e4p Evidence for Practice

Motor Considerations in Children & Youth with Autism Spectrum Disorder

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Introduction

This document contains a brief overview of information on motor considerations in children and youth with autism spectrum disorder (ASD). It is intended to provide clinicians working with this population with relevant background information on the: (1) presence and impact of motor impairments in children with ASD; (2) possible neuroanatomical/physiological causes of motor impairments; (3) recommended motor skill measurement tools; (4) effectiveness of motor interventions or other interventions affecting motor outcomes.

How was the literature review completed?

An electronic search was performed in November 2014 to identify systematic reviews, meta-analyses and guidelines focusing on motor considerations in children with ASD. Databases searched included: PubMed MEDLINE, CINAHL, EMBase PEDro, OT Seeker, Trip Database, Cochrane Database and Rehabilitation Reference Centre. Keywords used in the search included: 'Autism', 'Autistic Disorder', 'Motor', 'Motor Skills', 'Motor Skills Disorder', 'Physiotherapy', 'Physical Therapy', 'Occupational Therapy.' Where possible, relevant subject headings were combined with keywords to ensure search thoroughness. To be included, systematic reviews, meta-analyses and guidelines had to meet the following inclusion criteria: (1) adhere to basic Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA)[1] criteria or AGREE-II criteria for guidelines;[2] (2) be published between 2004 and 2014; and (3) pertain to the presence, impact, cause or measurement of motor impairments and/or motor interventions or interventions targeting motor impairments in 0-19 year-olds with a diagnosis of ASD. Twelve reviews met the inclusion criteria (see Figure 1). The AMSTAR[3,4] scale was used to rate the quality of all included systematic reviews and meta-analyses (see Tables 1,2 & Appendix III) while American Academy for Cerebral Palsy and Developmental Medicine levels of evidence[5] were assigned to intervention studies (see Table 2 and Appendix II). Two reviewers assigned both evidence levels and AMSTAR scores with consensus scores reported. Finally, the International Classification for Functioning, Disability and Health[6] (ICF) was used to describe intervention study outcomes.

What is autism spectrum disorder (ASD)?

The term **ASD** encompasses a broad range of symptom severity and subsequent impacts on function.[7] **Core features** according to the Diagnostic and Statistical Manual of Mental Disorders, 5th Edition (DSM-V)[8] include: (1) impairments in social communication; (2) repetitive behaviours; and (3) restricted interests.

ASD affects more than 1% of the population[9] with boys outnumbering girls 4:1.[10] **Causes** of autism are believed to be multi-factorial and include genetic, epigenetic, non-genetic as well as environmental factors.[7] **Diagnosis** is based on whether the child meets the accepted DSM-V[8] ASD criteria. Diagnosis would ideally occur as early as possible by considering possible red flags in social communication, language, play and visual or other sensory and motor skills at ages 12-18 months[7,11] to maximise benefits of intervention[12,13] while minimising costs to families and society.[14]

Are children and youth with ASD at risk for motor impairments?

Research has long postulated that there are differences in motor skills between individuals with and without ASD.[15] One overview of systematic reviews[16] and two systematic reviews[17,18] on 'motor activity' and 'motor coordination' in ASD met inclusion criteria for this review.

Miyahara (2013) published a moderate quality overview of systematic reviews and meta-analyses on movement differences, effect of movement-based interventions and underlying motor mechanisms in ASD.[16] This overview included 12 studies of which four pertained to the presence of motor differences and impairments.[16] Of these four studies, only two[17,18] met our inclusion criteria of adhering to PRISMA systematic review guidelines.[1]

