

Physiotherapy interventions to improve gross motor skills in
people with an intellectual disability aged six years and older:
a systematic review.

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Abstract

Intellectual disability is a life-long condition occurring during the early developmental years, resulting in impaired learning ability, reduced adaptive behaviour skills, and decreased functional independence. It affects approximately one percent of the world's population, and affected individuals have poorer health outcomes. People with an intellectual disability may benefit from specific teaching and learning approaches in therapy interventions which accommodate their cognitive and behavioural needs.

Gross motor skills (GMSs) are larger movements of the body, such as standing and walking, which are typically attained before the age of six. Deficits in GMSs may occur due to congenital conditions, such as cerebral palsy or Down syndrome, in which there occurs altered neuromuscular coordination and tone. GMS deficits can negatively affect a person's functional independence.

People with an intellectual disability who also suffer from GMS deficits can benefit from physiotherapy interventions to help improve their GMSs. Previous research has reported improvements in walking and balance for this population. Much research has supported early intervention programmes for children aged under six years. There is a comparative lack of research for people with an intellectual disability aged older than this, and no prior systematic review. A systematic review would inform clinicians and consumers regarding identifying effective interventions.

The object of this thesis was to conduct a systematic review which investigated the effectiveness of physiotherapy interventions to improve GMSs in people with an intellectual disability aged six years and older. The data sources for identifying quantitative research were: PubMed, CINAHL, Embase and ProQuest. Reference lists of relevant identified papers were hand-searched. Papers published in English from 1-1-2008 to 22-10-14 were considered for inclusion. Types of eligible study designs were randomized controlled trial (RCT), pseudo-RCT, repeated measures, and case report.

Overall, 866 potential articles were identified, of which 42 were retrieved for full-text review, and seven were finally included. Critical appraisal was conducted by two reviewers independently using the Joanna Briggs Institute (JBI) appraisal checklists; no papers were excluded following critical appraisal. Data extraction was performed using JBI Meta Analysis of Statistics Assessment and Review Instrument (MAStARI) data extraction instruments.

High heterogeneity between the studies precluded meta-analysis of the results, and a narrative synthesis was completed instead. Two RCTs, two pseudo-RCTs, two repeated measures studies and one case report were included. Studies varied in regard to participants' intellectual disabilities, and also regarding the interventions used. All interventions were well tolerated with negligible adverse effects. Significant improvements were reported for: cadence and non-dimensionalized gait velocity following body-weight supported gait training; cadence following lower limb strengthening exercises; and for the Gross Motor Function Measure-88 measure following adapted Judo training. These results suggest that task-specific training may be useful. However, based on the critical appraisal the overall quality of evidence was low.

The systematic review found limited evidence supporting physiotherapy for improving GMSs in people with an intellectual disability. Further research is needed to validate the early significant findings identified in this review and to define effective physiotherapy approaches which meet the learning needs of people with an intellectual disability.

Declaration

I certify that this work contains no material which has been accepted for the award of any other degree or diploma in my name in any university or other tertiary institution and, to the best of my knowledge and belief, contains no material previously published or written by another person, except where due reference has been made in the text. In addition, I certify that no part of this work will, in the future, be used in a submission in my name for any other degree or diploma in any university or other tertiary institution without the prior approval of the University of Adelaide and where applicable, any partner institution responsible for the joint award of this degree.

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List of abbreviations

| Abbreviation | Meaning |
|--------------|--|
| ABS | Australian Bureau of Statistics |
| APA | Australian Physiotherapy Association |
| BMI | Body Mass Index |
| BOT-2 | Bruininks-Oseretsky Test of Motor Proficiency, 2nd edition |
| BWS | body-weight supported |
| CP | cerebral palsy |
| DMD | Duchenne Muscular Dystrophy |
| DS | Down syndrome |
| GMAE | Gross Motor Ability Estimator |
| GMFCS | Gross Motor Function Classification Scale |
| GMFM | Gross Motor Function Measure |
| GMS | gross motor skill |
| HEP | home exercise programme |
| ICF | International Classification of Functioning, Disability and Health |
| ID | intellectual disability |
| ITT | Intention to treat |
| JBI | The Joanna Briggs Institute |
| MAStARI | Meta-Analysis of Statistics Assessment and Review Instrument |
| MDC | minimum detectable change |
| MID | minimum important difference |
| MMSE | Mini-Mental State Examination |

| | |
|---------|---|
| MS | multiple sclerosis |
| N | No |
| N/A | not applicable |
| NDIS | National Disability Insurance Scheme |
| PBWSTT | partial body-weight supported treadmill training |
| PICO | Population, Intervention, Comparator, Outcome |
| PWS | Prader-Willi syndrome |
| QOL | quality of life |
| RCT | randomized controlled trial |
| SD | standard deviation |
| SR | systematic review |
| Ss | sample size |
| SWAPS | Supported Walker Ambulation Scale |
| OGS | Observational Gait Scale |
| UN CRPD | United Nations Charter on the Rights of Persons with Disabilities |
| WHO | World Health Organization |
| Y | Yes |

1 Introduction

1.1 Overview

Intellectual disability, as defined by the World Health Organization (WHO)'s International Classification of Diseases Working Group on the Classification of Intellectual Disabilities,⁽¹⁾ is diagnosed when a person has marked impairments in adaptive behaviour and learning, with onset occurring in the individual's developmental years. The impairments are life-long and result in lower independence in managing life activities.⁽¹⁾ Commonly, other areas of personal development may also be impaired in a person with an intellectual disability, including gross motor skill (GMS) impairments. Deficits in GMS development can result in further limitations in functional independence for a person with an intellectual disability.

