the manual’s predecessor, the DSM-IV [15], 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 and intellectual function expressed by people across the autism spectrum requires that a wide range of services and approaches be employed to reflect the heterogeneity of the condition.

The term ASD is still used widely internationally and the guideline’s first edition [1] was prescient in recognising the movement toward considering autism as a spectrum condition. However, increasingly many people (particularly adults) in the autism community, whether formally or self-diagnosed, prefer to use identity-first language to refer to themselves as being autistic, autists or Aspies. This recognises autism as a central part of their identity, rather than being a person “with autism”. Some prefer to describe themselves as being on the autism spectrum, or as having autism or Asperger’s. Where used, the acronym ASD is sometimes defined as autism spectrum *difference* rather than *disorder.* In the UK, the term Autism Spectrum Condition (ASC) is sometimes used instead of ASD. In this supplementary report, the term “person on the autism spectrum” or “autistic person” is preferred, and refers 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) [14]. 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 [9]. These outputs (detailed in **Section 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 literature review updating the published evidence was conducted by INSIGHT Research and disseminated to the LGG as pre-reading for a one day, face-to-face meeting on 25 November 2019. At the meeting, the currency of the guideline was discussed in view of the updated evidence. Existing recommendations/good practice points were revised and new ones developed based on the considered evidence. These are described, accompanied by the LGG’s rationale and additional notes, in **Section 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.

# Summary: New recommendation and good practice points

Summary Table I: New recommendation relevant to physical activity and autism

|  |  |  |
| --- | --- | --- |
| **Reference** | **Revised recommendation** | **Grade** |
| 2.3.9a | Physical activities provide benefits across social, cognitive and behavioural domains in addition to general well-being 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.

Summary Table II: New good practice points relevant to physical activity and autism

|  |  |  |
| --- | --- | --- |
| **Reference** | **New Good Practice Points** | **Grade** |
| 2.3.9b | When supporting individuals participating in a physical activity or programme, their preferences should be respected and needs accommodated | ✓ |
| 2.3.9c | Further research that targets meaningful outcomes for the autistic community is essential | ✓ |
| *also* 4.1.6 | Further research that targets meaningful outcomes for the autistic community is essential | ✓ |

**Note**: Where a consensus-based recommendation is based on the experience of members of the Living Guideline Group, it is referred to as a good practice point.

# 1 Introduction

## 1.1 Background

### Physical activity interventions to improve health

The health benefits of physical exercise for virtually everyone are irrefutable and well established across many domains [16]. Regular physical activity is an effective primary and secondary preventative strategy for more than 25 chronic medical conditions with common risk reductions of 20%–30% [17]. These include the prevention and treatment of coronary heart disease, hypertension, stroke, type 2 diabetes, obesity, breast and colon cancers, falls, depression, anxiety, dementia, osteoporosis, fractures; and obesity [18]. These improvements in general and health-related quality of life, better functional capacity and better mood states are evident across a broad range of participants, representing males and females, children and adults, and diverse ethnic populations [19].

In efforts to encourage physical activity from a young age, New Zealand developed amended guidelines for sustainable physical activity in school communities, mandated for all state and state-integrated schools in 2006 [20]. Physical activity was added as a priority in the curriculum, and specifically as an area in which students are to develop high levels of competence, alongside literacy, numeracy, science and technology (*clause 5, National Education Guidelines)*. Additionally, the National Administration Guidelines, added a clause concerning the need to develop teaching and learning programmes, “giving priority to regular quality physical activity that develops movement skills for all students, especially in years 1–6” (*clause 1, National Administration Guidelines).* Despite these efforts, New Zealand children are amongst the most inactive in the world. New research funded by the World Health Organisation found that New Zealand ranked 138th out of 146 countries in the physical activity of children aged
11–17 years. In this age range, the prevalence of insufficient physical activity (less than one hour of moderate exercise a day) was over 80% for boys and over 90% for girls in New Zealand [21].

### Physical activity of people on the autism spectrum

People on the autism spectrum appear to be particularly at risk for physical inactivity. Some recent indicators suggest that individuals on the autism spectrum tend to be less active, and are more likely to be overweight and obese, than their typically-developing counterparts. Analyses from the 2016–2017 National Survey of Children’s Health (United States) [22] compared weekly physical activity, sedentary behaviour, and body mass index classification among adolescents diagnosed with and without autism spectrum disorder. After adjustment for covariates, adolescents with ASD diagnoses were found to engage in less physical activity, and were more likely to be overweight and obese, than their peers without ASD. As parent-reported autism severity increased, the adjusted odds of being overweight and obese increased and physical activity participation decreased. A recent systematic review and meta-analysis reported that children with ASD had a 41.1% greater risk (p<0.018) for the development of obesity [23].

A literature review of the physical health of people with ASD [24] concluded that people with ASD have a high rate of comorbid medical conditions and are at increased risk for chronic disease compared with their peers. Whilst many risk factors for poor physical health are likely the same, the needs and challenges of the autism condition and its comorbidities may make autistic people more susceptible and vulnerable to those same risk factors [25].

Commonly co-occurring factors that can impact on physical health include psychopharmacological treatment, disordered sleep, food selectivity, gastrointestinal disturbances, social anxiety, and metabolic disturbances [5, 26, 27]. Many individuals on the autism spectrum also have motor impairments (including hypotonia, apraxia, toe-walking, delayed gross and fine motor milestones, and reduced ankle mobility) to a degree that they have been considered by some to be a core characteristic of autism [28-30]. Such impairments can be a significant barrier to physical activity participation among people with ASD [31]. Alongside reduced motor proficiency (including muscular strength and endurance) [32], balance and gait challenges prevalent in people on the autism spectrum can also increase their risk of falls [26].

Physical exercise interventions often occur in social environments, and surveys have suggested that intrapersonal factors are most commonly identified as leading to barriers, as well as facilitators, of physical activity in the ASD population [33]. Supporting these findings, qualitative research with parents and children themselves has identified bullying, social comparisons, and not feeling welcome to participate, as reasons for children diagnosed with ASD to feel excluded or to self-exclude from physical education activities. Other challenges include interventions not being adapted for their needs, instructors not having awareness of autistic challenges, lack of adult supervision, sensory concerns, and fear of injury [34, 35]. With these barriers and the challenges already routinely faced by autistic people, exercise can seem to be less of a priority [36].

Together, the challenges and comorbidities of autism, as well as social barriers around the autistic person, can place people on the autism spectrum at risk for being less physically active as well as being at risk for poor health outcomes that could be improved by physical activity.

### Physical activity interventions in ASD

Investigations of interventions aiming to increase physical activity have become a prominent area of research in autism, with the number of publications growing dramatically over the last decade. Evidence for benefit has also accumulated to the extent that physical exercise was identified as an evidence-based practice by the US’s National Professional Development Center on Autism Spectrum Disorder (NPDC) in 2015 [37, 38].

A wide range of activities have been investigated therapeutically, including aerobic activities (eg, running, swimming, exercises), recreational activities (eg, walking, hiking), sports (eg, basketball, tennis), specific motor skill training, martial arts, yoga, horse riding, and dance movement. Aerobic activities including jogging and aquatics have been the most commonly investigated intervention [37].

One of the strengths of physical activity interventions is their accessibility and affordability, being able to be introduced into one’s home or community without the need for expensive equipment or specialists [39]. Group-based activities provide increased opportunities for social interaction, teamwork and interpersonal communication [40].

One activity gaining research attention is exergaming, a form of digital gaming which requires physical movement to play (eg, the Nintendo Wii™ which uses motion capture technology to track actual movements and represent them on the screen, allowing virtual sport or action gaming). Such activities can be undertaken in a private environment, and offer flexible multi-play options which allow opportunities for social interaction with peers [41, 42].

Notably, physical activity interventions for individuals on the autism spectrum target domains beyond physical health, fitness, balance, flexibility, and cardiovascular risk factors. Other areas potentially impacted include social skills, communication skills, language, cognitive functioning, attention, academic engagement, specific motor skills, self-injurious behaviours, aggression, sensory skills, and repetitive patterns of behaviour [36, 43-45].

Mechanisms for how physical movement and exercise may affect such domains are not yet well understood.

An association between motor deficits and social deficits in autism has been proposed such that interventions that improve motor skills may also improve social functioning [46].

With respect to repetitive behaviours, it has been suggested that physical exercise or “movement breaks” can have a strong sensory, perceptual-motor component and produce a pleasant internal consequence for the individual and may assist them in self-regulation. Due to a similarity in biofeedback responses, targeted physical exercises may potentially replace undesired stereotypical behaviours. An alternative possibility is that physical activity may improve the synthesis of dopamine and serotonin and potentially reduce stereotypical behaviour through that pathway [47].

Researchers have suggested that there may be indirect pathways between physical exercise and cognitive gains, including elevated self-efficacy, mood, and other psychological functioning [48]. It has also been hypothesised that, in turn, executive functioning improvements after exercise may lead to behavioural improvements [39].

It is not fully known how much an improved outcome relies on the physical movement common to all interventions of this class (though varying in intensity) and how much is due to accompanying features. For example, social elements incidental to a physical activity may themselves be key components, such as the social contact provided by an activity through interactions with peers, family members and instructors/coaches, particularly when delivered to groups. Informal peer contact and group belonging are also common features of effective recreational interventions included in social skills training [7].

In addition to exercise, a horse riding intervention includes contact with animals. This feature is common to other therapies which do not include core exercise components, such as animal assisted therapy (zootherapy), equine therapy, hippotherapy, service dogs, dolphin encounters, elephant therapy, and pet therapy.[[2]](#footnote-2) Animal therapy evaluations have reported improvements in social interaction, language and communication, motor skills, positive emotions, and stress for individuals on the autism spectrum [49].

## 1.2 Physical exercise in the existing autism guideline

Physical exercise is largely absent from the current New Zealand Autism Spectrum Disorder Guideline. A brief section on “Other health issues’ (page 73) notes the lack of reliable research on the topic, captured in Recommendation 2.3.9 (emphasis added): “Research should be undertaken to identify the needs of people with ASD with regard to constipation, allergies, medication reactions, menstruation *and exercise*” (see **Table 1.1**).

Table 1.1: Recommendations relevant to physical activity in the guideline

|  |  |  |
| --- | --- | --- |
| **Reference** | **Recommendation** | **Grade** |
| 2.3.9 | Research should be undertaken to identify the needs of people with ASD with regard to constipation, allergies, medication reactions, menstruation and exercise. | C |

**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.

Exercise is also referred to briefly in Chapter 3 of the guideline under positive behaviour supports with reference to its health benefits and a related reduction of behavioural problems (page 118).

The current update will permit revision (including removal, rewording or replacement) of the existing related recommendation in the guideline, as well as the development of new recommendations and Good Practice Points (GPP).

## 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 for effective physical activity interventions for individuals on the autism spectrum
* consider this evidence as it supplements the guideline [9] in order to inform the LGG’s revision of existing recommendations/good practice points and/or the development of new ones.

# 2 Systematic review of effectiveness of physical activity for people on the autism spectrum

This chapter describes the findings of a systematic review relating to physical activity interventions for individuals 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 question is:

* What is the effectiveness of physical exercise interventions for individuals on the autism spectrum?

### Identification and selection of studies for inclusion

Search strategies were limited to English language publications from 1 January 2004, to ensure capture of articles published since the search was conducted for the original guideline [1]. Studies already appraised for a relevant research question in the guideline’s first or second edition [1, 9] were excluded from the current review regardless of date of publication.

Seven bibliographic, health technology assessment, and guideline databases were accessed in the systematic search (see **Appendix 1** for details). The search was conducted on 1 August 2019 and updated on 6 September 2019. Following removal of duplicates, 1442 potentially relevant articles were identified.

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

In order to consider the “best evidence” within the hierarchy of evidence [50], criteria relating to study designs was then applied. This led to a restriction to level I evidence (systematic reviews and meta analyses) and level II evidence (additional randomised controlled trials; that is, where not already included in appraised reviews). Studies were also limited to those published since 1 January 2014 as secondary studies were understood to capture previously published primary research. This reduced the number of potentially relevant articles identified to 1017.

Selection criteria for included and excluded studies are summarised in **Table 2.1**.

Table 2.1: Inclusion and exclusion criteria for selection of studies

|  |  |
| --- | --- |
| **Characteristic** | **Inclusion criteria** |
| Publication type | Studies published in the English language in peer reviewed journals. |
| Participant characteristics | Participants aged two years or older diagnosed with Autism Spectrum Disorder (ASD).For samples of mixed disabilities, studies were included if at least 51% of participants were diagnosed with ASD, and/or where results were reported separately for the eligible sub-group. |
| Scope | The key focus was investigating the effectiveness of eligible physical activity interventions for people diagnosed with ASD. |
| Intervention | Intervention was primarily a physical activity involving some physical exertion that is:* planned
* structured
* repetitive
* purposeful.

Interventions may be given individually or in groups. |
| Comparator | For experimental studies, the comparison/control group was a non-physical control activity, wait-list control, or no intervention. |
| Outcomes | Included measures from one or more of the following primary outcome domains:* social functioning and communication
* behavioural functioning
* cognitive functioning.

Secondary outcomes reported but not sufficient on their own for inclusion:* skill development
* fitness.
 |
| Study design | Identification of “best evidence” led to further refinement of criteria to include:* Level 1 [50] evidence: systematic reviews and/or meta-analyses that have a clear and relevant review question, use at least one electronic bibliographic database, and include at least one study meeting aforementioned inclusion criteria for the current review
* Level II [50] evidence: randomised controlled trials where they have met other selection criteria for the current review, and not already reviewed in included Level I research.
 |

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

|  |  |
| --- | --- |
| **Characteristic** | **Inclusion criteria** |
| Publication date | Again to identify best evidence publications were further limited to those published between 1 January 2014 and 6 September 2019 inclusive. |
| Publication type | The following were excluded* correspondence, dissertations, editorials, commentaries, narrative reviews, expert opinion, book chapters, conference abstracts, poster presentations, unpublished data, and animal studies
* interventions where exercise was used as a punishment (“contingent exercise”)
* interventions providing encouragement or suggestions for how to conduct exercise, in an educational capacity only
* studies evaluating role playing, imitation, or sensory integration interventions
* single case reports.
 |
| Language  | Non-English language articles |
| Scope | Studies which were not deemed relevant to the research question or nature of the review, including those describing service provision, or development of an intervention, outcome measure or index without explicit evaluation of an eligible intervention |

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 for inclusion in the review. 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,

### Participants

The study population were people aged at least 2 years old and diagnosed with Autism Spectrum Disorder (ASD). Whilst the guideline [9] defines autism spectrum disorder as classified by or consistent with DSM-IV [15] or DSM-5 [14] diagnostic criteria, studies were not limited by how ASD status was 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 diagnosed with ASD.