Fournier et al. (2010) completed a moderate quality meta-analysis of differences in motor coordination between pre-schoolers, school-aged children and adults with and without ASD.[17] After analysing the 51 included studies comparing individuals with ASD to typically developing peers, the authors identified a large effect size (ES = 1.2) in motor coordination deficit in ASD based on a variety of assessments of motor coordination including movement/reaction time, movement accuracy/error, adaptation rate, gait velocity, excursion of centre of pressure, balance stability and standardized test scores such as the Bruininks-Oseretsky Test of Motor Proficiency (BOTMP), the Movement Assessment Battery for Children (MABC), the Peabody Developmental Motor Scales (PDMS), the Physical and Neurological Examination for Soft Signs (PANESS) and the Vineland Motor Standard Score.[17] When Miyahara considered the types of motor measures as a moderator variable and completed a post-hoc analysis solely of standard assessments of motor coordination in his overview of systematic reviews, the effect size for these five studies was even larger (ES=2.91; SE=0.581; $p<0.01$; $Z=5.01$; $I^2=93.48$; 95% CI=1.774-4.051) supporting the presence of differences in motor coordination between individuals with and without ASD.[17]

Williams et al. (2004) published a low quality meta-analysis on the difference in action imitation between 281 infants, pre-schoolers, school-aged children and adults with and without ASD.[18] Results of the pooled 17 studies indicated a significant difference between individuals with and without the diagnosis ($p<0.05$).[18]

In summary, although motor impairments are not a diagnostic core feature of ASD, research supports the presence of differences in motor skills and coordination (ICF dimensions of body function and activity) between children with and without ASD.

Why are impairments in motor functioning in children with ASD of concern?

It has been postulated that the presence of motor impairments may have implications in other important domains for children with ASD.[19] One low quality systematic review investigated the impact of motor development on social cognition and language. Leonard & Hill (2014) reviewed 43 studies of which 13 pertained to motor function differences in infants through school-aged children with ASD to evaluate differences in social cognition and language.[20] Although the authors report mixed results, the majority of studies reviewed identified a positive correlation between development of motor skills and development of social cognition and language in children with ASD.[20]

Is there a physiological or anatomical cause for motor impairments in children and youth with ASD?

Models of motor control and motor learning suggest that motor skill learning and performance are a product of multiple brain regions with particular importance of basal ganglia and cerebellum activity.[21-23] In his overview of systematic reviews and meta-analyses, Miyahara (2013) included four studies pertaining to the neurophysiological nature of ASD.[16] Of these, three met inclusion criteria.[24-26]

Philip et al. (2012) completed a moderate quality meta-analysis of three studies pertaining to motor tasks in individuals with ASD.[24] When asked to complete a button pressing task while in an MRI scanner, children and adults with ASD showed significantly different activity patterns than their typically developing peers in motor areas of the brain (cerebellum, basal ganglia and pre-central gyrus) as well as brain systems directed to attention (basal ganglia, inferior and superior parietal lobules.)[24] The authors postulated that hypo-activation in the left inferior parietal lobule and hyper-activation in the right inferior frontal gyrus may impact the capabilities of individuals with ASD with respect to observation and execution of model movements.[24]

Stanfield et al. (2008) completed a high quality meta-analysis of 46 studies involving more than 800 pre-schoolers, school-aged children and adults with ASD.[25] Studies included volumetric analysis of brain size using MRI. After adjusting for age and IQ, the cerebrum and cerebellum of individuals with ASD was larger than that of typically developing peers, while the corpus callosum was found to be smaller.[25] The authors postulated that this difference in cerebellar size and possible disorganised connection to the cerebrum might play a role in the movement disturbances seen in individuals with ASD.[25]

Nickl-Jockschat et al. (2012) completed a low quality meta-analysis of 16 studies analysing morphometric MRI studies in 277 school-aged children and adults with ASD and identified a link between disturbances in specific brain regions (left peri-central region, left putamen, right caudate, right parietal operculum) and sensory-motor impairment in ASD.[26]

Although these three meta-analyses suggest a link between neurophysiology and motor impairments, Miyahara suggests that none of the studies sufficiently account for the differences in movement in individuals with and without ASD to support causality.[16]

What tools can be used to measure motor skills in children and youth with ASD?

Although our search did not identify any systematic reviews on **motor skill measurement** in ASD, a Best Evidence Statement (BESt) published by the Cincinnati Children's Hospital Medical Centre (2009)[27] was identified as being the highest level of available evidence.