Various physical impairments can result in GMS deficits in people with an intellectual disability: these include altered muscle tone such as quadriplegic or diplegic hypertonia, as occurs in central nervous system diseases such as cerebral palsy,^(2, 3) or generalised low muscle tone with concurrent decreased muscle strength and impaired balance, as occurs in various syndromes including Down Syndrome⁽⁴⁾ and Prader-Willi syndrome.^(5, 6) Conversely, people with an intellectual disability may also suffer a degree of delay in their GMS development⁽⁷⁾ without any overt neuro-muscular impairment – the gross motor delay is instead associated with difficulty in acquiring new skills, including GMSs, due to primary learning difficulties arising from the individual's intellectual disability.^(7, 8) Levels of physical activity may similarly be decreased in people with an intellectual disability⁽⁹⁾ due to reasons such as difficulty moving freely without the support of carer input or facilitative equipment.⁽¹⁰⁾

There is high clinical relevance for carefully appraising the impact of an intellectual disability and any GMS deficits upon an individual's ability to participate freely in life's activities, and for defining intervention goals for improving GMSs. The WHO International Classification of Functioning, Disability and Health (WHO ICF)⁽¹¹⁾ provides paradigms for appraising such needs; these paradigms can be utilized by clinicians including physiotherapists.

Physiotherapists are instrumental in assisting to improve a client's GMSs in order to help overcome functional disabilities and promote improved levels of physical activity. Physiotherapists may assist in the attainment and retention of improved GMSs in clients with GMS deficits arising from a developmental disability such as cerebral palsy⁽¹²⁾ or resulting from an acquired cause such as a cerebro-vascular accident.⁽¹³⁾ Physiotherapists may work as a sole professional, or within a multi-disciplinary team with other health professionals. A multi-disciplinary team approach is often more effective in meeting the needs of clients with complex disabilities resulting from intellectual and physical challenges.⁽¹⁴⁾

Multi-disciplinary team early intervention models of care are usually provided to children aged less than six years of age with any type of developmental disability.⁽¹⁵⁾ These programmes have been a health focus for approximately the last 25 years⁽¹⁶⁾ and afford medical practitioner oversight.⁽¹⁷⁾ Early intervention programmes have been identified by an international expert panel as being a key clinical input for improving the health outcomes of individuals with an intellectual disability.⁽¹⁸⁾ Similarly, a United Kingdom national consultative panel identified that focusing on improvements in mobility for children with neurological disabilities resulting in movement deficits and cognitive impairment is of paramount importance during the developmental years.⁽¹⁹⁾ In contrast to this, there is comparatively less focus on the provision of therapy interventions for persons aged six and older, including the provision of physiotherapy services delivered either as a stand-alone service or within multi-disciplinary team interventions;^(20, 21) this is more so particularly for individuals with severe disabilities.⁽²²⁾ This is despite the need to carefully plan the provision of health care services, including physiotherapy, for people with an intellectual disability throughout the lifespan.^(21, 23)

No previous systematic review was identified which investigated physiotherapy services for improving GMSs for people with an intellectual disability aged six years and older. Accordingly, the primary objectives of the current review were to identify the best available evidence regarding physiotherapy interventions to improve GMSs in people with an intellectual disability aged six years and older, and the

effectiveness of these interventions. The secondary objectives of this review were to identify physiotherapy interventions for improving levels of physical activity in this population, and the effectiveness of these interventions.

1.2 Gross motor skills (GMSs)

GMSs are larger movements of the body, many of which are attained during early lifespan development.⁽²⁴⁾ In particular, there are a number of fundamental GMSs that emerge before six years of age. These skills include head and neck control in lying, rolling skills, independent static and dynamic balance for sitting and standing, and various forms of locomotion including walking and running.⁽²⁴⁾ The typical attainment of GMSs contributes to the ability to perform daily activities at an age-appropriate level throughout life. For example, typical GMS development in infancy contributes to improved visual and cognitive engagement. Later, integration of mature GMSs enables safe and independent walking with reduced risk of falling throughout the adult years.^(25, 26) In contrast, deficits in mobility skills can result in a greater risk of falls and injury from falls.^(27, 28)

The attainment of more advanced GMSs, such as climbing, jumping, hopping and ball skills, usually occurs during the preschool and school years. More advanced GMSs support an individual's ability to participate in sport and varied recreational and vocational activities,^(29, 30) and thus to also improve their level of physical activity.

1.3 Physical activity

Physical activity relates to the non-sedentary activity performed by an individual.⁽³¹⁾ Physical activity can be measured in different ways, for example the time a person spends in non-sedentary versus sedentary activity,⁽³¹⁾ and the level of their general physical endurance⁽³²⁾

1.4 Defining GMS deficits

GMS deficits can be defined with reference to the WHO ICF model⁽³³⁾ (figure 1). According to this model, GMS deficits can be categorised as activity limitations, resulting from various factors which can be categorised under ICF domains of biomedical, psychosocial⁽³⁴⁾ and/or environmental⁽³⁵⁾ factors. Additionally, GMS deficits may restrict a person's level of participation in their life roles and responsibilities. It is important that the health practitioner holistically assesses a client's GMS needs, giving regard to the ICF model, to ensure that accurate and thorough clinical deductions are made.

1.4.1 GMS deficits and reduced Quality of Life (QOL)

Particular attention has been given to assessing clients' QOL in regard to their physical disabilities. In particular, GMS deficits have been reported to be associated with reduced QOL, and to result in having a negative impact upon family interactions and work.⁽³⁶⁾ Additionally, it has been found that an individual's perceptions of their physical impairments is a greater determinant of QOL than the actual severity of their physical impairments.⁽³⁷⁾ These findings illustrate the relationship between GMS deficits and psychosocial factors, and also the need to carefully assess GMSs primarily, and to appraise secondary issues contributing to, or arising from, the GMS deficits.

1.5 Physiotherapy

Physiotherapy clinical work aims to improve a person's physical movement, comfort and functioning utilising a range of physical interventions.⁽³⁸⁾ The profession of physiotherapy has its own unique and specific professional registration and ethical requirements,⁽³⁹⁾ and clinical paradigms and scopes of practice.⁽⁴⁰⁾ Physiotherapy management of a client incorporates a client-centred approach to intervention planning which includes clinical assessment and reassessment throughout the duration of the intervention.