### 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 physical activity interventions for people diagnosed with ASD.

### Intervention

Included studies evaluated interventions that focused on (as the core of their intervention) undertaking a physical activity that involves some *physical exertion* and is *planned, structured, repetitive and purposeful* (p. 128) [13]. This definition, also used in key systematic reviews on this topic [31, 39], is inclusive of a range of exercise interventions that have been employed in this population in both a recreational and therapeutic setting [39]. Interventions may be given individually or be group-based.

Areas of activity included the following:

* sports-based and aerobic activities such running, jogging, swimming, weight training, cycling, rebound therapy (trampolining), gym training
* recreational activities such as walking, hiking, etc
* martial arts-based activities such as tai chi
* relaxation training with significant physical components such as yoga
* creative therapies such as dance movement therapy
* gaming activities involving physical movement (eg, exergaming)
* other interventions in which physical activity is a significant component such as equine therapy (horse-back riding).

### Comparator

Where applicable for experimental study designs, the comparison/control group could be a non-physical control activity (eg, usual activity such as educational programme), waitlist control, or no intervention.

### Settings

The study took place in any setting, including a physical education, physical activity, or sport setting, defined as follows [28]:

* a physical education setting: traditional team or individual, organised, sport-specific gameplay settings that occur outside of educational settings
* physical activity setting: activity taking place outside of an educational setting
* sport-specific based setting: recreational activities such as walking, hiking, playing at a playground, or lab-based activity.

### Outcomes

Included studies needed to include one or more of the following primary outcome domains:

* social functioning and communication (eg, social responsiveness, social motivation, social interaction, communication skills, communication, language ability, emotional recognition)
* behavioural functioning (eg, restricted, repetitive behaviour, stereotypic behaviour, adaptive skills, aggression, self-injury, other problem behaviours)
* cognitive functioning (eg, executive functioning, attention, academic performance, time on-task)
* additional secondary outcomes also reported but which in the absence of a primary outcome did not meet criterion for inclusion:
* skill development (eg, sport, locomotor, manipulative, balance, sensory)
* fitness (eg, muscular strength/endurance, cardiovascular endurance, body composition, skill-related fitness, health-related fitness).

### Study designs

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

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 [50] (see **Appendix A1.3** for further details of levels of evidence). The goal was to identify “best evidence” representing the higher levels of the evidence hierarchy and only in their absence, include lower order evidence.

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

* Level I evidence; that is, 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 (solely or separately as a synthesised sub-group), and included at least one study meeting other selection criteria (ie, apart from study design) for the current review outlined above.
* Level II evidence; that is, randomised controlled trials (RCT) where meeting other selection criteria for the current review, and where not reviewed in included Level I studies.

### Publication date

After refinement of eligibility criteria to identify best evidence (secondary evidence and additional RCT), studies were limited to those published between 1 January 2014 and 6 August 2019 inclusive.

### Exclusions

Research papers were **excluded** if they:

* were not published in the English language
* were non-systematic reviews, correspondence, editorials, expert opinion articles, commentaries, news reports, trade magazines, case reports, book chapters, articles published only in abstract form, conference proceedings, poster presentations, dissertations, unpublished work, animal studies, single case reports, and non‑empirical research
* described service provision (without explicit evaluation of the intervention)
* described development of an intervention, outcome measure or index (without explicit evaluation of the intervention)
* interventions where exercise was used as a punishment (often called “contingent exercise”)
* interventions providing encouragement or suggestions for how to conduct exercise, in an educational capacity only.

### 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 [51] 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 since 1 January 2014, 1017 unique abstracts were identified. After applying inclusion and exclusion criteria, 14 studies were eligible for inclusion in the review: nine secondary studies (systematic reviews and meta analyses), and five randomised controlled trials.

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 (as defined in **Appendix 1**, **Table A1.1**), SIGN study quality rating, 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

Nine secondary studies on the review topic were identified which included systematic reviews using selection criteria overlapping with the current review.

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

#### Scope

Of the nine secondary studies identified relevant to this topic published since 2014, one was a clinical practice guideline [52], four were stand-alone systematic reviews [39, 41, 53, 54], and four were reviews which also included meta analyses [31, 40, 48, 55].

With respect to the scope of interventions included, the terms physical activity and physical exercise appear to have been used interchangeably in the literature and within reviews.

Studies ranged from very broad reviews of interventions in ASD generally [52], to more focused reviews on subsets of intervention types.

In terms of physical activities, one review was restricted to exergaming [41]; another related to group-based delivery of interventions [40]; and others restricted outcomes evaluated to psycho-social functioning [54] social and communication outcomes [40], or cognitive functioning [48].

With respect to population, one review considered people diagnosed with ASD, ADHD, or both [48], and reported impact on cognitive outcomes separately for each diagnostic group. Two-thirds of the appraised secondary reviews had selection criteria that limited studies to those involving children and young people diagnosed with ASD; one was restricted to individuals in their primary school years [40], but more commonly studies included individuals aged up to their early 20’s. Three reviews [41, 48, 52] did not have an age restriction for included studies, though in actuality the identified studies relevant to the current review related to people up to age of 25 years. And so regardless of scope, the included systematic reviews published since 2014 relate to children and young adults on the autism spectrum.

#### Country

Reviewers were based across developed countries comparable to New Zealand, including three reviews from the United States [31, 41, 53], two from Australia [40, 48], two from Canada [39, 54], one from Hong Kong [55], and one guideline from Scotland [52].

#### Quality

Review quality varied widely, with three reviews rated (using the SIGN checklist) as being of low quality (-) [53-55], four of acceptable quality (+) [31, 40, 41, 48], and two of high quality (++) [39, 52].

Key review limitations included using narrow or restricted search strategies which reduce confidence that all relevant studies had been identified; lack of reporting of inter-rater reliability; lack of appropriate appraisal checklists to assess methodological quality; lack of methodological critique; inconsistent or inaccurate descriptions of study design; inclusion of poorly controlled studies; and for meta analyses, a lack of sensitivity analyses. Reviews also were impacted by the limitations of the evidence base, particularly when results were synthesised across disparate studies in meta-analyses. Included studies were highly divergent with respect to type of intervention, duration (per session) and dose (ie, how many weeks the exercise programme was applied). Limitations of research in this area, including in the current review, are discussed in further detail in **Section 2.4.**

#### Narrative summary of secondary studies

Below, studies are narratively synthesised and presented in order (broadly) from least to most relevant to the current review. This relates to breadth of scope operationalised by how many randomised controlled trials were identified relevant to the current topic (lowest to highest).

Physical exercise was only one small topic considered by the high quality Scottish clinical practice guideline by the **Scottish Intercollegiate Guidelines Network (2016)** [52] on assessment, diagnosis and interventions for individuals on the autism spectrum. The review identified a meta-analysis by Sowa et al (2012) [45] that pre‑dated the inclusion period for the current review, but included an RCT evaluating horse riding [56]. The guideline concluded that physical exercise may be beneficial for improving motor skills and social interaction for individuals with ASD.

Physical activity was also a sub-component of a larger, low quality scoping review by **Lee et al (2016)** [54] which considered movement-based interventions (defined as full body movement through space) for children diagnosed with ASD. Only two studies within this broader review related to physical activity, including one RCT on the Japanese martial art, Kata [57]. Reviewers undertook a thematic analysis of what was described as emerging evidence for the benefits of movement-based interventions (including physical exercise) for children with ASD.

Table 2.2: Characteristics of secondary studies

|  |  |  |  |  |  |  |
| --- | --- | --- | --- | --- | --- | --- |
| **Author/s (year)** | **Country** | **Evidence level, Quality** | **Population** | **Intervention, Outcomes** | **Search strategy and analysis** | **Results/Recommendations** |
| Sam et al (2015) [55] | Hong Kong | I- (low quality) | Children with ASD | Exercise interventionsImpact on exercise mastery, physical fitness, and social competence | Narrow search strategy with few databasesOne researcher using appraisal checklists and briefly described resultsMeta-analysis performed | 8 studies, including 3 RCTs [56, 58, 59]Evidence that exercise interventions were moderately effective for improving exercise mastery and social competenceNon-significant effect on physical fitness |
| Bremer et al (2016) [39] | Canada | I++ (high quality) | Children with ASD (0–16 years) | Exercise interventionsImpact on overall behaviour | Moderately broad search strategyTwo researchers independently applied eligibility criteria with high agreement, and appraised studies using SIGN checklists | 13 studies, including 2 RCTs [57, 58]Moderate to large improvement on stereotypic behaviour, social-emotional improvement, cognition and attention, particularly for martial arts (from 2 RCTs) and horse ridingLess evidence of effectiveness for swimming, yoga, dance interventions |
| Lee et al (2016) [54] | Canada | I- (low quality) | Children with ASD | Within broader review of movement-based interventions:Physical activityImpact on psychosocial outcomes | Moderately broad search strategyOne researcher appraised studies using appraisal tool but no critique providedThematic analysis provided | 2 studies relevant to current review, including 1 RCT [57]Emerging evidence on the use of movement-based interventions for improving psychosocial outcomes for children with ASD |
| Scottish Intercollegiate Guidelines Network (2016) [52] | Scotland | I++ (high quality) | Individuals with ASD | Within broader guideline on assessment, diagnosis and interventions in ASD:Physical activitiesAny outcome | Moderately broad search strategyTwo researchers independently applied eligibility criteria and appraised studies using SIGN checklists | One meta-analysis of small trials [45], including one RCT [56]Exercise may be beneficial for improving motor skills and social interaction for individuals with ASD |
| Tan et al (2016) [48] | Australia | I+ (acceptable quality) | Individuals with ASD | Within broader review including ADHD:Physical exercise interventionsImpact on cognitive functioning | Moderately broad search strategyAuthors independently applied eligibility criteriaMeta-analysis performed | 6 studies relevant to current review, including 1 RCT [60]Evidence for moderate effect on improving duration of being on-task, evident for majority of participants. No moderators of effect identified |

*Continued next page*

**Table 2.2: Characteristics of secondary studies** *(continued)*

|  |  |  |  |  |  |  |
| --- | --- | --- | --- | --- | --- | --- |
| **Author/s (year)** | **Country** | **Evidence level, Quality** | **Population** | **Intervention, Outcomes** | **Search strategy and analysis** | **Results/Recommendations** |
| Dillon et al (2017) [53] | US | I-(low quality) | Children and young people with ASD (1-21 years) | Exercise interventionsAny outcome | Search strategy limited by use of single search term (“exercise”)Authors independently applied eligibility criteria with high agreement. Study designs inconsistently reported | 23 studies, including 4 RCTs [57-59, 61]Concluded from one well-conducted quasi-experimental study that exercise is an evidence-based practice for school aged children with ASD |
| Healy et al (2018) [31] | US | I+ (acceptable quality) | Children and young people with ASD (2-22 years) | Physical activity interventionsAny outcome | Broad search strategyAuthors independently applied eligibility criteria but no checklists reportedMeta-analysis performed | 29 studies, including 7 RCTs [57-59, 62-65].Found a moderately positive effect, particularly for social functioning, and locomotor skills, and improvement of skill-related fitness, and muscular strength/endurance.Non-significant effects for body composition and cardiovascular endurance |
| Fang et al (2019) [41] | US | I+ (acceptable quality) | Individuals with ASD  | Exergaming interventionsImpact on cognitive performance, social-emotional behaviours, or physical ability | Narrow search strategyAuthors independently applied eligibility criteria and appraised studies using quality checklistsThematic coding used to categorise outcomes | 10 studies, including 1 RCT [64]Exergaming is effective in improving physical fitness, executive function, and self-perceived motor skills performanceNon-significant effects for improving emotional control and motor skill development |
| Howells et al (2019) [40] | Australia | I++(high quality) | Children with ASD (5-13 years) | Group-based physical interventionsImpact on social and communicative outcomes | Broad search strategyTwo researchers independently applied eligibility criteria and used quality checklistsMeta-analysis performed | 11 studies, 7 in meta-analysis, including 4 RCTs [57, 58, 62, 63]Significant small to medium improvement in overall social functioning, but non-significant effect for communication |

**Key**: ASD=Autism Spectrum Disorder; SIGN=Scottish Intercollegiate Guidelines Network; US=United States of America

A recently published systematic review of acceptable quality from the US by **Fang et al (2019)** [41] considered a subset of physical activity, exergaming; a form of digital gaming which requires physical movement to play using motion capture technology. Gaming was said to provide opportunities for moderate to vigorous physical activity. The review included 10 primary studies, one of which was an RCT which evaluated use of a game accessed on a Nintendo Wii™ [64]. Thematic analysis found significant improvements in physical fitness, executive function, and self-perceived motor skills performance, but no significant effects on emotional control and actual motor skill development.

An Australian systematic review of acceptable quality by **Tan et al (2016)** [48] considered the impact of physical activity on cognitive outcomes for people diagnosed with ASD and/or ADHD. Six studies reported on people on the autism spectrum, including one RCT evaluating tri-cycling [60]. A meta-analysis indicated a moderate effect on improving duration of being on-task, evident for the majority of participants. Multiple regression analyses across all 22 studies regardless of participant diagnosis investigated the possibility of moderators of effect (including gender, age, diagnostic group, control group, exercise type, exercise duration) but none were identified.

A high quality Canadian systematic review by **Bremer et al (2016)** [39] considered the behavioural impact of exercise interventions on children on the autism spectrum up to the age of 16 years. Of 13 studies identified, two were RCTs, both evaluating martial arts interventions [57, 58]. The reviewers reported moderate to large improvement on stereotypic behaviour, social-emotional skills, cognition and attention, noting the results were strongest for martial arts and horse riding interventions. They reported weaker evidence of effectiveness for swimming, yoga, and dance interventions on these outcomes.

A low quality review of the effectiveness of exercise interventions by Hong Kong based researchers **Sam et al (2015)** [55] included a meta-analysis of eight studies, including three RCTs evaluating horse riding, martial arts (kata), and mind-body exercise, respectively [56, 58, 59]. The reviewers concluded there was evidence that exercise interventions were moderately effective for improving exercise mastery and social competence, but not for physical fitness.