This evidence synthesis[27] recommended the use of standardized testing of motor skills only when indicated. Consideration should be given to the fact that standardized testing may not be well tolerated by individuals with ASD[28-30] and may result in an inaccurate reflection of the individual's ability.[31] Scores may be inflated for those who thrive in the highly structured standardized test setting while scores may be reduced for those who have difficulty with motivation, interest or understanding.[32] In addition, any modification to the test administration process would require careful interpretation of results.[27]

When motor skill testing is recommended, the following assessment tools can be administered depending on the child's age:[27]

Peabody Developmental Motor Scale, 2nd Edition (PDMS-2)

The PDMS-2 is a standardized test with six sub-tests assessing motor functioning in children 0 through 6 years of age.[33] A number of studies have reported the use of the PDMS-2 in children with ASD. [34-36] The PDMS-2 has been shown to have excellent test-retest and inter-rater reliability.[33,37] It also has excellent concurrent validity with the Mullen Scale of Early Learning: AGS Edition[33,37] as well as with the Bayley Scales of Infant Development, 2nd Edition (Bayley-II) age equivalents and locomotion subscale; however it demonstrates poor concurrent validity with Bayley-II standard scores.[36] Note: To be able to utilize this test, the child must have a level of cognition that allows them to understand test item instruction.[35,36]

Bruininks-Oseretsky Test of Motor Proficiency, 2nd Edition (BOT-2)

The BOT-2 is a standardized test with four motor composite areas (and a total of eight subtests) assessing fine and gross motor functioning in children aged 4 to 21 years.[38] The BOT-2 included children with a variety of disabilities in their normative sample, including children with ASD.[38] The BOT-2 has been shown to have excellent inter-rater reliability in seven of eight subtests and all four composite scores (>0.90.)[38,39] The remaining subtest (Fine Motor Precision) had inter-rater reliability of 0.84.[38,39] The test-retest reliability for Total Motor Composite is ≥ 0.80 while the other composite scores and related subtests were <0.80 except for the Strength and Agility composite which was >0.80.[38,39] Evidence of validity in test

content, internal structure, clinical group differences, and relationships with other motor skills tests is described in the BOT-2 manual.[40]

Which motor interventions or interventions affecting motor outcomes are considered effective in children and youth with ASD?

In his overview of systematic reviews, Miyahara identified four systematic reviews pertaining to motor interventions,[16] of which three[41-43] met inclusion criteria. In addition, two more recent systematic reviews were identified through the literature search.[44,45]

Exercise

Three systematic reviews,[41-43] all included in Miyahara's overview,[16] evaluated the effect of exercise on a variety of outcomes in the body function and activity dimensions of the ICF in children with ASD.

Petrus et al. (2008) completed a moderate quality systematic review of seven studies on the effects of exercise interventions on stereotypic behaviours in 25 preschool and school-aged children with ASD.[41] Included studies were small (n<6) and used case-series or case study designs (AACPDM Level of Evidence IV). Intervention consisted of hydrotherapy, jogging, walking and ball playing.[41] Reported intervention duration was of 6 to 20- minute intervals with unclear frequency and number of sessions.[41] Overall results suggest improvements in stereotypical behaviour after exercise, although because of the low level of evidence of included studies and small sample sizes, Miyahara concluded there is weak evidence to support the effectiveness of exercise in reducing these behaviours in children with ASD.[16]

Lang et al. (2010) published a moderate quality systematic review of 18 studies, some of which were included in the publication by Petrus et al.,[41] on the effects of exercise on behaviour, academics and physical fitness in 64 pre-school and school-aged children as well as adults with ASD.[42] As in Petrus et al.,[41] studies included small sample sizes and the majority of studies utilized a time-series analysis. Intervention included teaching exercise (e.g., jogging in the majority of cases) to participants with ASD by using modelling, physical guidance, verbal reinforcement and contingency management.[42] Intervention frequency and duration varied widely.[42] Improvements after exercise included decreased levels of stereotypy, aggression, off-task behaviour, and elopement with increases in on-task behaviours, academic responding and appropriate motor behaviours.[42]