Physiotherapists may work alone or within a multi-disciplinary team^(41, 42) alongside other professions such as occupational therapists, medical doctors and nurses. A physiotherapist may implement interventions directly, or delegate and supervise the intervention activity to another capable person; such persons may be family members or carers of the client, therapy assistants, or gym trainers.^(39, 40)

1.5.1 Physiotherapy clinical assessment

Physiotherapy clinical assessment comprises a subjective assessment (in which the client and their carers are interviewed), and an objective assessment (which includes assessment of physical processes and GMSs of the client). The WHO ICF (figure 1) provides relevant outcome domains which can be used to frame the clinical assessment process.⁽⁴³⁾ A client's GMSs can be directly assessed and re-assessed using recognised outcome measurement tools.^(44, 45) Thorough clinical assessment of GMSs can be enhanced by appraising the client's level of engagement in general physical activity such as sport and recreation.

1.5.2 Types of physiotherapy interventions

Physiotherapy interventions are targeted at improving specific physical impairments (such as weakness) or activity limitations (such as decreased balance or impaired gait). Examples of types of physiotherapy interventions for specific physical issues include improving muscle strength and function through exercise and rehabilitation^(46, 47) (including strengthening⁽⁴⁸⁾ and stretching exercises⁽⁴⁹⁾ and aquatic therapy),⁽⁵⁰⁾ training of specific GMSs⁽⁵¹⁾ including gait education,⁽⁵²⁾ and improving a client's balance strategies^(53, 54) which may help to decrease the risk of falls.⁽⁵⁵⁾ Exercises may also be prescribed to aid reduction in pain,^(56, 57) or recovery of physical function following surgery.⁽⁵⁸⁾

1.5.2.1 *Settings for physiotherapy interventions*

Settings for interventions can be varied: interventions can be provided in a clinical setting such as an acute hospital ward,⁽⁵⁹⁾ a rehabilitation hospital,^(60, 61) or an outpatient clinic,^(56, 62) or in a community setting,⁽⁶³⁾ such as a school,⁽⁶⁴⁾ a residential care facility,⁽⁶⁵⁾ or the client's home.^(66, 67)

1.5.2.2 *Optimising engagement in physiotherapy interventions*

It is important that a client's base-line cognitive profile and needs are considered when planning and providing physiotherapy interventions, so as to optimise the client's learning during therapy.⁽⁶⁸⁾ When learning is optimized, it is more likely the client will be able to recall, utilize and integrate the skills learnt into their daily life.⁽⁶⁸⁾

1.5.3 Physiotherapy to improve GMS deficits

Physiotherapy can assist people of various ages to attain improved GMSs, and also be able to regularly practice using these skills in familiar contexts. Such practice enhances the integration of the skills into daily life and also makes it more likely that they will be successfully used in less familiar settings when required.

1.5.4 Task specific practice

One clinical approach used for improving GMSs is incorporation of task specific practice; this involves practicing the GMS of interest (such as walking or standing independently) in its entirety or in its component parts.^(69, 70) This is in contrast to intervention approaches in which the therapy is designed to support physical impairments contributing to the GMS deficit^(71, 72) such as strengthening⁽⁴⁸⁾ and stretching exercises⁽⁴⁹⁾ (section 1.5.2).

1.6 Intellectual disability

Intellectual disability, or intellectual developmental disability, as defined by the WHO,⁽¹⁾ is diagnosed when a person has significant impairment in cognitive functioning and adaptive behaviour, with onset occurring in the person's developmental years.^(73, 74) People with an intellectual disability experience learning difficulties^(75, 76) which impair their ability to acquire new intellectual and physical skills, and to meet the demands of daily living.⁽⁷⁷⁾

The term 'intellectual disability' replaces earlier terminology such as 'mental retardation'.⁽⁷⁸⁾ This review will use the definition for intellectual disability provided by the WHO,⁽¹⁾ which has also been used in a previous meta-analysis of the prevalence of intellectual disability internationally.⁽⁷⁹⁾ This review will also use person-focused phrasing ('person/people with an intellectual disability') when describing a person affected by having an intellectual disability. This approach to phrasing is endorsed by the American Physical Therapy Association for their 'Physical Therapy' peer-reviewed journal,⁽⁸⁰⁾ and is in contrast to condition-focused phrasing (for example, 'intellectually disabled person').

Under the WHO ICF (figure 1), intellectual disability can be termed an impairment of body functions (intellectual capacity) resulting from a diagnosed disorder or disease. Causes for intellectual disability⁽⁸¹⁾ are many, and include genetic reasons (including Down syndrome⁽⁸²⁾), various syndromes (for example Prader-Willi syndrome⁽⁸³⁾), or conditions resulting from insult to the central nervous system occurring during the developmental years (such as cerebral palsy⁽⁸⁴⁾).

Specific clinical traits can be present with distinct diagnoses for intellectual disability, including the level of severity of intellectual disability and the types of physical impairments. For example, certain conditions are more likely to result in mild intellectual disability such as occurs in Prader-Willi syndrome,⁽⁸³⁾ a condition also characterised by hyperphagia, obesity, low muscle tone, decreased muscle strength, and balance impairments. Down syndrome similarly includes features of low muscle tone and decreased strength, but the severity of intellectual disability can vary between individuals.⁽⁸²⁾ Other developmental conditions may

or may not result in an intellectual disability, such as Autism Spectrum Disorder⁽⁸⁵⁾ and cerebral palsy.⁽⁸⁴⁾ In such conditions there is variability in the severity of intellectual disability when this does present. Interestingly, for cerebral palsy, intellectual function was found to correlate to levels of physical function in a large observational study of youth with cerebral palsy.⁽⁸⁶⁾

1.6.1 Prevalence of intellectual disability: internationally; nationally

A systematic review and meta-analysis conducted by the George Institute and the WHO which investigated the prevalence of intellectual disability internationally, reported the overall rate to be 10.37 per 1000. In this review, data was included specifically from studies with participants with an intellectual disability which had onset during the developmental years.⁽⁷⁹⁾ Meta-analysis was conducted, and, within this, four Australian studies were included, for which the prevalence ranged from 7.02 to 11.53 per 1000 persons.⁽⁷⁹⁾