Another low quality review by **Dillon et al (2017)** [53] in the US identified 23 studies, including four RCTs, two of which evaluated martial arts [57, 58], one mind-body exercise [59] and one considered table tennis training [61]. This review used very limited search terms and inconsistent terminology for describing study designs, but concluded on the basis of one well conducted study (which was not an RCT) that “exercise is an evidence-based practice for school aged children with ASD”.

A recently published, high quality Australian systematic review by **Howells et al (2019)** [40] considered the impact of group based physical interventions on social and communicative outcomes for primary school aged children on the autism spectrum. Eleven studies were identified, and seven were included in a meta-analysis. Of these, four were RCTs, two of which evaluated martial arts [57, 58], and two evaluated horse riding [62, 63]. The review of studies of children aged between 5 and 13 years found a significant small to medium improvement in overall social functioning, but no improvement in communication outcomes.

Finally, is the systematic review and meta-analysis (of acceptable quality) from US based researchers **Healy et al (2018)** [31] which had a broad scope closest to the current review. Of 29 studies identified, eight were RCTs, including two studies of martial arts [57, 58], two of horse riding [62, 63], one of exergaming [64], one of mind‑body exercise [59], and one of athletics [65]. The meta-analysis found a moderately positive effect (g=0.62), particularly for the development of manipulative skills, locomotor skills, skill-related fitness, social functioning, and muscular strength/ endurance. Effects were not significant for body composition and cardiovascular endurance outcomes. Attempts to determine moderators for effects were restricted by low sample sizes and high heterogeneity.

The included systematic reviews had overlapping scope, search strategies, and selection criteria, and therefore also overlapping included primary studies. It is important that review findings are therefore not summated or given additional weight where the same studies are reported. To emphasise this, a summary of the 10 unique randomised controlled trials identified by the systematic reviews published since 2014 are presented in **Table 2.3**. Information entered into this table has been obtained from the original primary studies to ensure consistency of data extraction and reporting. Note that these studies have not been formally appraised in the current review, and therefore their potential biases and limitations are not known. Moreover, as RCTs these studies represent a subset of the primary studies considered by the appraised systematic reviews.

Table 2.3: Characteristics of relevant randomised controlled trials identified in appraised secondary studies

|  |  |  |  |  |  |  |  |
| --- | --- | --- | --- | --- | --- | --- | --- |
| **Author/s (year)** | **Country** | **Sample size, gender** | **Mean age (range)** | **Intervention** | **Duration/dose of activity** | **Significant improvement in outcomes\*** | **Non-significant change in outcomes** |
| Bass et al (2009) [56] | US | N=3485% male | M=7 years(5–10) | Horse riding | 1-hour x 1/wk x 12 weeks=12 hours | * sensory profile
* social responsiveness
 |  |
| Bahrami et al (2012) [58] | Iran | N=3087% male | M=9 years(5–16) | Kata programme | 90 minutes x 4/wk x 12 weeks=72 hours | * reduced stereotypical behaviour
 |   |
| Borgi et al (2016) [62] | Italy | N=28100% male | M=9 years(6–12) | Horse riding | 60-minute x 1/wk x 25 weeks=25 hours | * social functioning
* motor skills
* executive functioning
 | * communication
* daily living skills
* executive functioning (planning, problem solving)
 |
| Chan et al (2013) [59] | China | N=4090% male | M=11 years(6–17) | Mind body exercises (*Nei Yang Gong*) | 1-hour x 2/wk x 4 weeks=8 hours | * self-control
* sociability
* sensory/cognitive awareness
* health/physical behaviour
 | * speech/language, communication
 |
| Dickinson & Place (2014) [64] | UK | N=10079% male | M=NR(5–15) | Exergaming | 15 minutes x 3/wk x 30 weeks=22.5 hours | * physical fitness
* reduced BMI
 | * flexibility
 |
| Favazza et al (2013) [65] | US | N=233X=80% male | M=4 years(3–5) | Athletics (motor skill programme) | 30-minute x 3/wk x 8 weeks=12 hours | * motor skill development
* school readiness
* social/play skills
 |  |
| Gabriels et al (2015) [63] | US | N=11687% male | M=10 years(6–16) | Horse riding | 45 minutes x1/wk x 10 weeks=7.5 hours | * reduced irritability
* reduced hyperactivity
* social responsiveness
* language (word use)
 |  |
| Movahedi et al (2013) [57] | Iran | N=3087% male | M=13 years(5–16) | Kata programme | 30– (building up to) 90-minute x 4/wk x 14 weeks=56 hours approximately | * social interaction
 |  |

*Continued next page*

**Table 2.3: Characteristics of relevant randomised control trials identified in appraised secondary studies** *(continued)*

|  |  |  |  |  |  |  |  |
| --- | --- | --- | --- | --- | --- | --- | --- |
| **Author/s (year)** | **Country** | **Sample size, gender** | **Mean age (range)** | **Intervention** | **Duration/dose of activity** | **Significant improvement in outcomes\*** | **Non-significant change in outcomes** |
| Pan et al (2017) [61] | Taiwan | N=22100% male | M=9 years(6–12) | Table tennis skills | 70-minute x 2/wk x 12 weeks=28 hours | * motor skills (coordination, strength, agility)
* executive functioning
 |  |
| Tan (2013) [60] | Singapore | N=12100% male | M=5 years(2–6) | Tri-cycling | 15 minutes x 8 sessions=2 hours | * cognition (attention span)
 | * health-related quality of life
 |

**Key**: approx.=approximately; ASD=Autism Spectrum Disorder; BMI=body mass index; M=mean; N=total sample size (participants in intervention and control groups); NR=not reported; UK=United Kingdom; US=United States of America; /wk=per week; \* significant improvements in the group receiving the intervention compared with those in the control group.

### Primary studies

Five primary studies were appraised that reported on randomised controlled trials evaluating physical activity interventions for individuals diagnosed with ASD. These were published since (and therefore excluded from) included recent systematic reviews identified that were published since 2014. Study characteristics for the primary studies are presented in **Table 2.4.**

#### Sample characteristics

There were 304 participants across the five studies. Sample sizes for the studies ranged from 40 to 100 children/adolescents.

All of the five appraised primary studies related to school aged children recruited from mainstream or special education schools, with ages ranging from 5–16 years, and mean age (in the four studies where reported) ranging from 6–10 years. There were a majority of males, ranging from 70% to 100%, broadly reflecting the 4:1 male to female ratio that is commonly seen in studies of ASD prevalence [66].

The primary studies were conducted in a diverse range of countries, including the United Kingdom, Iran, Brazil, China, and Hong Kong.

#### Quality

Whilst inclusion criteria restricted studies to randomised controlled trials, the methodological quality varied, with four studies rated using the SIGN checklist as being of low quality, and one study from researchers in Hong Kong [67] of acceptable quality.

#### Interventions

Interventions were all delivered in small groups. One study [42] evaluated an exergaming intervention (the Nintendo Wii™), one considered basketball skills training [67], and three studies [27, 47, 68] investigated the effectiveness of general exercise programmes involving a range of strength, balance and ball skill activities. Intervention programmes involved sessions of between 15 and 60 minutes, 2 to 3 times per week, for 12 to 48 weeks, with total duration of between 18 and 48 hours.

#### Narrative summary of primary studies

A UK based study [42] investigated the impact of a brief, repeated exergaming intervention (using the Nintendo Wii) on the social functioning of 100 children (aged
2–16 years; 79% male) on the autism spectrum recruited from three schools. Students were randomly assigned to the treatment group or control group (receiving regular school physical education classes), controlling for age and gender. The intervention involved a range of sport games (aquatics, athletics, fencing, table tennis) involving 2 to 4 players and provided in 15 minute sessions three times per week over a school year (summing to approximately 30 hours). Children did not differ at baseline in demographic characteristics. A Social Behaviour at School questionnaire was administered (unblinded by condition) to teachers to assess social functioning pre- and post-intervention. ANCOVA analyses indicated that children receiving the exergaming intervention improved significantly in social functioning over time compared with the control group (p<0.001).

Investigating the effectiveness of multi-faceted exercise programmes was an Iranian RCT [47], whichrecruited 50 boys (Mean age=7.6 years; range=6–9 years) from schools specializing in educating children on the autism spectrum. A high proportion of the 250 children approached were ineligible due to various health conditions, or declined to participate. Following randomisation, half the sample received a 60 minute thrice-weekly programme of exercises including jogging, jumping, hopping, balance, and ball kicking, throwing, and catching for 12 weeks (summing to 36 hours). The treatment group, which was similar to the control group (receiving no intervention) at baseline, exhibited decreased stereotypy (judged on the GARS-2 by parents, teachers and observers unblinded to condition) between pre- and post-test assessments (p<0.01), unlike the controls.

Another study evaluating a varied exercise programme was undertaken in Brazil [27].Using a cluster randomisation process to randomise 64 participants to treatment and control (daily activity) groups at a ratio of 3:1, the sample included children aged 6–12 years (Mean age=9.4 years; range=6–8; gender not reported), a third of whom were obese. A significant number dropped out due to non-completion of assessments without any intention to treat analyses undertaken. Those in the intervention group received a programme of strength, balance and basic coordination exercises (including climbing, throwing, elastics, ascending steps, stepping, marching) in twice-weekly 40‑minute sessions over 48 weeks (totalling 64 hours). Whilst not differing at baseline in age or pre-test assessments, there was a difference in body dimensions and some biologic markers. At follow-up, regression models nested for psychotropic medication found that the physical exercise group (compared with controls) improved in CARS assessed autistic traits (effect size=1.05) including stereotypical behaviour, verbal and nonverbal social communication skills, cholesterol levels, and parent-reported psychological quality of life (effect size=1.66) and physical quality of life (effect size=1.05), measured on the CHQ-PF50. Glucose and triglycerides did not change.

In China, an RCT recruiting students from special education classes randomised 41 children (Mean age=6.12 years; range=5–8; 70% male) into treatment or control (usual daily activity) groups [68]. The intervention group received a 60 minute twice-weekly physical activity programme over 12 weeks (total 48 hours) of jogging and modified ball games in small groups. The programme actively reinforced communication through prompting, schedules, encouragement, use of visual materials (cue cards, activity schedule, screens) and rewards (stickers and toys). Groups did not differ at baseline in demographic or outcome assessments. A MANOVA Group x Time interaction was found, and follow-up analyses indicated that the intervention group improved in social skills (non-blinded teacher-rated Social Skills Improvement System (SSIS) total score) (p<0.005) whereas the controls did not. Subtests indicated improvement in communication, cooperation, social interaction, and self-control subscales (p<0.005), but not assertion, empathy or engagement. Social interaction was also improved on the ABLLS-R teacher-reported scale for the treatment group compared with the control group.

Finally, an RCT from Hong Kong [67], rated as being of acceptable quality (+), included 40 children (Mean age=9.9 years; range=8–12; 80% male) block randomised (for each 10 consecutive recruitments) into an intervention or waitlist control group. The intervention group received basketball skills training in twice-weekly 45 minutes sessions for 12 weeks (total=18 hours). Sleep quality was measured by a wearable actigraphy accelerometer and a sleep log, and executive functioning assessed blind to condition using two formal assessment tasks (Go Not Go task, and the Corsi block tapping task). With no baseline differences between groups, linear mixed modelling (LMM) analyses revealed a group by time interaction for actigraphy measured sleep, with follow-up analyses indicating improved sleep for the intervention group with respect to sleep efficiency and (on weekdays only) reduced wake after sleep onset time. Sleep onset latency and sleep duration did not differ between groups. Repeated measures ANCOVAS controlling for age found significant Group X Time interaction for an executive function test such that the exercise group improved in inhibition control whereas the control group didn’t. Working memory did not differ between groups.

Table 2.4: Characteristics of primary studies

|  |  |  |  |  |  |  |  |  |
| --- | --- | --- | --- | --- | --- | --- | --- | --- |
| **Author/s (year)** | **Country** | **Quality rating** | **Sample size, gender** | **Age (Mean, range)** | **Intervention** | **Duration of activity** | **Significant Improvement in outcomes\*** | **Non-significant change in outcomes** |
| Dickinson et al (2016) [42] | UK | low quality( – ) | N=10079% male | M=NR(7–16 years) | group exergaming (Nintendo Wii™) | 15 minutes x 3/wk x 40 weeks=30 hours | * improved social functioning
 |  |
| Moradi et al (2018) [47]  | Iran | low quality( – ) | N=50100% male | M=8 years(6–9 years) | group exercise programme (jogging, jumping, hopping, balance, ball skills) | 60 minutes x 3/wk x 12 weeks=36 hours | * reduced stereotypical behaviour
 |  |
| Toscano et al (2018) [27] | Brazil | low quality( – ) | N=64NR | M=8 years(6–9 years) | group exercise programme (climbing, throwing, elastics, ascending steps, stepping, marching) | 40 minutes x 2/wk x 48 weeks=64 hours | * reduced stereotypical behaviour
* improved social communication skills
* improved health-related quality of life
* improved psychosocial health
* improved metabolic health (cholesterol levels)
 | * glucose and triglyceride levels
 |
| Zhao & Chen (2018) [68]  | China | low quality( – ) | N=5070% male | M=6 years(5–8 years) | group physical activity programme (jogging, ball games) | 60 minutes x 2/wk x 12 weeks=48 hours | * social skills (communication, cooperation, social interaction, self-control subscales)
 | * assertion, empathy, or engagement subscales
 |
| Tse et al (2019) [67]  | Hong Kong | acceptable quality( + ) | N=4080% male | M=10 years(8–12 years) | group motor skills training (basketball) | 45 minutes x 2/wk x 12 weeks=18 hours | * sleep quality (some parameters)
* cognition (inhibition control)
 | * working memory
 |

**Key**: ASD=Autism Spectrum Disorder; M=mean; N=total sample size (participants in intervention and control groups); NR=not reported; UK=United Kingdom; /wk=per week

## 2.3 Synthesis of results

The nine systematic reviews included in the current review supported the potential for physical activity interventions to improve the lives of people on the autism spectrum across several domains.

Commenting on physical activity interventions’ effectiveness overall, Dillon et al (2017) [53] concluded that “exercise is an evidence-based practice for school aged children with ASD”. This statement is consistent with that of the US’s National Professional Development Center on Autism Spectrum Disorder (NPDC) [37, 38].

The high quality Canadian systematic review [39] reported moderate to large effect sizes for improvements in social functioning, cognitive outcomes and stereotypic behaviour. The authors observed that results were strongest for martial arts and horse-riding interventions, whereas evidence of effectiveness was weaker for swimming, yoga, and dance interventions.