Sowa and Meulenbroek (2012) published a low quality meta-analysis of 16 studies evaluating the effects of physical exercise on motor and social skills in 133 pre-school and school-aged children as well as adults with ASD.[43] Of these studies, seven evaluated individualised exercise programs while nine examined group programs.[43] Intervention consisted of jogging/walking, hippotherapy, general motor training, hydrotherapy and/or swimming, cycling, weight training and other leisure activities.[43] Reported intervention duration and frequency ranged from 20 to 90 minutes, one to three times per week, for two to 12 weeks.[43] All studies reported positive outcomes, with better outcomes in individualised versus group programs.[43] Results of this meta-analysis should, however, be cautiously interpreted as no control groups were used in the analysis and therefore confounding variables may be involved.[16]

Hydrotherapy

Mortimer et al. (2014) published a moderate quality systematic review of four studies evaluating the effects of Halliwick-based hydrotherapy on social interaction and behaviour in 44 preschool and school-aged children with ASD (National Health and Medical Research Council Grade of Recommendation=D; Poor.)[44] Intervention duration ranged from 60 to 90 minutes, two to three times per week, for a total of 10-14 weeks and all studies showed some improvement in social interactions or behaviours (including stereotypies) on various outcome measures (video analysis, School Social Behaviour Scale; Pediatric Quality of Life

Inventory, Computerized Evaluation Protocol of Interactions in Physical Education).[44] Positive outcomes should be interpreted with caution because included studies had low levels of evidence (two studies at level IV) or a moderate level of evidence but with a high risk of bias (one level III study with control group; one level III study without control group) in addition to small sample sizes.[44]

Treatment and Education of Autistic and Related Communication Handicapped Children (TEACHH)

Virues-Ortega et al. (2013) completed a moderate quality meta-analysis of 13 studies evaluating the effects of the TEACHH Method on perceptual and motor skills, behaviour, language and cognition in 172 preschool and school-aged children as well as adults with ASD.[45] The TEACHH program focuses on a close working relationship between caregivers and practitioners, adapts intervention to the individual and utilises structured teaching experiences.[46] Meta-analysis results of the Psychoeducational Profile - Revised scores from six studies (93 individuals total) suggest negligible to small magnitude effects on motor skills. Duration, intensity or location of intervention did not moderate effects of the intervention.[45] Due to the limited number of studies, small sample sizes, lower levels of evidence and low quality of included studies, results should be interpreted with caution.

In summary, all three reviews on exercise report positive outcomes from exercise-based interventions in individuals with ASD in the body function and activity dimensions of the ICF.[41-43] Limitations with these results include the lack of understanding of the extent that psychosocial factors played in the motor intervention.[16] In addition, hydrotherapy may positively influence a variety of outcomes in the body function, activity and quality of life domains of the ICF[44] while the TEACHH method only demonstrated a negligible impact at best on the activity domain.[45] Outcomes for all three types of intervention should be interpreted with caution due to the low levels of evidence employed by the majority of studies included in the reviews.

Summary

Although motor impairments are not considered to be a diagnostic core feature of children with ASD, research confirms the presence of motor impairments in this population and their influence on other areas of functioning. Standardised measures to assess these motor skills have been developed and should be used judiciously. In addition, research regarding motor interventions and interventions affecting motor domains is beginning to emerge, and indicates promising positive outcomes in a variety of motor outcomes as well as other elements of the body function and activity dimensions of the ICF. Future rigorous research in this population is warranted to help guide future intervention and help optimise functioning in children with ASD not only at the body function and activity levels but ultimately to improve outcomes at the participation level.

Support for researcher time to conduct this review was provided by the Sunny Hill Health Centre for Children Foundation. The author would like to acknowledge Dr. Susan Harris, Andrea Ryce, Stephanie Glegg, Janice Evans and Lori Roxborough for their assistance.