An Australian Bureau of Statistics (ABS) report (2012) on the prevalence of long-term health conditions resulting in disability, found that of Australians with a disability (4.2 million (18.5% of total population)) 5.6% reported having intellectual and developmental disorders.⁽⁸⁷⁾ In a 2008 Australian Institute of Health and Welfare (AIHW) report, the prevalence in Australia of persons having an intellectual disability with or without another disability was determined as 3%, and for persons having an intellectual disability as their primary cause of disability the prevalence was found to be 0.8% of the Australian population.⁽⁸⁸⁾

1.6.1.1 *Prevalence of intellectual disability in Australian children*

In a 2012 ABS report of children and disability, it was reported that 8.8% of 5-14 year olds and 3.4% of 0-4 year olds had a disability.⁽⁸⁹⁾ Of the children with disabilities, the prevalence of those with an intellectual disability was 61% of the 5-14 year olds and 29% of the 0-4 year olds; the discrepancy here is attributable to lower rates of formal cognitive testing in younger children.⁽⁸⁹⁾ In a 2008 AIHW report, data specifically

for Australian children found almost 200, 000 children with intellectual disabilities were attending schooling in 2003, and of these more than half were attending special education facilities.⁽⁸⁸⁾

1.6.2 Accommodation and education support

Persons with an intellectual disability reside in a variety of home settings (within the family home at any age, and, as an adult, in the community living either independently or semi-independently) and in respite or permanent supported accommodation settings.⁽⁹⁰⁻⁹²⁾ Schooling for children and youth with an intellectual disability can be undertaken in main-stream schools with increased teaching support staff available to meet the child's learning needs, or in a specialised educational setting.^(93, 94)

1.6.3 Lower health status and reduced QOL

People with an intellectual disability suffer higher social vulnerability,⁽⁹⁵⁾ decreased social integration,^(96, 97) and poor self- efficacy and esteem.^(98, 99) Additionally, there is a higher prevalence of mental health issues in people with an intellectual disability:⁽¹⁰⁰⁾ intellectual disabilities with associated mental health issues have been found to contribute to a reduced QOL, and interruption to vocational pursuits and interpersonal relationships.⁽¹⁰¹⁾ A person with an intellectual disability may also have a lower level of self-perceived health status which can result in decreased QOL.⁽⁹⁷⁾ The importance attributed to the issue of reduced QOL in people with an intellectual disability is demonstrated by the development of the WHO QOL measure for people with disabilities including intellectual disability.⁽¹⁰²⁾

1.6.3.1 *Difficulty accessing appropriate health care*

People with an intellectual disability can experience decreased access to mainstream health care services,^(103, 104) including for individuals with an associated mental health disorder.⁽¹⁰⁵⁾ This can further compound the impact of the intellectual disability and any concurrent reduced QOL, thus further inhibiting the person's ability to cope with and adapt to the demands of daily living.⁽⁷⁷⁾ In order to overcome these

potential risks and inequalities, more focused planning of the delivery of health services to people with an intellectual disability is warranted.

1.7 International and national documentary support for improving levels of health and health care delivery for people with an intellectual disability

There has been international commitment to addressing discrepancies in health status and access to health services experienced by people with an intellectual disability with a key selection of reports and legislative changes published which support these goals. These international and national documents provide the overarching ethical paradigms and political contexts for this review's topic of physiotherapy to improve GMSs in people with an intellectual disability, and are discussed below.

In 2008 the United Nations Convention on the Rights of Persons with Disabilities (UN CRPD) was internationally ratified by member nations⁽¹⁰⁶⁾ including Australia.⁽⁸⁷⁾ The principles of this document have been reflected in Australian governmental policy with pivotal changes to disability funding occurring with the implementation of the National Disability Insurance Scheme (NDIS).⁽¹⁰⁷⁾ One of the key priorities of this scheme was to enable consumers to have increased autonomy in choosing which disability services to engage and to what extent.⁽¹⁰⁸⁾ The UN CRPD has also been reflected in South Australian legislation, with amendments to the Disability Services Act 1993 passed in 2013.⁽¹⁰⁹⁾ This Amendment Act applies to all registered disability services providers, including health care providers, and outlines requirements supporting the rights and protection of persons with disabilities, giving reference to the UN CRPD.⁽¹⁰⁹⁾ Other pivotal international documents which have significantly affected the perceptions and understanding of disability health issues include two United States Surgeon General reports.^(110, 111) Both of these reports made broad-reaching analysis of, and recommendations for overcoming, the discrepancies in health status in people with any type of disability, including having an intellectual disability. The first of these, a 2002 national consultation report entitled 'Closing the Gap' assessed the health discrepancies and needs of persons with an intellectual disability;^(110, 111) and a subsequent 2005 report appraised ways to improve

the health of persons with disabilities more generally.⁽¹¹²⁾

1.8 The World Health Organization's International Classification of Functioning, Disability and Health (WHO ICF)

The clinical paradigms relevant to the systematic review presented in this thesis, which support appropriate and relevant assessment and intervention planning, are provided by the WHO ICF, which was initially published in 2001.⁽¹¹⁾ The ICF (figure 1) provides a conceptual framework for understanding and appraising the multi-factorial influences impacting upon an individual's function, disability and health. The multiple concepts of the ICF are interdependent, having influence on and being impacted by, the other concepts within the framework.