Considering all secondary evidence appraised, five reviews concluded that physical activity leads to improvements in social functioning [31, 39, 52, 54, 55], and three reviews found cognitive benefits [39, 41, 48] such as improved attention, being on task, and executive functioning.

Motor skills performance improvements were also observed in several systematic reviews, including in the SIGN guideline [52], for locomotor skills in the 2018 meta-analysis of Healy et al [31], and with respect to exercise mastery in the review of Sam et al [55]. Exergaming however was found to have no impact on motor skills performance, except when measured through self-perceived ratings [41].

Physical fitness had variable results. Improvements were reported for skill-related fitness, and muscular strength for interventions generally [31], and for exergaming interventions specifically [41]. However, in two reviews, no impact was reported for physical fitness [55], or for body composition and cardiovascular endurance [31].

A review of group-based interventions in primary school children [40] noted the lack of effect on communication outcomes. Another review found no impact of exergaming on emotional control [41].

The five primary studies appraised were largely consistent with the findings of the secondary reviews. Social functioning was improved in three trials investigating this outcome [27, 42, 68]. Cognition was improved in one trial [67] with respect to inhibition control but not working memory. The same trial also found an improvement in sleep quality from physical activity. Stereotypical behaviour was reduced in two trials [27, 47]. Health related quality of life and some metabolic health factors were also improved following a group exercise programme [27].

## 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 limited by the restriction to English language studies. Restriction by language may result in study bias, but the direction of this bias cannot be determined. However, it seems 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 three of the five included primary studies reported non-significant results for at least some of their outcome measures, and four secondary reviews investigated publication biases and found none [31, 40, 48, 55], with the exception of being possible communication outcomes, which were dealt with using the “trim and fill” adjustment method [31].

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.

The review had a broad scope. A pragmatic decision was made to focus the review on studies explicitly investigating structured physical activity as a key objective. Thus, articles were initially identified where they included any of the following as key terms in their title, abstract, or subject fields: “exercise”, “physical education”, “fitness”, “aerobic”, “physical activity”, “sport”, and “recreation” (alongside search terms relating to autism). This may have excluded studies where specific activities were identified (eg, table tennis) without reference to their purpose (eg, fitness), type (eg, sport, recreation) or being part of a class of interventions (ie, involving physical exercise or activity). By contrast, the search strategy was unrestrictive with respect to study design and sample size. Supplemental searching, including considering the reference list of all retrieved studies, and narrative reviews retrieved as background material, extended the search catchment.

The secondary reviews tended to be conducted by researchers in industrialised, developed and largely Western countries including three in the United States [31, 41, 53], two in Australia [40, 48], two in Canada [39, 54], one in Scotland [52], and one in Hong Kong [55]. However the randomised trials included in these reviews, as well as those appraised in the current review, were conducted in countries across a range of continents including Europe, Asia, South America, North America, and Australia.

It is noted that all 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)

The search, data extraction, synthesis and report preparation was performed by a single reviewer over a limited timeframe (July to October 2019). 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.

The nine appraised systematic reviews published since 2014 included two studies rated as being of “high quality” [39, 52] (using the SIGN critical appraisal checklist [51]), four rated as “acceptable quality” [31, 40, 41, 48], and three of “low quality” [53–55]. Poorer quality reviews used narrow search strategies, lacked supplemental searching, lacked inter-rater reliability reporting or double-coding in data extraction and appraisal, and lacked formal critical appraisal checklists. The recent systematic review of Howells et al [40] commented on the poor design of many studies on this topic, including uncontrolled studies, a lack of randomisation, and before-and-after studies which may overestimate treatment effects. Despite these issues, systematic reviews have tended to aggregate results from across such studies, without discussing their methodological limitations.

When looking specifically at the study designs of studies included in the nine appraised systematic reviews published over the last 6 years, 10 randomised controlled trials were identified. The critical appraisal of the more recently published primary studies included in the current review reveal how such RCTs can nevertheless be open to biases if not conducted well. Four of the included primary studies [27, 42, 47, 68] were rated to be “low quality” using the SIGN appraisal checklist, and one study from Hong Kong [67] was graded as being of “acceptable quality”. No randomised controlled trials appraised in the current review were rated as being of high quality.

Some of the key methodological limitations from these studies are discussed below.

#### Sampling and recruitment

Sample sizes ranged from 40 to 100 (M=61, *k=5*), or 12 to 233 (M=63, *k*=15) when including the 10 RCTs identified in the secondary reviews, which include those assigned to both intervention and control groups.

Limitations included poorly described or limited randomisation (including cluster randomisation and block randomisation) which can reduce the ability of ensuring similarly characterised samples. Ages in the five included primary studies ranged from 5 to 16 years (or 2–17 years in all 15 RCTs identified), and therefore no RCTs included individuals beyond school age. Gender representation was typically weighted towards male participants with all studies including 70% or more males, with one appraised RCT [47] and three RCTs identified in the included systematic reviews [60-62] being restricted to male participants only.

Observing that cognitive ability is often not reported in studies in this area, two systematic reviews suggested there may be a bias towards research conducted on children with higher levels of functioning [39, 48]. It was therefore encouraging to see that in the recently published trials appraised for the current review, two studies’ selection criteria extended to students with moderate to strong learning disabilities [42] and non-verbal IQ above 40 [67], rather than the more common cut-off for study inclusion of 70 [47].

In four of the included primary studies, recruitment was from convenience samples of between one and three special education schools, with no details on recruitment provided for the remaining study [67]. Drop-out rates were not always reported, and in one study [27], 29% of the sample were excluded from analyses due to non-completion of outcomes measures. Whether these initial participants differed from others at baseline was not reported, and biases by their exclusion are therefore unknown.

Sampling biases can affect results in unpredictable ways when studies have small samples, use inadequate recruitment and sampling frames, and have poor response rates and high drop-out rates. Such factors make it less likely that samples are representative of the perspectives and experiences of the broader population the intervention is intended for.

#### Interventions

One of the challenges in synthesising research for this topic is understanding the underlying mechanism for change across such a disparate group of interventions. The following range of activities have been investigated in RCTs in the current review: martial arts, horse riding, athletics, specific motor skills (eg, basketball, table tennis, tri-cycling), exergaming, and mind-body exercises. Aquatic skills have also been researched in this population [69].

It is possible that different activities offer unique physical experiences and benefits [39]. Interventions may also suit some people on the autism spectrum better than others. For example, the repetitive nature of martial arts training may be preferred by or more suited to a person who thrives on structure [39]. Some individuals may prefer group activities whilst others may prefer individual interventions tailored to their specific needs [45].

It is not always clear what the mechanism is that leads to improvements. The necessary components of physical activity interventions and the underlying processes of effectiveness have not been clearly established, particularly across the broad range of outcomes that have been measured in the research conducted to date. (This issue is discussed further under **Section 2.5** below.)

The feasibility of interventions which are empirically supported in research settings being effective and practical to apply in “real world” settings is also variable. Interventions that are too narrowly focused, complex, difficult to implement, costly, or do not meet the perceived needs of the community, impede implementation [53]. For example, some interventions that require high ratios of instructors to participants, specialised equipment, or highly qualified staff, may be difficult to offer in a general community or school setting with limited resources.

The impact of exercise extent and intensity is not well understood. In primary studies appraised in the current review, interventions were undertaken in sessions ranging from between 15 minutes and 60 minutes long, variable times per week, for between 12 and 40 weeks, and summing to 18 to 64 hours (and in the RCTs in appraised secondary studies, 2 to 72 hours). Such extreme variability in intervention frequency, intensity and duration make it challenging to determine optimal frequency and dosage for greatest improvement, or whether there is a ceiling effect after which benefits plateau [39]. Perhaps more importantly, whether benefits are maintained after the intervention ends is an area that has been very poorly attended to in research, with follow-up rarely conducted beyond the immediate completion of the intervention programme.

#### Assessment

Synthesising the research is also challenging because of the diverse range of outcome domains investigated and assessment tools used to measure those outcomes [31], and a lack of standardised assessment scales which have been validated in autistic populations that permit more reliable comparison of effects across studies.

Another methodological issue that can undermine confidence in study results was raised in the systematic reviews: lack of blinding to condition in assessment [40, 41]. That is, assessors or informants knowing whether the participant received the intervention or usual care. Unblinded studies cannot control for participants and observers biased towards seeing an improvement (reporting biases). Assessments can be influenced by expectations about the value of the programme, or perhaps a desire (conscious or not) to assist the researchers for the time and effort invested in offering the intervention. This can artificially inflate ratings of the effectiveness of an intervention. Lack of blinding was evident for all outcome measures in three of the included primary studies [42, 47, 68], and for parent/teacher assessments for another study [27], with only the Hong Kong based study including blinding of both assessors and data analysts [67].

Studies rarely collected direct observational or biometric data to validate whether behavioural improvements (eg, in increased social behaviour, academic performance, or cardiovascular fitness) were evident. Effect sizes were also not generally reported, with some studies comparing differences in group averages rather than individual improvements.

## 2.5 Future research

Future research into physical activity interventions for individuals on the autism spectrum should address the limitations of the current evidence base.

### Sampling and recruitment

Randomised controlled trials will be central to disentangling the factors contributing to benefits from physical activity interventions [31]. Larger samples (determined by power calculations) which are carefully recruited to address sampling biases will provide samples that are more representative of the community and increase the generalisability of findings.

With respect to age groups, greater focus on early childhood and later adolescence has been called for [39], as well as on adults, given the focus of much existing research on children attending primary school. Further, samples should represent a broader range of intellectual functioning, verbal ability, motor skills and coordination, support needs, and economic, cultural and ethnic backgrounds. Not every intervention can or should be applied for every individual. Well-powered and diversely characterised studies are essential to permit the systematic investigation of whether there are sub-populations who are more or less likely to benefit from interventions.

### Interventions

Future research should evaluate interventions delivered in naturalistic settings. This could include home-based exercise programmes delivered by family [39], and community-based, organised group activities (such as team sports) [40]. Another research gap are investigations into different procedures for teaching or maintaining exercise [44].

Feasibility and acceptability are also important. For example, interventions evaluated in research studies often involve high participant to instructor ratios. Such resource intensive programmes may not be practicable in community and home settings. Whether the activity appeals to an individual is also an important consideration. Any intervention that is experienced as burdensome, unappealing, or boring, is unlikely to be taken up or adhered to. Interventions which are engaging, stimulating, and fun, are therefore more likely to be effective. Offering individual choice of intervention type is likely to be crucial to ensure take up of activity over the longer term. Targeting a person’s areas of interest can also be helpful in order to establish their “buy-in” and to maintain their level of motivation. The potential benefit of one activity over another is likely to be less important than the benefit of persisting with any physical exercise over doing nothing because the offered activity did not appeal.

As discussed above, the necessary components of physical activity interventions and the underlying processes of effectiveness have not been clearly established. Researchers have tended to investigate a range of outcomes without necessarily presenting clear ideas of underlying pathways to explain the expected association with the activity. Future intervention research should aim to clearly delineate how an intervention’s components are underpinned by *sound theory*, to allow for reproducibility of research, and achieve real-world impact and generalisability of findings [31]. Understanding the mechanism by which exercise impacts on outcomes is likely to lead to improvements to how the interventions are offered and used to make programmes more effective and efficient for people on the autism spectrum [44].

Some intervention components may be more effective (and more necessary) than others. Rigorously conducted research is needed to assess the contribution of different elements of physical activity programmes that lead to optimal outcomes. Moderators and mediators of effectiveness could include features such activity type, overall duration, intensity, delivery (in groups or individually), setting, and location. For example, future research could explicitly randomise different frequency and doses of physical activity, different forms of physical activity controlled for intensity [39], and compare an offer of choice of activity over no choice.

### Assessment

Outcomes considered across the studies evaluated in the current review varied greatly in terms of outcome domain being measured, as well as assessment tools employed. More consistent use of validated, standardised instruments would provide more reliable indicators and permit meaningful study comparisons, syntheses and meta-analyses. Where possible, outcomes should also be completed by independent, blinded-to-condition assessors from a range of informants. Measures of exercise intensity have been lacking, and the use of activity and physiological monitors could provide more reliable indicators of adherence to a programme [39]. The reporting of effect sizes, and the number of individuals achieving clinically significant benefits and not just group mean changes [48], is also recommended. Finally, longer term follow-up is needed to see whether benefits, including physical, social, cognitive, and behavioural, are maintained over time or cease when the activity programme ends. It is encouraging in this regard that a randomised controlled trial is currently under investigation evaluating a 14-week cross-fit programme for autistic children which includes an 8-week follow-up assessment [70].

## 2.6 Systematic review conclusions

This systematic review updates evidence for the New Zealand Autism Spectrum Disorder Guideline [9] with respect to the effectiveness of physical activity interventions for people on the autism spectrum. Following a comprehensive database search and citation searching of primary and secondary studies published since 2014, 1017 unique abstracts were identified. After applying inclusion and exclusion criteria, 14 studies were included for critical appraisal: nine systematic reviews, and five primary studies collecting original data.

This represents a significant upsurge in research in this field over the last decade given that physical activity was barely mentioned in the original NZ guideline (2008) [1].

The current review suggests that there is evidence across a reasonable body of primary and secondary evidence including RCTs that physical activity interventions can provide benefits across a number of domains for individuals on the autism spectrum. More specifically:

* There is consistent evidence that physical activity interventions appear to show significant improvements on social functioning outcomes such as social interaction skills.
* Cognitive functioning and behavioural functioning measures are also improved in most studies evaluating outcomes in these domains.
* There is little evidence that communication is improved by physical activity. However the lack of communication effects may be due to small sample of studies including such outcomes [40], leading analyses to lack power to identify small improvements.
* Motor skill development and general physical fitness, considered secondary outcomes of the current review, were also improved, most notably in studies where specific motor skills were explicitly taught [61, 65].

Possible explanations for discrepancies between studies include heterogeneity of participants, variability in intervention and duration, variability in outcome domains and measures employed, and the quality of studies themselves [40]. Research studies, including randomised controlled trials, were commonly limited by smaller samples. However it should be noted that despite relatively small samples, medium to large effect sizes were found for improvements in stereotypic behaviour, social functioning, and cognition/attention outcomes [39].

There is currently a research gap in understanding the impact of physical activity for pre-school aged children and adults. It is also not clear whether benefits found in some studies are likely to apply to participants with different levels of cognitive ability or ASD severity.

Assessment was limited by a lack of blinded, objective indicators. Follow-up to determine maintenance of effects are infrequent in this literature and it is therefore difficult to assess whether benefits are sustained over the longer term. It is also not clear whether there is an optimal level or dose (frequency and duration) of activity to maximise benefits. Notably two studies which did include follow-up and had larger doses of physical activity had some of the largest impacts on social functioning and communication [40].