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Table 1: Motor Function and Physiology/Neuroanatomy Systematic Reviews & Meta-Analyses Characteristics

	Overview of SRs/MAs in All Domains Relating to Motor Function	Motor Function SRs/MAs			Physiology/Neuroanatomy SRs/MAs		
		SRs/MAs Included in Miyahara (2013) Overview of SRs		SRs/MAs Published Since Miyahara (2013)	SRs/MAs Included in Miyahara (2013) Overview of SRs		
First Author	**Miyahara[16]**	**Fournier[17]**	**Williams[18]**	**Leonard[20]**	**Nickl-Jockschat[26]**	**Philip[24]**	**Stanfield[25]**
Type of Review	Overview	MA	SR	SR	MA	MA	MA
Topic of Review	Motor function Physiology/ Neuroanatomy Intervention	Motor function differences	Motor function Differences	Motor function Differences	Structural differences	Brain activation differences	Structural differences
Outcome of Interest	All	Motor coordination	Action imitation	Social Cognition Language	Structural changes	Brain activation	Brain volume
ICF Dimension	Body function Activity Quality of life	Body function Activity	Activity	Activity	Body function	Body function	Body function
Number of Included Studies (n)	12	51	21	43	16	3*	46
Design of Included Studies	SR & MA	Comparative (TD)	Comparative	Comparative	Comparative	Comparative (TD)	Comparative (TD)
Dx of Participants	-	ASD	ASD	ASD DCD SLI	ASD	ASD	ASD
Age of Participants	-	Preschool School-age Adult	Infant Preschool School-age Adult	Infant Preschool School-age	School-age Adult	Preschool School-age Adult	Preschool School-age Adult
Total Number of Participants	-	CA	281 ASD CA TD	CA	580 277 ASD 303 TD	CA	> 1600 > 800 ASD > 800 TD
AMSTAR Score[3]	5 - Moderate	7 - Moderate	3 - Low	3 - Low	3 - Low	5 - Moderate	8 – High

Abbreviations - ASD: autism spectrum disorder; CA: can't answer; DCD: developmental coordination disorder; ICF: International Classification of Functioning, Disability and Health; SLI: specific language impairment; MA: meta-analysis; SR: systematic review; TD: typically developing.

AMSTAR Quality Rating:[4] High Quality: 8 to 11; Moderate Quality: 4 to 7; Low Quality: 0 to 3