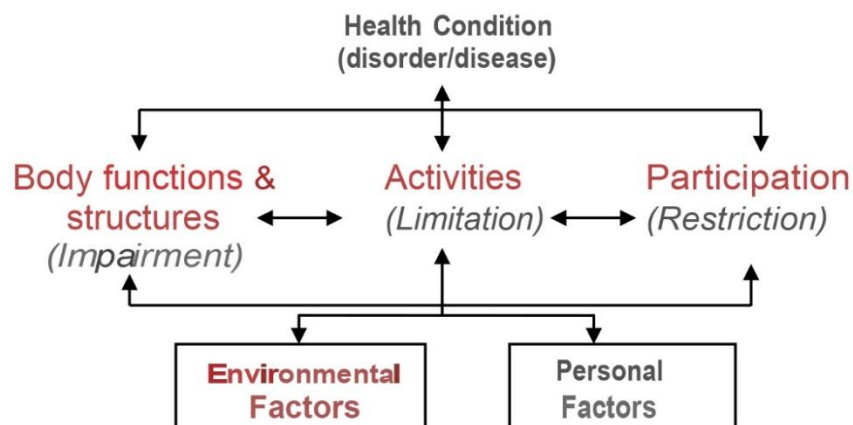
The ICF supports utilising a clinical approach of assessing more than just the diagnostic label of a client's condition, by situating the person's clinical diagnosis within a psycho-socio-environmental context. By doing so, this approach enables identification of fixed and/or modifiable factors that impact upon the individual's experience of their diagnosis, which may be ameliorated through various interventions.

Consideration of the ICF when assessing the needs of persons with an intellectual disability is particularly pertinent, and the goals and objectives of the current systematic review were developed with reference to the ICF paradigm.

Figure 1: The International Classification of Function, Disability and Health (ICF): interrelation of framework paradigms

Taken from http://sydney.edu.au/health-sciences/aidarrp/icf/multi_dimensional.shtml; ⁽¹¹³⁾ based upon WHO ICF Beginners Guide. ⁽¹¹³⁾

ICF: Interaction of Concepts



1.9 Use of the ICF when researching the needs of individuals with complex disabilities

The ICF has been previously used in primary research to investigate the needs of people with physical and cognitive disabilities. An overview report investigating the use of the ICF in primary research studies to appraise physical function in people with an intellectual disability found that the tripartite ICF domain, which is comprised of body structure and function, activity, and participation was more commonly utilized when assessing this population, rather than using these domains separately, and that there was variability in how the ICF term Activity was applied in studies.⁽¹¹⁴⁾ Similarly, in a systematic review investigating the use of the ICF in primary interventions studies for children with cerebral palsy, it was found that the ICF components of activity and also body structure and function were most commonly used.⁽¹¹⁵⁾

A specific disability assessment tool developed by the WHO which was developed based upon the WHO ICF - The WHO ICF Disability Assessment Schedule - has previously been used to assess functional abilities and deficits in people with complex disability resulting from central nervous system trauma (either

head injury or spinal cord injury).⁽¹¹⁶⁾ The results of this analysis highlighted personal deficits in the study participants related to either cognition or physical impairment or both.⁽¹¹⁶⁾ This study illustrates the usefulness of the ICF when appraising the complex needs of people with cognitive and physical impairments.

1.9.1 The ICF and how it relates to this review

For the purposes of the current review, the primary outcome of GMSs was considered under the ICF domain of activity, and GMS deficits were considered as a type of activity limitation. The secondary outcome of level of physical activity was considered under the ICF domain of participation; reduced levels of physical activity were considered a participation restriction (figure 1).

1.9.1.1 *Defining GMS deficits with respect to the ICF*

GMS deficits can be due to a variety of causes, such as muscle weakness or impaired central nervous system control (which can be grouped under the ICF domain of physical impairments) due to various health conditions (considered under disorders or diseases). Additionally, activity limitations arising from deficits in GMS such as walking, standing and general balance abilities, can restrict a person's functional independence⁽³³⁾ and, as per the ICF, create participation restrictions such as reduced ability to undertake usual daily activities or vocational pursuits. For the current review, studies which reported outcome data assessing GMS activity limitations were considered for selection.

1.9.1.2 *Relating physiotherapy assessments to the ICF*

Physiotherapy for improving GMSs in people with an intellectual disability should incorporate the assessment of potential clinical factors which may have contributed to any GMS deficits. Assessment may address the ICF domains of physical impairment (for example muscle weakness, or decreased muscle tone), activity limitation (such as reduction in gait speed, or impaired standing balance) and participation restriction (including reduced engagement in vocational activities or sport), as outlined in the WHO ICF

(figure 1).⁽³³⁾ A physiotherapy assessment should also incorporate other ICF domains concerning the individual's co-morbidities as well as personal and environmental factors.

1.9.1.3 *Relating physiotherapy interventions to the ICF*

Physiotherapy interventions designed to improve GMS deficits may focus on the ICF domains of activity (such as task-specific training interventions for example gait training) or on processes categorised within the domain of body structure and function (such as muscle strengthening, or balance exercises). For physiotherapy interventions that aim to improve levels of physical activity, these same ICF domains may be considered as well as the ICF domain of participation (for example, involvement in team sports).

1.10 **GMS deficits in people with an intellectual disability across the lifespan**

Specific GMS deficits, particularly physical mobility, have been identified in people with an intellectual disability of all ages, including infants, school-aged children and adults.

Impaired GMSs in early development have been found in infants with Down syndrome, including delay in acquiring independent standing.⁽¹¹⁷⁾ School-aged children with an intellectual disability, including a mild intellectual disability, have been shown to have deficits of balance and standing abilities.^(118, 119)

Additionally, eight-year-old children with Down syndrome have been found to have significantly lower gross motor function and lower health related QOL compared to age-matched normative results.⁽¹²⁰⁾

Adolescents and young adults with Down syndrome have been shown to have altered lower limb biomechanics; these physical limitations may be related to altered muscle co-ordination of agonist-antagonist muscle groups.⁽¹²¹⁾ Additionally, in youths with Down syndrome, balance impairments related to increased postural sway during weight-bearing activities have been reported.⁽¹²²⁾ Impaired balance has also been identified in youths with an intellectual disability aged up to 22 years, and has been attributed to impaired vestibulo-ocular responses.⁽¹²³⁾ For adults with an intellectual disability, a previous systematic

review reported there were limitations in general mobility and walking; however, this review found a lack of quality evidence for this finding.⁽¹²⁴⁾