Whilst several systematic reviews have attempted to conduct meta-analyses, synthesising the current evidence base is challenging given the diversity of interventions, outcomes, and measures involved. It is not currently possible to offer specific recommendations about the most successful and necessary components, delivery, and duration of physical activity interventions. Ideally such research should be undertaken in naturalistic settings, offering a range of appealing and easy to implement activities that groups of/or individuals on the autism spectrum choose to participate in. Beyond scheduled activity programmes, a clear objective should be to encourage people on the autism spectrum (as for neurotypical people) to incorporate physical activity into their daily routine as an enduring lifestyle.

# 3 Recommendation development

The Living Guideline Group (LGG) was tasked with considering the systematically reviewed evidence reported in **Section 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 existing recommendations and good practice points (GPP) as well as the development of new ones.

The text and graded “strength of evidence” of recommendations and GPP (see **Appendix 1**, **Table A1.2**) were revised/developed at an all-day face-to-face meeting. The LGG’s decisions are presented below, and summarised in **Summary Tables I** and **II** (p. x). 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

This update was informed by a systematic review updating the evidence for the effectiveness of physical activity interventions for people on the autism spectrum.

Physical activity interventions involve repeated gross-motor movements requiring physical exertion. In the current review, these are defined as planned, structured, repetitive, and purposeful.

The health benefits of physical exercise for virtually everyone are irrefutable and well established across many domains [16]. Regular physical activity is an effective primary and secondary preventative strategy for more than 25 chronic medical conditions with common risk reductions of 20%–30% [17].

People on the autism spectrum are less likely to engage in physical activity, and are at greater risk for the development of obesity [22, 23]. Whilst many risk factors for chronic medical conditions are likely the same, individuals on the autism spectrum who often have co-occurring conditions may be more susceptible.

### Existing Recommendations and Good Practice Points

One existing recommendation in the Guideline [9] was considered for revision by the Living Guideline Group.

* **Original Recommendation 2.3.9:** “Research should be undertaken to identify the needs of people with ASD with regard to constipation, allergies, medication reactions, menstruation and exercise.” (Grade C)

**Unchanged**

**Rationale**: Recommendation was left unchanged as it remains accurate.

### New Recommendations

* **New Recommendation** 2.3.9a: “Physical activities provide benefits across social, cognitive and behavioural domains in addition to general well-being and should be considered for children and young people on the autism spectrum.” Grade B

**Rationale**: There is consistent evidence that physical activity interventions appear to show significant improvements on social functioning outcomes such as social interaction skills. Cognitive functioning and behavioural functioning measures are also improved in most studies evaluating outcomes in these domains.

**Additional text:** Although the existing evidence related primarily to children and young people, it is likely to apply to adults.

**Rationale**: Three of the appraised systematic reviews [41, 48, 52] did not have an age restriction for included studies, and included study participants aged up to 25 years.

### New Good Practice Points

* New Good Practice Point 2.3.9b: “When supporting individuals participating in a physical activity or programme, their preferences should be respected and needs accommodated”. ✓
* New Good Practice Point 2.3.9c: “Further research that targets meaningful outcomes for the autistic community is essential.” ✓

**Rationale**: Research outcomes to date have primarily focused on addressing the core features of autism. However, there is increasing demand from the autistic community for research into outcomes that are important to them. It is unclear the extent to which the reviewed research outcomes meet these expectations.

* New Good Practice Point 4.1.6: “Further research that targets meaningful outcomes for the autistic community is essential.” ✓

**Rationale**: It is recognised that whilst Good Practice Point 2.3.9c was developed in response to the research considered in the review of physical activity interventions, it was also relevant to other research in the guideline, and particularly to that presented in Part 4 on “Treatment and Management of ASD”. For this reason, the GPP is also presented prominently in Part 4’s summary of recommendations in its Introduction section.

**Summary Tables I** and **II** (p. x) presents the revised recommendation and new Good Practice Points.

# Appendix 1: Methods

## A1.1 Contributors

This systematic review was conducted, and the report prepared, by **Marita Broadstock** (INSIGHT Research). The recommendations and good practice points were developed and graded by the LGG based on discussion of the systematic review’s body of evidence as it updates that considered in the guideline [9].

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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 1 August 2019 and updated on 6 September 2019. 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)
* SPORTDiscus (Ovid)
* 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 syntax is offered used for databases accessed through EBSCOHost:

(exercise# OR physical education OR fitness OR aerobic OR physical activit\* OR sport# OR recreation\*)

AND

(autism spectrum disorder OR autism OR autist\* OR ASD OR asperger#)

These searches were made in the title, abstract and subject fields, and were limited to English language, 2004-current, human, and peer reviewed academic journals. Hand searching of journals was not undertaken. Refinement of scope to consider best evidence led to restriction of the publication dates to 1 January 2014 to current.

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 [50]

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, so as to rank them in terms of quality from most robust (level I) to least (level IV) [50] (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 a sufficient body of level I and II evidence to permit study designs to be restricted to systematic reviews and meta analyses covering relevant research questions published since 1 January 2014.

## 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:

* study design (ie, experimental or quasi-experimental), level of evidence, SIGN quality rating
* sample characteristics including age, gender, participation rate
* setting
* duration
* whether intervention was manualised
* outcomes measured (including scales employed and completed by whom)
* results
* authors’ conclusions
* key methodological strengths and weaknesses
* funding source of study
* for secondary papers, included primary studies that meet selection criteria for the current review.

## 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 SIGN quality checklists from the Scottish Intercollegiate Guidelines Network [51] 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).

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** (+): 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 2019 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 revised affected recommendations (and good practice points) from the guideline [9] and developed 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 [9] 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 [50]:

* 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 [9].

## 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 through an online survey. 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 six organisations/individuals representing a 75% response rate. The lead researcher (INSIGHT Research) collated feedback and drafted revisions for the LGG to consider. Amendments were finalised by group consensus. 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

AS Asperger’s syndrome

ASD Autism Spectrum Disorder

BMI Body Mass Index

EBP Evidence-based Practice

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

M mean

ns not significant

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

NHMRC National Health and Medical Research Council (Australia)

NR not reported

p. page number

PDD Pervasive Developmental Disorder

PDD-NOS Pervasive Developmental Disorder – Not Otherwise Specified

RCT randomised controlled trial

SIGN Scottish Intercollegiate Guidelines Network

UK United Kingdom

US United States of America

vs versus

/wk per week

### Tests, scales and measures

ABLLS-R ABLLS-R=Assessment of Basic Language and Learning Skills-Revised;

APAT Adapted Physical Activity Taxonomy

BDS Backward Digit Span test

CBTT Corsi block tapping task

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

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

FDS Forward Digit Span test

GARS Gilliam Autism Rating Scale

GNG Go Not Go task

PEDro Physiotherapy Evidence Database

QUIPS Quality in Prognostic Studies Tool

SDQ Strengths and Difficulties Questionnaire

SRS-2 Social Responsiveness Scale

SSIS-RS Social Skills Improvement System Rating Scales

TAT Tinetti Assessment Tool

WHrT Waist to Height Ratio

### Databases

CDSR Cochrane Database of Systematic Reviews

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

|  |  |
| --- | --- |
| **Animal assisted therapy (zoo therapy)** | Therapy that uses pets to improve the physical and mental health of people. In autism specifically, it can be used to help children learn empathy, communication and social skills. It can also be used to help children with ASD manage their behaviour. |
| **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. |
| **Case-control study** | Patients with a certain outcome or disease and an appropriate group of controls without the outcome or disease are selected (usually with careful consideration of appropriate choice of controls, matching, etc.) and then information is obtained on whether the subjects have been exposed to the factor under investigation. |
| **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. |
| **Cohort study** | Subsets of a defined population can be identified who are, have been, or in the future may be exposed or not exposed in different degrees, to a risk factor or factors hypothesised to influence the probability of occurrence of a given disease or other outcome. Subjects are followed from a well-described starting point to determine whether the outcome/disease occurs (either retrospectively, or prospectively). The control group of people not exposed to the risk factor can be identified within the population-based cohort, and be matched by confounders known to be associated with the outcome (eg, age, sex), or can be obtained from an historical cohort. Studies usually involve the observation of a large population, for a prolonged period (years).A prospective cohort study is where groups of people (cohorts) are observed at a point in time to be exposed or not exposed to an intervention (or the factor under study) and then are followed prospectively with further outcomes recorded as they happen.A retrospective cohort study is where the cohorts (groups of people exposed and not exposed) are defined at a point of time in the past and information collected on subsequent outcomes, eg, the use of medical records to identify a group of women using oral contraceptives five years ago, and a group of women not using oral contraceptives, and then contacting these women or identifying in subsequent medical records the development of deep vein thrombosis. |
| **Convenience sampling** | Also known as grab sampling, accidental sampling, or opportunity sampling, Convenience sampling is a type of non-probability sampling that involves the sample being drawn from that part of the population that is close to hand. |
| **Criterion sampling** | Involved selecting cases that meet some predetermined **criterion** of importance. For example: Every person that meets selection criteria within a service environment is invited until a certain sample size criterion has been met. |
| **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. |
| **Dolphin therapy** | Any kind of intervention (treatment or service) which involves dolphins. This interaction may include touching or kissing the dolphin, or getting into the water and taking a ride holding onto the dolphin’s dorsal fin. |
| **Effect size** | A quantitative measure of the strength of a phenomenon, a standardised measure of the size of the difference between two groups. |
| **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. |
| **Equine-assisted therapy** | An alternative multimodal intervention that involves utilizing a horse to enhance core impairments of ASD. Can include grooming, walking alongside, and horse riding activities. |
| **Exergaming** | Technology-driven physical activities, such as video game play, that requires participants to be physically active or exercise in order to play the game. |
| **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. |
| **Hippotherapy** | A form of physical, occupational and speech therapy in which a therapist uses the characteristic movements of a horse to provide carefully graded motor and sensory input. |
| **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 recognised 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. |
| **Non-randomised, experimental trial** | The unit of experimentation (eg, people, a cluster of people) is allocated to either an intervention group or a control group, using a non-random method (such as patient or clinician preference/availability) and the outcomes from each group are compared.This can include:* a controlled before-and-after study, where outcome measurements are taken before and after the intervention is introduced, and compared at the same time point to outcome measures in the control group.
* an adjusted indirect comparison, where two randomised controlled trials compare different interventions to the same comparator, ie, the placebo or control condition. The outcomes from the two interventions are then compared indirectly.
 |
| **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. |
| **Pet therapy** | A form of animal therapy involving a pet animal (eg, a dog, hamster). |
| **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. |
| **Purposive sampling** | A non-probability sample that is selected based on characteristics of a population and the objective of the study. Purposivesampling is also known as judgmental, selective, or subjective sampling. |
| **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 subjects in a population are randomly allocated into groups to receive or not receive an experimental preventive or therapeutic procedure, manoeuvre, or intervention. The groups are compared prospectively. |
| **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). |
| **Service/assistant dog** | A dog trained to assist a person with autism to help them gain independence and the ability to perform activities of daily living and improve their quality of life. |
| **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 |
| **Zoo therapy** | See *animal assisted therapy* |

# 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

| **Dickinson et al (2016) [42]** |
| --- |
| **Country, study, aims** | **Participants** | **Selection criteria** | **Intervention, control, outcomes** | **Results** | **Conclusions, quality issues** |
| **Country:** UK**Study type:** randomised controlled trial (RCT)**Evidence level:** II**Study Quality** (SIGN checklist): – (low quality)**Aims:** to examine if a computer-based physical activity programme could improve the social functioning of children with ASD. | **Setting:** Recruited from one of three schools with specialist education classes.**Participants:** 100 children diagnosed with ASD; 67% aged7–10, 33% aged 11–16 years; 79% male. Participants pooled from three schools and randomly allocated into treatment or control groups controlling for age and genderTreatment group (TG): N=50Control group (CG); N=50**Dropout:** none reportedRandomisation method or how age and gender were controlled between groups was not reported. | **Inclusion:** confirmed diagnosed with ASD; aged7–-16 years; moderate or severe learning disability.**Exclusion:** physical illness or disability that would adversely affect their use of the computer game, comorbid mental health problems requiring use of psychoactive medication.**Follow-up:** pre-test, immediately post-test only.**Fidelity:** researcher made random visits to participating schools to monitor intervention delivery. | **Treatment (TG):** use of Nintendo Wii™ motion-capture game and software package of multi-player sports games (aquatics, athletics, fencing, table tennis), in addition to routine school physical education classes: 15 minutes sessions 3x week, for a school year (about 40 weeks) (total dose=30 hours). Classes led by teacher and assistants with games involving 2–4 players.Control (CG): control receiving routine school physical education classes.**Outcomes****Staff Questionnaire:** Social Behaviour at School (child and adolescent versions): social functioning (completed by teachers).**Blinding:** Assessments were not blind to condition. | At baseline, no significant differences between groups in age group, gender, parental employment, family functioning, or social functioning. TG more likely to come from one-child families than CG (18% cf 50%).**Key findings**Analyses were performed separately for girls and boys in two age groups (7–11 and 11–16 years). Mann Whitney U test showed social functioning improved over time for the TG but not for the CG group (in both age ranges).ANCOVAs investigated these changes:* Children improved significantly in social functioning in the TG compared with CG (*F*=66.18, p<0.001).
* Considering boys only, improvements were evidence for both younger boys (*F*=45.99, p<0.001), and older boys (*F*=4.46, p<0.05).
* There was no significant improvement for girls in either age group.
 | **Author conclusions:** computer-based activity programme has the potential to produce improvement in social functioning, as part of a physical education programme.**Reviewer’s comments:** ASD diagnoses verified. Moderate sample size. Assessment was not blind to condition. The non-blinded, teacher-completed social functioning scale had not been used for people with ASD before and has no formal psychometric data available, being designed to measure the impact of an intervention rather than provide as a measure of absolute social skills. Children and family members did not complete any outcome measures. Sample sizes for girls within age groups were very small, reducing power to identify real effects. Level of physical exertion not physiologically measured. Maintenance not assessed.The computer game encouraged multi-player use and peer interactions and/or physical activity could have been the mechanism for improvements in social functioning.**Source of funding:** none reported. |