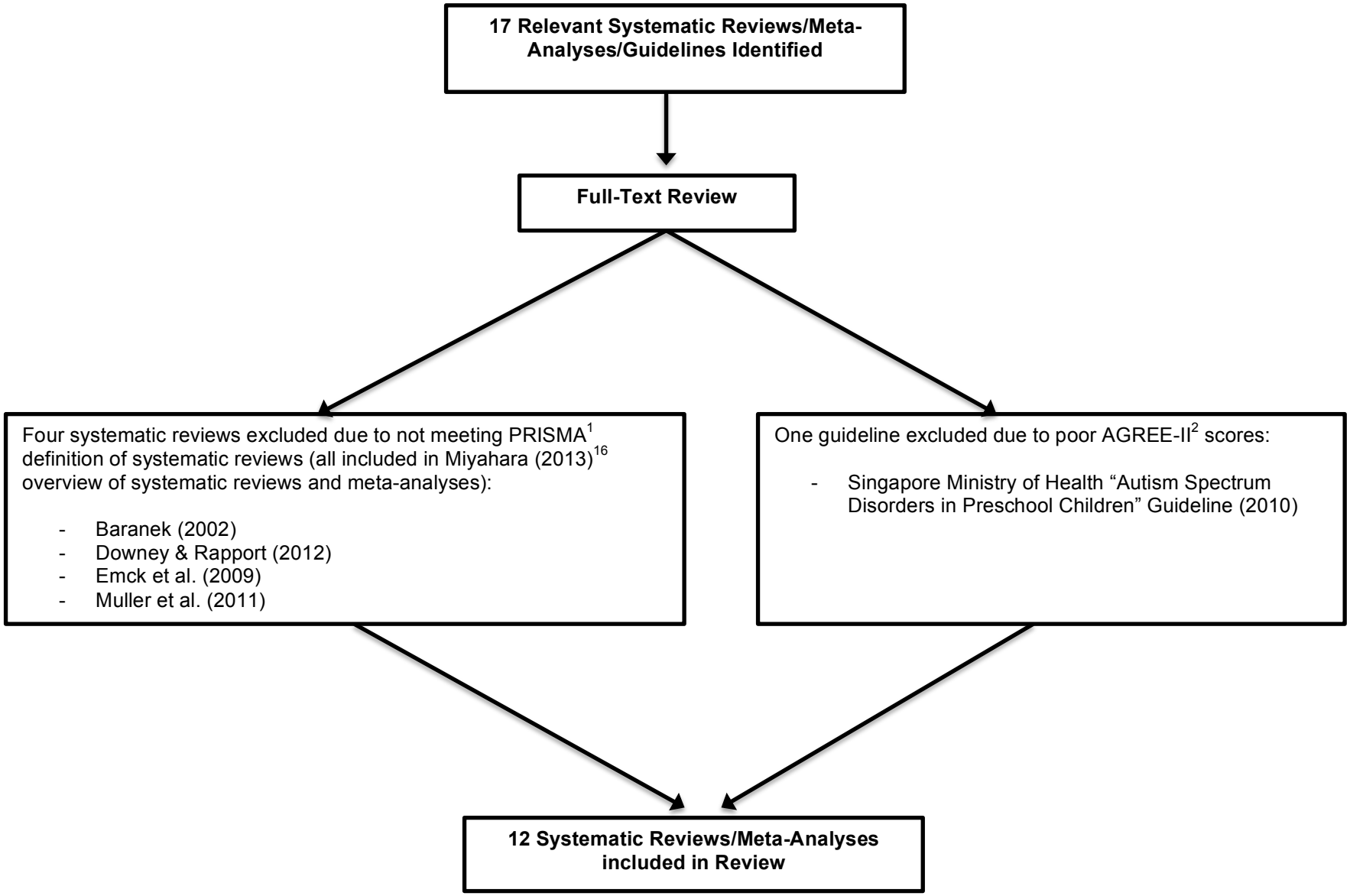
Table 2: Intervention Systematic Reviews & Meta-Analyses Characteristics

	Intervention SRs/MAs Relating to Motor Function				
	SRs/MAs Included in Miyahara (2013) Overview of SRs			SRs/MAs Published Since Miyahara (2013)	
First Author	**Lang[42]**	**Petrus[41]**	**Sowa[43]**	**Mortimer[44]**	**Virues-Ortega[45]**
Type of Review	SR	SR	MA	SR	MA
AACPDM Level of Evidence[5]	III	III	III	III	III
Topic of Review	Treatment Effect of Exercise	Treatment Effect of Exercise	Treatment Effect of Exercise	Treatment Effect of Hydrotherapy	Treatment Effect of TEACHH Method
Outcomes of Interest	Behaviour Academics Physical fitness	Stereotypic behaviours	Motor skills Social skills	Social interaction Behaviour	Perceptual & motor skills Adaptive behaviours Language & cognition,
ICF Dimension	Body function Activity	Body function	Activity	Body function Activity Quality of life	Activity
Number of Included Studies (n)	18	7	16	4	13
Design of Included Studies	Case Series	Case Series	Case Series	Case Series	Case Series
Participant Diagnosis	ASD	ASD	ASD	ASD	ASD
Age of Participants	Preschool School-age Adults	Preschool School-age	Preschool School-age Adults	Preschool School-age	Preschool School-age Adult
Total Number of Participants	64 ASD	25 ASD	133 ASD	44 ASD	172 ASD
AMSTAR Score[3]	4 - Moderate	6 - Moderate	3 - Moderate	6 - Moderate	7 - Moderate

Abbreviations - ASD: autism spectrum disorder; ICF: International Classification of Functioning, Disability and Health; MA: meta-analysis; SR: systematic review.