There may be specific deficits in sensorimotor integration which are unique to some people with intellectual disability, and which may require careful clinical attention: for example, deeper proprioceptive sensation for improving static balance in individuals with Down syndrome.⁽¹²⁵⁾ Additionally, decreased ability to grade muscle co-contraction at the ankle in response to external perturbations has been reported in adults with an intellectual disability (compared to adults without an intellectual disability).⁽¹²⁶⁾ A greater degree of co-activation of the primary anterior and posterior ankle muscles was noted resulting in stiffer responses of the ankle in response to the perturbations. The authors of this study hypothesized that these were due to deficits in the integration of sensory and motor function, possibly occurring at the cerebellar level.⁽¹²⁶⁾

1.10.1 Obesity and GMS deficits

The influence of obesity upon GMS deficits in people with an intellectual disability has been previously investigated. Research investigating differences in gait patterns between obese people with Down syndrome, non-genetically obese participants, and healthy non-obese participants found that participants with Down syndrome and who were obese had the slowest gait and were the most restricted in terms of cadence and step length. The author postulated that this was due to reduced motor development resulting from obesity in early development.⁽¹²⁷⁾ A similar study comparing the balance abilities of participants with Prader-Willi syndrome to non-genetically obese participants as well as healthy controls found that balance was the most impaired in the Prader-Willi syndrome group.⁽¹²⁸⁾ The balance deficits were attributed to the individuals with Prader-Willi syndrome having small foot size, low muscle tone and strength, and obesity.⁽¹²⁸⁾ It has been noted that particular attention is needed to address ankle strength and function in adults with genetic obesity and low muscle tone as occurs in Down syndrome and Prader-Willi syndrome.^(121, 127, 128)

1.11 The impact of GMS deficits in decreasing functional independence for people with an intellectual disability

When a person with an intellectual disability has concurrent GMS deficits, there is likely to be further difficulties in participating in education, recreation and vocational employment beyond what could have been experienced if only an intellectual disability was present. This has been identified as an important clinical issue for people with an intellectual disability.⁽¹²⁴⁾

Optimising a person's ability to be independent within a community setting is a key goal for people with intellectual disability.⁽¹²⁹⁾ Improved motor function has been shown to result in increased levels of functional independence in adults with an intellectual disability living within a community setting, with a concurrent lower requirement for formal care supports.⁽¹²⁹⁾ Additionally, improved GMSs have also been shown to result in improved performance in activities of daily living for children with an intellectual disability.⁽¹³⁰⁾ Children with Down syndrome have progressed from a sedentary to a non-sedentary lifestyle following improvements in motor function.⁽³¹⁾ All of these findings indicate that there is clear benefit in seeking to overcome GMS deficits in people with an intellectual disability. Nonetheless, people with an intellectual disability may struggle to overcome GMS deficits due to difficulties with adaptive behaviours and learning new skills.⁽¹²⁴⁾ Specific therapies aimed at improving GMSs in people with an intellectual disability are indicated.

1.12 Physical activity in people with an intellectual disability

Physical activity has been investigated for people with an intellectual disability, with various positive benefits reported in response to being physically active. There is evidence to suggest that integrating improved levels of physical activity into daily life can improve adaptive skills for people with an intellectual disability.⁽¹³¹⁾ It has also been found that people with an intellectual disability experience improved health and well-being with increased levels of physical activity.⁽¹³²⁾ Optimisation of levels of physical activity where possible in this population is an imperative.

Different approaches for improving levels of physical activity in people with an intellectual disability have been proposed including modifying the person's general lifestyle activities, or, in contrast, prescribing a structured exercise programme.⁽¹³³⁾ Issues affecting compliance, however, need to be considered in order to optimise sustained uptake of an exercise programme.⁽¹³³⁾

There is clinical relevance in accurately measuring physical activity, for example walking, in people with intellectual disability who are vulnerable to losing their physical endurance. This is particularly relevant for people with Rett Syndrome⁽¹³⁴⁾ - a neurodegenerative disorder characterized by typical early growth followed by slowing of development.

1.12.1 Physical activity and GMSs in people with an intellectual disability

A relationship between general GMS development and levels of physical activity has been reported for individuals with an intellectual disability. Children with an intellectual disability who have higher GMS abilities, in particular with object control, have been found to show greater participation in sport.⁽¹³⁵⁾ Improvements in general motor proficiency and maintaining GMSs have been found to relate to increased levels of physical activity in people with Rett syndrome.^(134, 136) Levels of physical activity have also been shown to improve in children with Down syndrome following bicycle skill training using a specific modified bicycle.⁽³¹⁾

1.13 Assessment of GMSs in people with an intellectual disability

Therapy interventions should be based upon ongoing clinical assessment and review; the careful assessment of clients is essential to ensuring the provision of safe and effective interventions. This is particularly true for clients with an intellectual disability who may have greater difficulty providing feedback of their experience of any intervention due to their increased likelihood of experiencing difficulties with communication⁽¹³⁷⁾ and adaptive behaviour.⁽¹⁾ Accordingly, physiotherapists should give careful attention to any indication of pain or distress made by a client with an intellectual disability.⁽¹³⁸⁾ There is also

clinical indication to use assessment tools which are validated for the specific clinical diagnosis and severity of intellectual disability of the client. The use of such tools helps to minimise the potential confounding impact of the client's cognitive difficulties on their ability to successfully complete the assessment process.