| **Moradi et al (2018) [47]** |
| --- |
| **Country, study, aims** | **Participants** | **Selection criteria** | **Intervention, control, outcomes** | **Results** | **Conclusions, quality issues** |
| **Country:** Iran**Study type**: randomised controlled trial (RCT)**Evidence level**: II**Study Quality** (SIGN checklist): – (low quality)**Aims:** to examine the effect of perceptual-motor exercises on the reduction of stereotypical behaviour in children with ASD (also considered vitamin D supplementation, not reported here). | **Setting**: Recruited from one of three “specialised institutions for youth with ASD” where activity and assessment took place.Of 250 approached, 139 were ineligible, 11 families chose not to participate, and 50 were randomly allocated to other treatment groups not reported here.**Participants**: 50 boys; M age=7.62 years (SD=1.15); diagnosed with ASD.Randomised into control or treatment groupTreatment group (TG): N=25Control group (CG); N=25**Dropout**: noneRandomisation method not reported. | **Inclusion:** confirmed diagnosed with ASD; aged 6–9 years; IQ more than 70; had vitamin D serum level <30ng/ml; sound sight and hearing; lack of orthopaedic or respiratory disorders; lack of seizure attacks in previous two years.**Exclusion**: gastrointestinal. feeding problems or malnutrition; genetic or autoimmune disorders; anaemia; neurological diseases; metabolic disorders; endocrine, cardiovascular, pulmonary, and liver or kidney disease; history of severe head trauma or stroke; receiving vitamin D.**Follow-up:** pre-test, immediately post-test only.**Fidelity**: instructor supervised by researcher. | **Treatment (TG)**: received instructor-led group programme of exercises including jogging, jumping, hopping, balance, ball kicking, ball throwing, ball throwing and catching; one hour session, 3 x week, for 12 weeks (total dose=36 hours).**Control (CG)**: control receiving no intervention, placebo (for vitamin D).**Outcomes**GARS-2: Stereotypy subscale, assessing previous week’s behaviour through consensus judgement in interviews with parents, teacher, care-givers, and direct observation.**Blinding:** Assessments were not blind to condition. | At baseline, no significant differences between groups in age, autism severity, IQ, vitamin D level.**Key findings**:Paired samples t-test showed stereotypy decreased between pre- and post-test for the treatment group (p=0.01), but not for the control group (p=0.51).An ANCOVA demonstrated that the three treatment groups (2 others relate to different interventions not reported here) differed from the control group but an analysis specifically for this TG was not reported. | **Author conclusions**: perceptual motor exercises significantly reduced stereotypical behaviours in children with ASD.**Reviewer’s comments**: ASD diagnoses independently verified. Small sample of participants without ID and with many health exclusions. Control group had placebo tablet (to control for another treatment arm). Assessment was not blind to condition. Level of physical exertion not physiologically measured. Potential for effects relating to other aspects of group activity and not physical movement *per se*. Maintenance not assessed.**Source of funding**: none reported. |

| **Toscano et al (2018) [27]** |
| --- |
| **Country, study, aims** | **Participants** | **Selection criteria** | **Intervention, control, outcomes** | **Results** | **Conclusions, quality issues** |
| **Country:** Brazil**Study type**: randomised controlled trial (RCT)**Evidence level**: II**Study Quality** (SIGN checklist): – (low quality)**Aims:** to examine the effects of an exercise-based intervention on the metabolic profile, autism traits, and perceived quality of life of children with ASD. | **Setting**: Recruited from a paediatric centre for people with ASD.**Participants**: 64/90 children diagnosed with ASD; M age 8.4 years; gender not reported; 43 with autism, 9 with Aspergers, 12 with developmental disorder; 21 with no prescribed medication, 3 with stimulants and antiepileptics, and 40 with antipsychotics; 27% were overweight and 33% were obese.Cluster randomisation to TG and CG was applied at a ratio of 3:1 (67 to 23, respectively) to address potential dropout and poor compliance.Treatment group (TG): N=46Control group (CG); N=18**Dropout**: 26 dropped out (21% of those allocated to CG, and 31% of those allocated to TG) due to non-completion of outcome measures, and (for some on TG) not attending >90% of programme. | **Inclusion:** confirmed diagnosed with ASD; aged6–12 years.**Exclusion**: non-attendance of more than 10% of sessions.**Follow-up:** pre-test, immediately post-test only**Fidelity**: not reported. | **Treatment (TG)**: use of exercise-based intervention involving strength, balance and basic coordination exercises (eg, climbing, throwing, elastics, ascending steps, stepping, marching): 40 minutes sessions 2 x week, for 48 weeks (total dose=64 hours). Classes of up to 3 children led by first author (physical educator) and parents/guardians.**Control (CG)**: control receiving usual daily activity.**Outcomes**Anthropometry measurements including stature, body mass index (BMI), and waist to height ratio.Biological markers by fasting blood test: plasma glucose, triglycerides, cholesterol levels.CHQ-PF50 (completed by parents/guardians): physical health, psychological health.CARS ASD traits (completed by clinical psychologist)**Blinding:** All tests were blind to condition except the parent/guardian completed CHQ-PF50. | At baseline, no significant differences between groups in age, autistic traits, physical or psychological health. However there were differences in groups for body dimensions, and some biological markers.**Key findings**:Baseline characteristics which varied at baseline included in multilevel regression models, nested for psychotropic medication which varied for body dimensions.The TG compared with the CG showed significant:* increases in HDL-C cholesterol (5.2 mg/dL, 95%CI 2.2–8.1 mg/dL, effect size=0.67)
* decreases in LDL-C cholesterol (‑7.7 mg/dL, 95%CI -14.5 and -0.9 mg/dL, effect size=0.43)
* decreases in total cholesterol (‑10.1 mg/dL, 95%CI -19.0 to1.3 mg/dL, effect size=0.88)
* decreases in CARS autistic traits (-8.1, 95%CI -12.2 to -4.0, effect size=1.05)
* increases in parental assessed quality of life for physical health (13.3, 95%CI 7.7–18.9, effect size=1.05), and increases in psychosocial health (15.2, 95%CI 9.8-20.7, effect size=1.66).

No change in glucose or triglycerides. | **Author conclusions**: children exposed to intervention compared with control group showed significant positive effects in their improved metabolic health, reduced autistic traits (particularly in stereotypical behaviour and improvements in verbal and nonverbal social communication skills), and physical and psychological health (perceived by parents).**Reviewer’s comments**: ASD diagnoses verified. Moderate sample size. High drop-out rates due to high outcome non-completion rate suggests potential biases in excluding students who were not well and/or for whom the intervention did not appeal. Intention to treat analyses not undertaken. Use of a non-blinded, parent-completed physical and psychological health scale, and a blinded, clinician reported autism traits scale. Level of physical exertion not physiologically measured. Maintenance not assessed.**Source of funding**: none reported. |

| **Zhao & Chen (2018)** **[68]** |
| --- |
| **Country, study, aims** | **Participants** | **Selection criteria** | **Intervention, control, outcomes** | **Results** | **Conclusions, quality issues** |
| **Country:** China**Study type**: randomised controlled trial (RCT)**Evidence level**: II**Study Quality** (SIGN checklist): – (low quality)**Aims:** to investigate the effects of structured physical activity program on social interaction and communication of children with ASD. | **Setting**: Recruited from special education school for children with ASD. Activity took place in fitness room on in the playground of the school.**Participants**: 41/50 children diagnosed with ASD; M age 6.1 years; 70% male; 33 with autism, 7 with Aspergers syndrome, 1 with PDD.Randomised into groups:Treatment group (TG): N=25Control group (CG); N=20**Dropout**: 4 (16%) children dropped out of the TG and 5 (20%) from the CG (reasons not reported). | **Inclusion:** confirmed diagnosed with ASD; aged 5–8 years.**Exclusion**: none reported.**Follow-up:** pre-test 1 week prior to intervention period, interim-test (after 6 weeks), and post-test one week after intervention period.**Fidelity**: not reported. | **Treatment (TG)**: use of structured physical activity programme of jogging and modified ball games in small groups (n=5) and whole groups, targeting social interaction and communication of children with ASD: 60 minutes sessions 2 x week, for 12 weeks (total dose=48 hours). One teacher for every 5 students, and one volunteer assistant for every 2 students.**Control (CG)**: control receiving usual daily activity.**Outcomes** (completed by)SSIS: subscale for social skills (teachers)ABLLS-R: social interaction (teachers)Qualitative data about programme collected through semis-structured interviews and open ended questionnaires (parents and volunteers).**Blinding:** not reported so assumed not blinded. | At baseline, no significant differences between groups in age, gender, diagnosis, or social skills (SSIS) or social interaction (ABLLS-R).**Key findings**Repeated measures Group (Treatment vs Control) X Time (pre vs post) MANOVA found:* significant Group X Time interaction for Social Skills: F(2,38)=36.52, p<0.05, Wilks Lambda=0.342).
* Follow-up analyses confirmed TG improved in SSIS overall score between pre- and post-test (p<0.005), but not the CG.
* Analysis of SSIS 7 sub-domains: TG cf CG improved in *communication, cooperation, social interaction*, and *self-control* subdomains (𝑝<0.005), but not for *assertion, empathy* or *engagement* subdomains.
* Two-way repeated measure ANOVA found significant improvement in ABLLS-R social interaction for TG cf CG (𝑝 <0.005).

Qualitative responses from parents and volunteers were transcribed and themes identified. These relate to the programme encouraging steps in social interaction including positive acceptance and eye contact, cooperative play and positive communication, and positive engagement and interactions. | **Author conclusions**: a specially structured physical activity program positively influenced social interaction and communication skills of children with ASD, especially in social skills, communication, prompt response, and frequency of expression.**Reviewer’s comments**: ASD diagnoses verified. Moderate sample size. Assessment not reported as blind to condition. Level of physical exertion not physiologically measured. Maintenance not assessed.**Source of funding**: none reported. |

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| **Tse et al (2019) [67]**  |
| **Country, study, aims** | **Participants** | **Selection criteria** | **Intervention, control, outcomes** | **Results** | **Conclusions, quality issues** |
| **Country:** Hong Kong**Study type**: randomised controlled trial (RCT)**Evidence level**: II**Study Quality** (SIGN checklist): + (acceptable quality)**Aims:** to examine the impact of physical activity on sleep quality and cognition in children with ASD. | **Setting**: Intervention occurred in school hall/gymnasium of “each participating school”. How children were recruited and how many schools were involved was not reported.**Participants**: 40/50 children diagnosed with ASD; M age 9.9 years; 80% male; 6 participants were taking regular medication during the study with no reported side effects, including hydrocortisone (n=2), fusidic acid cream (n=3) and risperidone (n=1).Block randomised into groups (5 into each group for every 10 consecutively recruited).Treatment group (TG): N=19Control group (CG); N=21**Dropout**: 6 (12%) children of the TG and 4 (8%) from the CG were excluded due to missing data (poor performance on GNG task, failing to respond correctly to at least 50% of the Go Trials) | **Inclusion:** confirmed diagnosed with ASD; aged 8–12 years; non-verbal IQ above 40; ability to follow instructions, perform physical intervention and executive function measures, no prior basketball skill training, no history of reading disabilities.**Exclusion**: medical conditions that limited physical activity capacity (eg, asthma, epilepsy).**Follow-up:** one week pre-test prior to intervention period, and one week post-test, no follow‑up.**Fidelity**: Attendance rate of 97.9%. Actigraphy assessment and sleep log were significantly correlated (*r=*0.18–0.33). | **Treatment (TG)**: basketball skills training: 45 minutes, sessions 2 x week, for 12 weeks (total dose=18 hours). Staff/participant ratio1:3–1:2.**Control (CG)**: waitlist control, no basketball intervention asked to follow their normal daily routine without any “additional exercise programme”.**Outcomes**SRS-2: social impairment, at baseline only (parents)*Sleep parameters:* SE (sleep efficiency: proportion of sleep time since going to bed); WASO (wake after sleep onset); SOL (sleep onset latency); SD (sleep duration), measured by wearable actigraphy accelerometer, and sleep log.*Executive functions* (with order counterbalanced across participants):GNG Task: False Alarm (FA) errors indicating poorer inhibition control.CBTT (Corsi block tapping task), FDS (forward digit span) test, BDS (backward digit span) test: measures visual-spatial and auditory working memory.**Blinding:** Assessors and data analysts were blind to condition.  | At baseline, no significant differences between groups in age, gender, BMI, IQ, medication use, SRS-2, sleep, executive functioning.**Key findings**:Linear mixed modelling found Group X Time interactions (actigraphy):* For *weekday* assessments, TG improved in sleep efficiency and reduced wake after sleep onset time (*d*from 0.66–0.77; all *ps*<0.001); for CT opposite occurred (*d*, 0.59–0.68; all *ps*<0.001). No difference in sleep onset latency, or sleep duration.
* For *weekend*, the TG improved in sleep efficiency (*d*=0.003, *ps*<0.01) whereas the CT group didn’t change (*p*>0.05). No differences in reduced wake after sleep, sleep onset latency, sleep duration.
* Similar results for sleep log.

Repeated measures ANCOVAS controlling for age found significant Group X Time interaction for inhibition control (FA) in GNG task (*F*(1,37)-6.58, *p*=0.02) such that:* TG had significantly improved (ie, fewer) FA at post-test (*t*(20)=2.55, *p*=0.02) but not in the CG (*p*=0.72).
* No improvement in working memory (CBTT, FDS, BDS) found in either group (ps>0.05).
* Inhibition control (FA) correlated with actigraphy-measured Sleep Efficiency (*r=*-0.43, *p*=0.006).
 | **Author conclusions**: physical activity can improve sleep quality and cognition among children with autism spectrum disorder, but specific physical activity may be required to benefit individual executive functions.**Reviewer’s comments**: Recruitment and number of schools not reported. Power calculation used to determine sample size. Some drop outs were excluded due to high FA rates indicating poorer inhibition control, with no intention to treat investigating their sleep measures. Valid and reliable assessment measures. Assessment was blind to condition. Level of physical exertion not physiologically measured. Baseline general physical activity not measured. Control group may have altered their exercise knowing they were in the control group. Odd that their sleep worsened over time (measurement effects could be at play). The TG may have altered physical activity throughout the week because of the tasks (for example, practising their new skills). No statistical accounting for multiple tests and possibility of chance effects. Maintenance not assessed.**Source of funding**: Research Grants of Hong Kong Special Administrative Region, China. |

**Key:** ABLLS-R=Assessment of Basic Language and Learning Skills-Revised; ADHD=attention deficit hyperactivity disorder; AS=Asperger’s syndrome; ASD=Autism Spectrum Disorder; BDS=Backward Digit Span test; BMI=Body Mass Index; BOT-2=Bruininks-Oseretsky Test; CARS=Childhood Autism Rating Scale; CBTT=Corsi Block Tapping Task; CHQ-PF50: Child Health Questionnaire, Portuguese version; DSM-IV=Diagnostic and Statistical Manual of Mental Disorders, 4th edition; DSM-5=Diagnostic and Statistical Manual of Mental Disorders, 5th edition; FA=False Alarm error (in GNG task); FDS=Forward Digit Span test; GNG=Go Not Go task; ID=intellectual disability; IQ=intelligence quotient; ns=not significant; PDD=pervasive developmental disorder; GARS-2=Gilliam Autism Rating Scale-2; RCT=randomised controlled trial; SIGN=Scottish Intercollegiate Guidelines Network; SRS=Social Responsiveness Scale; SSIS-RS= Social Skills Improvement System Rating Scales; UK=United Kingdom; US=United States of America.