AMSTAR Quality Rating:[4] High Quality: 8 to 11; Moderate Quality: 4 to 7; Low Quality: 0 to 3

Appendix I. Systematic Reviews/Meta-Analyses/Guidelines Excluded from Synthesis



Appendix II: AACPDM - Levels of Evidence for Group Intervention Studies (December 2008)[5]

Level	Group Intervention Studies
I	Systematic review of randomized controlled trials (RCTs)
	Large RCT (with narrow confidence intervals) (n>100)
II	Smaller RCTs (with wider confidence intervals) (n<100)
	Systematic reviews of cohort studies
	"Outcomes research" (very large ecologic studies)
III	Cohort studies (must have concurrent control group)
	Systematic reviews of case control studies
IV	Case series
	Cohort study without concurrent control group (e.g. with historical control group)
	Case-control study
V	Expert opinion
	Case study or report
	Bench research
	Expert opinion based on theory or physiologic research
	Common sense/anecdotes

AACPDM: American Academy for Cerebral Palsy and Developmental Medicine.

Appendix III: AMSTAR Scores[2] for Included Reviews*

	All Motor Dimension SRs	Motor Functioning SRs/MAs			Physiology/Neuroanatomy SRs/MAs			Intervention SRs/MAs				
		SRs/MAs Included in Miyahara (2013)		SRs/MAs ≥ 2013	SRs/MAs Included in Miyahara (2013)			SRs/MAs Included in Miyahara (2013)			SRs/MAs Published ≥ 2013	
Item Number	**Miyahara[16]**	**Fournier[17]**	**Williams[18]**	**Leonard[20]**	**Nickl-Jockschat[26]**	**Philip[24]**	**Stanfield[25]**	**Lang[42]**	**Petrus[41]**	**Sowa[43]**	**Mortimer[44]**	**Virues-Ortega[45]**
1. Was an a priori design provided?	Y	Y (CA)	Y (CA)	Y	Y (CA)	Y (CA)	Y (CA)	Y (CA)	Y (CA)	Y (CA)	Y	Y
2. Was there duplicate study selection and data extraction?	CA	CA (Y)	CA	CA	CA	CA	Y	Y	CA (Y)	CA	Y	CA
3. Was a comprehensive literature search performed?	Y	Y	Y	Y	N	Y	Y	Y	Y	N	Y	Y
4. Was the status of publication (i.e., grey literature) used as an inclusion criterion?	N	Y	N	N	N	Y	Y	N	Y	CA	N	Y
5. Was a list of studies (included and excluded) provided?	N	Y	N	N	N	N	N	N	N	N	N	N
6. Were the characteristics of the included studies provided?	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	N
7. Was the scientific quality of the included studies assessed and documented?	Y	Y	N	N (Y)	CA	N	Y (NA)	N (Y)	Y	CA	Y	Y
8. Was the scientific quality of the included studies used appropriate in formulating conclusions?	Y	CA	N	N	CA	N	CA	N (Y)	Y	CA	Y	Y
9. Were the methods used to combine the findings of studies appropriate?	NA	N	NA	NA	Y	Y	Y	NA	NA	N	NA	Y
10. Was the likelihood of publication bias assessed?	NA	Y	NA	NA	N	N	Y	NA	NA	Y	NA	Y
11. Was the conflict of interest included?	N	CA	N	N	N	CA	CA	N	N	N	N	N
Total AMSTAR Score	5	7	3 (4)	3	3	5 (4)	8 (6)	4 (5)	6	3 (2)	6	7
AMSTAR Quality Rating	Mod	Mod	Low (Mod)	Low	Low	Mod	High (Mod)	Mod	Mod	Low	Mod	Mod

CA: Can't Answer; Mod: Moderate; N: No; NA: Not Applicable; Y: Yes,
*Parentheses indicate AMSTAR scores assigned by Miyahara, 2013[16] that are discrepant from this author's scores.

AMSTAR Quality Rating:[3] High Quality: 8 to 11; Moderate Quality: 4 to 7; Low Quality: 0 to 3