1.13.1 GMS outcome assessments for people with an intellectual disability

Outcome assessment tools have been developed which accommodate clients' needs relating to having an intellectual disability and impairments in adaptive functioning. Previous research has shown that GMS outcome assessment can be reliably undertaken for adults with varied levels of intellectual disability including mild to moderate levels,⁽²⁷⁾ and severe levels,⁽¹³⁹⁾ and for specific diagnoses including Down syndrome.^(140, 141) Additionally, the well-known 'modified Berg Balance Scale' has been tested for feasibility and reliability for adults with profound intellectual disability and sensory impairments.⁽¹⁴²⁾ Other tools include outcome assessments specifically for individuals with an intellectual disability and a recent history of falling,⁽¹⁴³⁾ and for measuring general GMSs⁽⁷³⁾ and balance.⁽¹⁴⁴⁾

1.14 Therapy interventions for improving GMSs in people with an intellectual disability

A person's ability to acquire improved GMSs involves cognitive skills of learning and behavioural adaptation, both of which are more difficult for individuals with an intellectual disability. Accordingly, for this population, usual teaching and learning approaches used in therapy interventions may need to be modified in order to optimise efficacy. For example, novel approaches which incorporate unique and specific assessments and interventions could be helpful. However, therapies for this client group should be regularly reviewed and researched to avoid the use of unsubstantiated therapies.⁽¹⁴⁵⁾

Various types of interventions prescribed by a range of health and exercise professionals have been reported for overcoming GMS deficits in people with an intellectual disability. Two physical education studies in the field of physical education have been previously reported: improvements were noted in

running, walking and obstacle course negotiation for adolescents with an intellectual disability following the use of fitness training machines;⁽¹⁴⁶⁾ and improvements in balance were found following training with rehabilitation balls and varied weight-bearing surfaces.⁽¹²⁵⁾

Modification to the usual approaches for using therapeutic treadmill training equipment may better meet specific GMS learning needs of people with an intellectual disability. Treadmill training combined with the use of supportive supra-malleolar orthoses has been found to be beneficial for facilitating gait in toddlers with Down syndrome.⁽¹⁴⁷⁾ As well, treadmill training conducted with inclination of the treadmill walking surface has been found to optimise gait biomechanics in people with Down syndrome.⁽¹⁴⁸⁾ Previous systematic reviews of partial body-weight supported treadmill training (PBWSTT) has been found to be effective in improving gait in children and youth with motor impairments, with and without intellectual disability;⁽¹⁴⁹⁾ and a later review reported evidence supporting the use of PBWSTT in children with Down syndrome.⁽¹⁵⁰⁾

Due to many people with an intellectual disability experiencing difficulty accessing and/or engaging with mainstream physiotherapy, it would appear that modified or novel therapy approaches to improving physical function could be considered in order to optimise participants' engagement and motivation. One example is hippotherapy, or therapeutic horse-back riding, which has been shown to improve balance in children with and without an intellectual disability,⁽¹⁵¹⁾ and sit-to-stand ability in adolescents with an intellectual disability.⁽¹⁵²⁾ Another novel approach, trampoline training, has been shown to improve physical fitness and balance in youth with an intellectual disability.⁽¹⁵³⁾ The use of interactive computer games have also been shown to improve GMSs in a previous occupational therapy study of children with Down syndrome.⁽¹⁵⁴⁾ The use of Wii games have similarly been reported to result in improved dynamic balance in a single case of childhood onset acquired brain injury.⁽¹⁵⁵⁾ Physiotherapists may consider the use of such novel adjuncts in interventions to achieve improvements in GMSs for clients with an intellectual disability.

1.14.1 Teaching and learning approaches to optimise client engagement

Individuals with an intellectual disability require specific therapy approaches that consider the individual's learning and psychological needs.⁽¹²⁴⁾ Optimising participation and engagement are important when implementing therapy interventions for clients with intellectual disabilities due to these clients having comparatively less motivation, greater impairments in learning, and more difficulty adapting to change than their healthy peers.^(1, 156) In order to achieve optimal therapeutic outcomes, physiotherapy interventions need to be carefully planned with consideration of these issues in order to optimise the client's ability to participate freely in the therapy programme. For example, when working with children with an intellectual disability, careful consideration of intellectual impairments such as those related to processing of information, and the known negative impact a deficit in this can have upon their motor development,⁽¹⁵⁷⁾ should be incorporated into physiotherapy intervention planning.

A range of therapy approaches for optimising learning of improved GMSs have been reported for individuals with an intellectual disability. These include optimising extrinsic rather than intrinsic modes of learning. Types of extrinsic inputs include: peer modelling of skills as a singular approach or with additional positive reinforcement;^(158, 159) home based modelling of skills;⁽¹³⁰⁾ encouragement of adherence to therapy interventions;⁽¹⁶⁰⁾ and the use of an external rather than an internal focus of attention, for example focusing on throwing an object at a specified target rather than upon the body's movements needed to achieve such a manoeuvre.⁽¹⁶¹⁾ Another teaching model for motor learning in which extrinsic factors are modified is error reduction. In this, the practice environment is simplified to ensure there is less likelihood of errors occurring during practice; this results in less conscious effort being used by the participant when learning the skill. This contrasts to the need to use a greater degree of conscious effort in resolving challenges experienced when making errors in GMSs within a less controlled environment.⁽⁶⁸⁾ The use of extended periods of training should also be considered for this population: training over a two year period has been reported to be successful for individuals with profound intellectual disabilities

acquiring new motor skills.⁽¹⁶²⁾ These factors could be effectively applied by physiotherapists when working with clients with intellectual disabilities in order to optimise clients' learning of motor tasks. Clients with profound multiple disabilities have additional challenges arising from having restricted verbal and non-verbal communication. Effective clinical engagement with this population requires giving careful attention to the client's vocal and non-vocal expressions which indicate their response to the intervention.⁽¹³⁷⁾

1.14.2 Consideration of specific health issues

There can be specific health issues for clients with an intellectual disability which impact upon the planning of physiotherapy clinical goals and interventions. These health issues may relate to specific developmental issues for a given syndrome, for example progressive loss of motor skills⁽¹⁶³⁾ in conditions which result in functional deterioration over time such as Rett Syndrome,⁽¹⁶⁴⁾ and also for non-progressive conditions which commonly result in secondary progressive deterioration of GMSs such as cerebral palsy.⁽¹⁶⁵⁾ It is important in such conditions to prevent or minimise deterioration in physical function.