Table A3.2: Evidence Tables for included secondary studies

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| **Sam et al (2015) [55]** |
| **Country, study type, aim** | **Search strategy** | **Appraisal methods** | **Results** | **Conclusions** |
| **Country:** Hong Kong**Study type**: systematic review and meta-analysis**Evidence level**: I**Study Quality** (SIGN checklist): – (low quality)**Aim**: Evaluate the effectiveness of exercise interventions on exercise mastery, physical fitness, and social competence in children with ASD. | **Databases**: MEDLINE, ProQuest, Scopus, Web of Science “related articles”, and “generally accessible websites” of major Journal publishers, Google Scholar, “and etc”.**Search**: Searched from 2004–2014. Transparent selection criteria. Range of keywords relating to organised, intervention, and population. No supplemental searching reported.**Selection criteria**: children (not defined) with a diagnosis of ASD; involved controlled interventions that involved “some kind of physical exercise”; included quantitative measures of behavioural effects.Excluded: non-controlled studies, non-English studies, unpublished articles (conference papers). | **Method:** One author assessed titles and abstracts for relevance, extracted data, and appraised studies.Quality was assessed using Jadad scale for intervention studies.Effect sizes (Cohens *d*) were calculated.A meta-analysis was conducted. Observation of funnel plots suggested no publication bias. | **Included**: 249 unique titles identified, 8 met criteria for inclusion in the review (n=173, *n*range 10–40; *age* range 5–19 years).Evaluated exercises included: aquatic exercise (n=4), horse riding (n=1 study), kata martial arts techniques (n=1), Chinese mind-body exercise (n=1), and treadmill walking (n=1).Four studies were quasi-experimental controlled studies, one was single subjects experimental study, and 3 were RCTs [56, 58, 59].**Key findings**From the meta-analysis:* significant improvement in exercise mastery functioning; *k*=5 studies, *d*=0.57,CI(0.22–0.92), p<0.05
* significant improvement in social competence; *k*=4 studies, *d*=0.58,CI(0.21–0.95), p<0.05
* non-significant effect for physical fitness; *k*=5 studies, *d*=0.36, CI(-0.02, 0.74), p>0.05.
 | **Author conclusions**: Current evidence showed that exercise interventions were effective for ASD children in exercise mastery and social competence.**Reviewer’s comments**: Narrow search strategy with few of the main databases and no supplemental searching (leading to a small number of abstracts screened). One researcher involved. Appraisal checklists used. Brief description of results. Only 3 studies were RCTs.**Source of funding**: non reported**Included RCTs (intervention)**:* Bass et al (2009) [56] – horse riding
* Bahrami et al (2012) [58] – martial arts/kata
* Chan et al (2013) [59] – mind-body exercise.
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| **Bremer et al (2016) [39]** |
| **Country, study type, aim** | **Search strategy** | **Appraisal methods** | **Results** | **Conclusions** |
| **Country:** Canada**Study type**: systematic review**Evidence level**: I**Study Quality** (SIGN checklist): ++ (high quality)**Aim**: Review the impact of exercise interventions on the overall behaviour of individuals with ASD from birth to 16 years of age. | **Databases**: ERIC, MEDLINE, ProQuest Nursing, PubMed, and SPORTDiscus.**Search**: Searched from database commencement to July to September 2014. Transparent selection criteria. Broad range of keywords relating to population, intervention, age, outcomes. No supplemental searching reported.**Selection criteria**: studies with: at least one participant aged 0–16 years with a diagnosis of ASD/PDD; evaluated the effectiveness of exercise interventions; compared to no or different intervention; were randomised and non-randomised controlled trials, cohort studies or pilot studies; included one or more outcomes measuring stereotypic behaviours, cognition/attention, social-emotional behaviours, additional behaviours; and were published in the English language.Excluded: educational only interventions (ie, suggestions to engage in exercise); studies appraised as low quality; non research articles, books/chapters, conference proceedings, abstracts, lectures, consensus development statements, systematic reviews, meta-analyses, and guidelines. | **Method:** Two researchers independently assessed titles and abstracts for relevance (relevant, possibly relevant, irrelevant), and resolved disagreements about eligibility through discussion and consensus. Single researcher extracted data into evidence tables, and second researcher independently reviewed extracted data for accuracy.Two researchers independently appraised internal validity using SIGN checklists. Where consensus on quality rating was not reached, a third reviewer was involved.Effect sizes were calculated for controlled studies.A meta-analysis was not conducted due to heterogeneity of samples, interventions and outcomes. | **Included**: 90 unique publications identified, 22 were screened as eligible for appraisal (inter-rater agreement of 93.8%) and were critically appraised, and 13 met criteria for inclusion (91% inter-rater agreement).Five groups of interventions: jogging, horseback riding, martial arts, swimming or yoga/dance.Studies were mostly conducted in school or community settings, and included ages of middle childhood. There was a wide range of intervention frequency ranging from once to 4 times per week and total duration ranged 80 minutes to 56 hours.**Key findings*** Physical exercise interventions: can result in improvements to numerous behavioural outcomes including stereotypic behaviours, social-emotional functioning, and cognition and attention.
* Two RCTs evaluating martial arts reported large effect sizes on the impact on stereotypic behaviour (0.9) [58], and social-emotional behaviour (1.4) [57].
* There was a moderate effect of horse-back riding on socio-emotional outcomes; however this was non-significant for a study investigating the impact of this exercise modality on parent-child interactions and sensory profile.
 | **Author conclusions**: Exercise can be an effective behavioural intervention for children and youth with ASD for decreasing stereotypic behaviours and improving social-emotional functioning, cognition and attention. The greatest benefits appear to be for horseback riding and martial arts interventions, with more limited benefits for swimming and yoga/dance.**Reviewer’s comments**: Reasonably broad search strategy using explicit criteria, with eligibility determined independently by two researchers with excellent inter-rater agreement. SIGN checklists used for critical appraisal. Only 2 RCTs identified, both of which found significant improvements in outcomes (both were related to martial arts interventions) [57, 58].**Source of funding**: None**Included RCTs (intervention)**:* Bahrami et al (2012[58] – martial arts
* Movahedi et al (2013) [57] – martial arts.
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| **Lee et al (2016) [54]** |
| **Country, study type, aim** | **Search strategy** | **Appraisal methods** | **Results** | **Conclusions** |
| **Country:** Canada**Study type**: systematic review**Evidence level**: I**Study Quality** (SIGN checklist): – (low quality)**Aim**: As part of a wider review of movement-based interventions (MBI), included a section on the efficacy physical activity interventions targeting psychosocial outcomes for children with ASD. MBI defined as intentional full body movement of the child through space in which the movement is child-directed, proposed by an adult, or guided, but not imposed. | **Databases**: Embase, Ovid MEDLINE, CINAHL, and PsychINFO.**Search**: Searched from database commencement to November 2013, updated to September 2015. Search terms not provided. Broad range of keywords relating to population, intervention, age, outcomes. Supplemental searching included snowballing technique to check references of selected studies.**Selection criteria**: studies with children with a diagnosis of autism; considered interventions where movement was the core attribute; targeted psychosocial outcomes regardless of methodology.Excluded: studies where cognitive and/or behavioural components were the focus of the intervention; studies not meeting quality appraisal criteria on the MMAT; syntheses or reviews of existing evidence; theoretical and empirical articles; grey literature; editorials. | **Method:** The first author “consulted with healthcare researchers”.Single researcher extracted data using a charting form.Appraised internal validity using Mixed Methods Appraisal Tool (MMAT). Excluded 2 studies not meeting quality criteria (criteria not defined).Results were synthesised using a descriptive numerical and thematic analysis. | **Included**: 7260 unique publications identified, and 14 met criteria for inclusion, only 2 of which related to physical activity and the current review, reported below. (Other MBI included role play, imitation, Ayres sensory integration).**Key findings*** One theme emerged was types of movement-based interventions, which included physical activity.
* Two studies identified which considered physical activity interventions, a cohort study of water exercise [71] and an RCT evaluating martial arts (karate kata) [57].
 | **Author conclusions**: there is emerging evidence on the use of MBIs with children that targets psychosocial outcomes.**Reviewer’s comments**: Reasonably broad search strategy though search terms not provided. One researcher conducted review, using a quality checklist for critical appraisal but no critique provided and unclear how it was used to exclude two studies. Only 2 studies including one RCT identified relevant to the current review [57] with brief overview of each.**Source of funding**: Richard and Edith Strauss Canada Foundation and by the Projet Strat’gique innovant du CRIR: The Rehabilitation Living Lab in a Mall.**Included RCTs (intervention)** relevant to the current review:* Movahedi et al (2013) [57] – martial arts.
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| **Scottish Intercollegiate Guidelines Network (2016) [52]** |
| **Country, study type, aim** | **Search strategy** | **Appraisal methods** | **Results** | **Conclusions** |
| **Country:** Scotland**Study type**: systematic review/guideline**Evidence level**: I**Study Quality** (SIGN checklist): ++ (high quality)**Review scope\***: part of a review up date of a Clinical Guideline on assessment, diagnosis and interventions for autism spectrum disorders.**Aim\***: to identify signs and symptoms for identifying adults for assessment. | **Databases**: Medline, Embase, Cinahl, PsycINFO, Cochrane Library.**Search terms**: Searched from 2006 and 2014 (as an update to a previous guideline). Transparent selection criteria used including “physical exercise” as a search term. Database searches supplemented by material provided by individual members of the Guideline development group. | **Method:** Each included paper was evaluated by two members of the group using SIGN methodological checklists. | **Included**: Identified a meta-analysis [45] of small trials of various physical activities concluded that exercise for individuals with ASD may be beneficial for improving motor skills and social interaction. Further, larger, more rigorous trials are needed to support the findings (See Section 6.5, p. 27). | **Author conclusions**: Further, larger, more rigorous trials are needed to support the findings that physical exercise may improve motor skills and social interaction for individuals with ASD.The Guideline recommends further research with respect to “more RCTs on physical exercise interventions in ASD” (Section 13.2, p. 51).**Reviewer’s comments**: Covers the key databases using explicit criteria, and appraised by two researchers using methodological checklists. Research topic peripheral to the current review.**Source of funding**: National Health Service Scotland, through Healthcare Improvement Scotland.* **Key evidence:** Meta-analysis of Sowa et al (2012) [45] which includes RCT by Bass et al (2009) [56] – horse riding.
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| **Tan et al (2016) [48]** |
| **Country, study type, aim** | **Search strategy** | **Appraisal methods** | **Results** | **Conclusions** |
| **Country:** Australia**Study type**: systematic review and meta-analysis**Evidence level**: I**Study Quality** (SIGN checklist): +(acceptable quality)**Aim**: Review efficacy of physical exercise interventions on improving cognitive functions of individuals with ASD (and/or ADHD, results for whom are not reported here). | **Databases**: ERIC, CINAHL, PubMED, Web of Science, and PsychINFO.**Search**: Searched from 1968 to January to June 2015. Transparent selection criteria. Broad range of keywords relating to population, intervention, and outcomes. Supplemental searching included hand-searching of references of retrieved papers, forward searches using Google scholar of articles that cited included articles, hand-searching of relevant Journals excluded from databases.**Selection criteria**: quantitative studies with at least one participant with a diagnosis of ASD or ADHD; evaluated the effectiveness of exercise interventions; included one or more outcomes measuring objective cognitive performance (ie, excluding self-report measures); published in Journals or dissertations.Excluded: abstracts | **Method:** Authors independently assessed titles and abstracts for relevance and resolved disagreements about eligibility through discussion and consensus.Effect sizes were calculated for controlled studies.A meta-analysis was conducted based on a random-effects model of data. Multiple regression model used to examine potential variables that account for efficacy of exercise on cognition. Sensitivity analyses conducted to evaluate publication bias. | **Included**: 32,767 titles identified, 22 met criteria for inclusion, 6 of which applied to participants with ASD (n=128, *n* range 4–76, *age* range 3–25 years) – only these will be reported here.Evaluated exercises included: jogging (n=3), exergaming (n=1), mixed exercises (n=1), one tricycling (n=1). Study design was not explicitly reported but 5 studies appeared to be single case experimental studies and one was an RCT [60].Five of the ASD studies evaluated on-task duration or simple learning tasks. One examined executive functions.**Key findings*** Mean effect size for 6 ASD studies of 0.471 95% CI .313–.603; p<0.01)
* There was a moderate effect of r=0.526 for the impact of exercise on on-task duration/simple tasks in individuals with ASD from 5 studies. Using binomial effect size display, reported that this translated to 76.3% of participants with ASD achieving benefits from exercise in these 5 studies.
* In multiple regression modelling, diagnosis (ASD or ADHD) was not a significant moderator of the relationship between exercise and cognition outcomes. Neither were sample size, age group, type of control, type of exercise, duration of exercise.
 | **Author conclusions**: Results support the efficacy of exercise on improving some aspects of cognition in young individuals with ASD.**Reviewer’s comments**: Reasonably broad search strategy using explicit criteria and supplemental searching. Inter-rater agreement not reported. No appraisal checklists reported. Study design selection criteria not clearly described but assumed to be controlled studies. Publication bias investigated. Meta-analysis with sensitivity analyses reported. Only 6 studies relevant to ASD, including one RCT.**Source of funding**: None reported**Included RCTs (intervention)**:* Tan et al (2013) [60] – tri-cycling
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| **Dillon et al (2017) [53]** |
| **Country, study type, aim** | **Search strategy** | **Appraisal methods** | **Results** | **Conclusions** |
| **Country:** USA**Study type**: systematic review**Evidence level**: I**Study Quality** (SIGN checklist): – (low quality)**Aim**: Review the use of exercise as an evidence-based practice (EBP) for individuals with ASD aged 1–21 years. | **Databases**: ERIC, SPORTDiscus, ProQuest Nursing, Science Direct, Ovid MEDLINE, and PsychINFO.**Search**: Searched from January 2006 to April 2016. Transparent selection criteria. Narrow range of keywords relating to “exercise” and ASD terms. Supplemental searching included hand-searching of references of retrieved papers.**Selection criteria**: studies including participants diagnosed with ASD aged between 1 and 21 years; used an experimental or quasi-experimental, correlational, single-subject, or qualitative study design; evaluated an exercise intervention; published in English language Journals and periodicals.Excluded: studies of participants diagnosed with other/secondary disabilities; systematic reviews and meta-analyses; and abstracts, books, unpublished papers, conference proceedings, book chapters; | **Method:** Reviewers assessed titles and abstracts for relevance; articles meeting selection criteria were appraised by one author using a checklist and ratings were independently rated by a second researcher to confirm agreement of rating level as strong, moderate or weak (100% agreement).Used the Adapted Physical Activity Taxonomy (APAT) to extract data. | **Included**: 169 titles identified, 23 met criteria for inclusion (considering participants aged 4–27 years).Studies reported on fitness focused exercise (n=7), aquatics (n=3), karate and martial arts training (n=3), yoga (n=2), dance (n=2), equine-assisted programming (n=2), relaxation training (n=2), table tennis skills (n=1) and exergaming (n=1) interventions.Interventions were attempted to address the following issues: social and behavioural (n=8), repetitive and stereotypical behaviours (n=5), health-related fitness (n=4), skill development (n=4), cognitive functioning (n=2), and time-on-task (n=2).Study designs were experimental (including 4 RCTs) or quasi-experimental studies, correlational; and single-subject designs.**Key findings*** 13 of the 23 studies had weak methods sections, and 17 of the 23 had weak results sections on the APAT.
* There was one well-designed and well-implemented quasi-experimental study [71] which examined the effects of a water skills swimming programme on the aquatic skills and social behaviours of 6–9 year old children diagnosed with ASD.
 | **Author conclusions**: From one well conducted study [71] of moderately good overall quality, the authors recommended that an exercise can be considered an EBP for school-aged children with ASD.**Reviewer’s comments**: Reasonably broad search strategy including some supplemental searching. However only the search term “exercise” was used which may miss physical activities, sports, or other eligible interventions. Adapted checklist used to extract data. One would also question whether relaxation training can be considered exercise. Study designs were inconsistently reported. For example, when the original studies classified as experimental were examined for the current review, five were found to be quasi-experimental, one was a before-and-after study, one was single subject experimental study, and 3 were RCTs. The one study [71] highlighted as being of moderately good quality was not an RCT but was quasi-experimental.**Source of funding**: None reported.**Included RCTs (intervention)**:* Bahrami et al (2012) [58] – martial arts
* Movahedi et al (2013) [57] – martial arts
* Chan et al (2013) [59] – mind-body exercise
* Pan et al (2017) [61] – table tennis skills.
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| **Healy et al (2018) [31]** |
| **Country, study type, aim** | **Search strategy** | **Appraisal methods** | **Results** | **Conclusions** |
| **Country:** USA**Study type**: systematic review and meta-analysis**Evidence level**: I**Study Quality** (SIGN checklist): +(acceptable quality)**Aim**: to determine the effect of physical activity interventions on young individuals diagnosed with ASD. | **Databases**: MEDLINE, ERIC, PsycINFO, SPORTDiscus, Child Development and Adolescent Studies, ProQuest, and PsycARTICLES.**Search**: Searched from database 1970 to an unspecified period (no paper published in 2017 or later was included). Transparent selection criteria. Broad range of keywords relating to intervention, setting, and population. Supplemental searching not reported.**Selection criteria**: the study’s participants had ASD and were aged between 2 and 22 years; was implemented an intervention or method; took place in a physical education, physical activity or sport setting; included a quantifiable outcome measure; was published in English after the year 1970.Excluded: none reported | **Method:** Two authors shared a process of searching databases and identifying potentially eligible titles. Abstracts were then screened independently by both authors, and full texts retrieved for those potentially eligible. Selection criteria were then applied by two independent authors with any disagreements resolved in discussion with a third author. Lead authors of potentially eligible articles were contacted for further information where necessary to apply criteria.One reviewer extracted data. Two coders independently entered study characteristics using established protocols (but not using appraisal checklists) with a third coder considering discrepancies with simple majority agreement determining the final code (acceptable coder agreement of *κ*=0.84).Effect sizes (through Hedges *g*) were calculated. A meta-analysis was conducted using a random-effects model. Sensitivity analyses conducted to evaluate effect of outliers and publication bias. Moderator analyses were conducted to consider the influence of selected demographic and methodological characteristics on the relationship between physical activity interventions and outcomes grouped into 6 constructs: body composition, muscular strength/endurance, cardiovascular endurance, locomotor skills, manipulative skills, skill-related fitness, and social functioning. | **Included**: 29 studies with 30 independent samples that met criteria for inclusion in the meta-analysis (n=1009, *n* range 5–233; *age* range 5–15 years).Evaluated exercises included: horse riding/equine assisted (n=4 studies), kata martial arts techniques (n=2), aquatic skills (n=4), yoga (n=2), dance (n=1), cycling (n=1), treadmill walking (n=1), run/jog (n=1), trampolining (n=1), mind body programme (n=1), and exergaming (n=1).Other studies included a range of combined or varying physical activities, or were non-experimental, cross-sectional studies of observed physical activity and its association with outcomes.Intervention duration ranged from less than 10 weeks (*k*=8 studies), 10-16 weeks (*k*=11), >16 weeks (*k*=7), with 3 studies not reporting duration.17 studies were described as experimental (including 7 RCTs), and 12 were described as quasi-experimental (though some were actually observational studies).**Key findings:**Significant effects found for:* manipulative skills (*k*=3, *g*=2.76, p<0.001)
* locomotor skills (*k*=6, *g*=1.60, p<0.001)
* skill-related fitness (*k*=12, *g*=1.07, p<0.001)
* muscular strength/endurance (*k*=7, *g*=0.78, p<0.01)
* social functioning (*k*=6, *g*=0.57, p<0.05).

Non-significant effects were found for body composition (*g*=-0.18) and cardiovascular endurance (*g*=0.10)Subgroup moderator analyses were likely to be imprecise due to low study numbers and high heterogeneity. | **Author conclusions**: The meta-analysis revealed an overall moderate-positive effect (g=0.62) for participants exposed to physical activity interventions, particularly for interventions targeting the development of manipulative skills, locomotor skills, skill-related fitness, social functioning, and muscular strength/endurance.**Reviewer’s comments**: Broad search strategy using explicit criteria but supplemental searching. Two researchers involved in several review phases, but inter-rater reliability not reported for all aspects. Publication bias investigated. No quality checklists were used. Thorough analysis and description of results although whether studies were RCTs was not reported. Included lower-order non-experimental/observational studies, and studies where control groups were of typically developing people (ie, before-and-after single arm studies).Original experimental papers were checked for the current review to ascertain that 7 studies were RCTs.**Source of funding**: none reported**Included RCTs (intervention)**:* Bahrami et al (2012) [58] – martial arts
* Borgi et al (2016) [62] – horse riding
* Chan et al (2013) [59] – mind-body exercise
* Dickinson et al (2014) [64] – exergaming
* Favazza et al (2013) [65] – athletics
* Gabriels et al (2015) [63] – horse riding
* Movahedi et al (2013) [57] – martial arts.
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| **Fang et al (2019) [41]** |
| **Country, study type, aim** | **Search strategy** | **Appraisal methods** | **Results** | **Conclusions** |
| **Country:** USA**Study type**: systematic review**Evidence level**: I**Study Quality** (SIGN checklist): +(acceptable quality)**Aim**: Review efficacy of exergaming interventions on individuals with ASD | **Databases**: Google Scholar, PubMED, Web of Science, Research Autism (website), and PsychINFO.**Search**: Searched from 2000 to 2018. Transparent selection criteria. Narrow range of keywords relating to autism, exergaming or active video gaming, and cognitive function or cognition. Supplemental searching was not reported.**Selection criteria**: peer reviewed studies published in English; with participants diagnosed with ASD by a physician; evaluated the effectiveness of exergaming interventions; and included measures of cognitive performance, social-emotional behaviours, or physical ability.Excluded: abstracts, conference reports, duplicates, book chapters, not peer reviewed, not relevant. | **Method:** Two authors independently assessed titles and abstracts for relevance and resolved disagreements about eligibility through discussion and consensus.Two authors used Physiotherapy Evidence Database (PEDro) scale to evaluate study quality with a third author involved to resolve disagreements.Thematic coding was used to categorise outcomes. | **Included**: 1024 titles identified, 10 met criteria for inclusion (*n* range 5–100, age range 5–21 years).Study designs were pre- and post-test studies (n=6), case-control studies (n=3), and there was one RCT [64] which investigated use of the Nintendo Wii three times per week over a year.Eight studies reported on physical fitness, and 6 reported on cognitive performance.**Key findings*** In studies overall, there were significant improvements in most physical and cognitive functions, and increased perceived skill performance, though not actual improved motor skill performance, nor emotional regulation.
* In the RCT, the exergaming group had improvements in decreased body mass, enhanced performance in speed and agility, increased broad jump performance, strengthened aerobic activity, and increases in sit-up performance.
 | **Author conclusions**: Exergame interventions can be considered an effective addition to standard physical activity treatment on individuals with ASD, with significant improvements in physical fitness, executive function, and self-perceived motor skills performance. Games provided opportunities for moderate-to-vigorous physical activity. However, effects on emotional control and actual motor skill development were not significant.**Reviewer’s comments**: Reasonably limited search strategy restricted to cognition outcomes, and with no supplemental searching. Inter-rater agreement not reported. Appraisal checklist used. Only one RCT identified.**Source of funding**: None reported**Included RCTs (intervention)**:* Dickinson et al (2014) [64] – exergaming
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| **Howells et al (2019) [40]** |
| **Country, study type, aim** | **Search strategy** | **Appraisal methods** | **Results** | **Conclusions** |
| **Country:** Australia**Study type**: systematic review and meta-analysis**Evidence level**: I**Study Quality** (SIGN checklist): ++(high quality)**Aim**: Review the effects of group-based physical activity for social and communicative outcomes in children with ASD. | **Databases**: CINAHL, MEDLINE, PsycINFO, SPORTDiscus, Embase.**Search**: Searched from database commencement to 1st March 2018. Transparent selection criteria. Broad range of keywords relating to organised, intervention, and population. Supplemental searching included hand-searching of references of retrieved papers.**Selection criteria**: participants with a formal diagnosis of ASD with a mean age of between 5 and 12.9 years; sample size of at least 5; evaluated the effectiveness of organised physical activity (OPA) which involved structured training sessions supervised by a coach or adult delivered in small groups with at least one 30-minute session per week; studies with or without a comparison group (of alternative non-physical activity or everyday activity); included social functioning and/or communication outcomes; published in English**Excluded**: none reported | **Method:** Two authors independently assessed titles and abstracts for relevance, resolving disagreements (<1%) through discussion. One author applied selection criteria to retrieved full texts with final selection finalised by discussion with a second author. Reference lists of retrieved papers were also independently screened by two independent reviewers and again selection finalised through discussion of discrepancies to achieve consensus.One reviewer extracted data. Two reviewers conducted critical appraisal of study quality using an adapted QUIPS Tool with discrepancies discussed to achieve consensus.Effect sizes (through Hedges g) were calculated for social functioning and communication outcomes for controlled studies.A meta-analysis was conducted using a random-effects model. Meta regression was not possible due to low *k* (too few studies). Sensitivity analyses conducted to evaluate publication bias (determined as unlikely for social functioning, and possible for communication outcomes where trim and fill adjustment procedure used) | **Included**: 3,188 unique titles identified, 11 met criteria for inclusion in the review (n=379, *n* range 13–116; *age* range 3–16 years) and 7 for inclusion in the meta-analysis (6 for social functioning, 4 for communication).Evaluated exercises included: horse riding (n=6 studies), kata martial arts techniques (n=2), soccer skills programme (n=1), multi-sport camp programme (n=1), and an outdoor adventure camp programme (n=1). Intervention duration ranged from one week to 25 weeks and frequency ranged from one session per week to a 5-day camp (ie, total dosage ranged from 6.5 to 56 hours). Two studies included follow-up assessments [57, 58].Eight of 11 studies included controls, 4 of which were RCTs [57, 58, 62, 63]**Key findings*** 8 of 10 studies found significant improvements in some aspect of social functioning. Pooled statistical results indicated:
* significant improvement in social functioning; *k*=6 studies, *g*=0.45, CI(0.19-0.72), p=0.001.
* non-significant effect for communication; *k*=4 studies, *g*=0.13, CI(-0.12, 0.38), p=0.13.
 | **Author conclusions**: The meta-analysis found a significant small-medium improvement in overall social functioning, but not communication. This review provides evidence in support of group-based organised physical activity participation by school-aged children with ASD.**Reviewer’s comments**: Broad search strategy using explicit criteria and supplemental searching. Two researchers involved in several review phases, but inter-rater reliability not reported for all aspects. Appraisal checklists used but modified. Publication bias investigated. Thorough analysis and description of results. Only 4 studies were RCTs.**Source of funding**: Jonathon and Simone Wenig Scholarship, National Disability Insurance Agency, Moose Foundation.**Included RCTs (intervention)**:* Bahrami et al (2012) [58] – martial arts
* Borgi et al (2016) [62] – horse riding
* Gabriels et al (2015) [63] – horse riding
* Movahedi et al (2013) [57] – martial arts
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**Key:** ADHD=attention deficit hyperactivity disorder; AS=Asperger’s syndrome; ASD=Autism Spectrum Disorder; DSM-IV=Diagnostic and Statistical Manual of Mental Disorders, 4th edition; DSM-5=Diagnostic and Statistical Manual of Mental Disorders, 5th edition; IQ=intelligence quotient; MBI=movement-based interventions; MMAT=Mixed Methods Appraisal Tool; PDD=pervasive developmental disorder; QUIPS=Quality in Prognostic Studies Tool; RCT=randomised controlled trial; SIGN=Scottish Intercollegiate Guidelines Network; UK=United Kingdom; US=United States of America

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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. Publications investigating animal therapy can be viewed here: <http://www.researchautism.net/publications?start=0&filters%5Bsubject%5D=Animal-Assisted%20Activities%20and%20Therapies> [↑](#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)