Specific health issues may also relate to medical interventions used to reduce the impact of physical impairments in clients with an intellectual disability. For example, anti-spasticity medication, including baclofen, may be used for individuals with associated muscle spasticity (hypertonia)⁽¹⁶⁶⁾ which can commonly occur in cerebral palsy. For individuals with Prader-Willi syndrome, low muscle tone, decreased strength and obesity (which can progress to morbid obesity) are problematic. Clinical interventions which improve muscle strength and development and also support weight loss in this population are indicated.⁽⁶⁾ These interventions can be provided as part of a multi-disciplinary team programme incorporating medical pharmaceuticals such as growth hormone^(5, 167) alongside physiotherapy interventions for improving strength and motor proficiency.⁽¹⁶⁷⁾

There is also a need to be aware of the range of clinical safety concerns which may be relevant when treating clients with an intellectual disability; for example atlanto-axial instability in clients with Down

syndrome.⁽¹⁶⁸⁾ This condition can result in spinal compression and warrants prospective surveillance under relevant guidelines.⁽¹⁶⁸⁾ A risk of atlanto-axial instability would preclude an individual from participating in more rigorous physical activity such as contact sport.⁽¹⁶⁹⁾

1.14.3 Multi-disciplinary team models of care

A range of multi-disciplinary team models of care can be highly appropriate for service delivery for people with an intellectual disability. These include multi-, inter- and trans-disciplinary approaches which incorporate physiotherapy with other allied health disciplines such as occupational therapy and speech therapy. These multi-disciplinary team approaches may be particularly useful where therapy services are in short supply, for example, in rural settings which commonly experience health workforce shortages⁽¹⁷⁰⁾ and a need for improved service funding and staff training.⁽¹⁷¹⁾ The importance of multi-disciplinary approaches to care for individuals with an intellectual disability and other concurrent health concerns has been previously highlighted in a 20 year follow-up study of individuals with Rett Syndrome registered in Australia.⁽¹⁷²⁾ The effectiveness of a multi-disciplinary team approach can be optimised through careful consideration of the input from each health profession; this is also indicated to prevent overlaps between professions.⁽¹⁷³⁾

1.14.3.1 *Multi-disciplinary team programmes for children with intellectual disabilities*

There are well documented and researched multi-disciplinary team approaches to early intervention for children aged under six years with developmental disabilities^(17, 174, 175) including intellectual disabilities.^(167, 176) An early intervention team may incorporate a range of allied health disciplines including physiotherapy, with its focus to address the multi-factorial effects of developmental disabilities including intellectual disabilities on the individual's growth and development.^(17, 174, 177) Reported evidence regarding effective early intervention care models supports the use of outcome measurement tools,⁽¹⁷⁸⁾ and the implementation of screening assessment for conditions resulting in developmental delay,⁽¹⁷⁹⁾ including

intellectual disability syndromes, such as Williams syndrome,⁽¹⁸⁰⁾ in order to enable prompt commencement of therapies.

1.15 Systematic reviews: overview

Systematic reviews provide a rigorous interpretation of a body of scientific literature concerning a particular domain of health care practice or policy. In a systematic review, defined objectives and methodologies guide the selection of studies, extraction of data, and the interpretative synthesis of data. Such predetermining of the approaches to be used minimises bias by the review's authors when conducting the review. These approaches may include publication of an *a priori* research protocol in a peer-reviewed journal and completing the development of comprehensive search strategies prior to commencing formal database searches. The final selection of studies to be included in a review should be based upon initial screening of the studies using pre-determined inclusion and exclusion criteria, and subsequently by the utilisation of appropriate critical appraisal checklists.⁽¹⁸¹⁾

There is a challenge for health care providers to maintain current knowledge in their clinical field due to the ongoing increases in primary and secondary research published, and the evidence base continually shifting and being updated. A range of newer approaches which endeavour to meet this challenge have been published in areas of clinical research which are relevant to the current review. It has been recommended that clinicians give careful consideration to input from clients, and also from the parents/carers of dependent or paediatric clients, and valuing this as an important source of clinical evidence.⁽¹⁸²⁾ Newer approaches for systematic reviews have also been utilised. These approaches include the use of an overview which synthesizes the results of previous systematic reviews and incorporates additional primary research findings to better answer a clinical question of concern. This approach has been reported for investigating the broad field of child health.⁽¹⁸³⁾ In the field of developmental disability, a comprehensive review has been utilised for researching evidence-based practice interventions for autism spectrum disorder.⁽¹⁸⁴⁾

1.15.1 Secondary research incorporating a range of study designs

Previously, RCTs have been seen as the best evidence to be considered for synthesis in systematic reviews. However, more recently there has been increased value placed upon alternate study designs being assessed within systematic reviews. For example, the Cochrane collaboration has extended the scope of the risk of bias assessment to include appraisal of issues specific to non-standard RCTs and pseudo-RCTs.⁽¹⁸⁵⁾

Systematic reviews which include non-experimental designs, such as descriptive and observational studies,⁽¹⁸⁶⁾ are indicated for domains of research for which it is difficult to conduct RCTs. This is relevant to the current review due to there being a number of reasons why conducting RCTs can be difficult in the field of intellectual disability research. These reasons include: studies in which randomisation would not be possible due to the psychological needs of the participants;⁽¹⁸⁷⁾ ethical concerns preventing an experimental study design; or where the inclusion and exclusion criteria of an RCT would limit generalisability to the broader clinical population.⁽¹⁸⁸⁾ Due to these issues, it was expected that few RCTs would have been conducted in this review's topic. Therefore, in order to optimise the identification of all published data for the current review, a range of experimental study designs were considered for inclusion, including controlled experimental designs, descriptive experimental designs and case report studies.

1.15.2 Evidence-based practice, Best Practice and systematic reviews

Evidence-based practice is a model of decision-making which enables clinicians to make informed decisions regarding health care provision for their clients based upon the best available evidence for clinical interventions with respect to safety and effectiveness, such as is available from systematic reviews. Results of systematic reviews are considered to provide a higher level of evidence compared to individual studies.⁽¹⁸⁹⁾ Evidence-based practice is based not only upon utilising the best available research

evidence, but also upon defining the clinical needs and preferences of the client, and combining these sources of evidence with the clinician's experience and knowledge.⁽¹⁹⁰⁾

Best Practice differs from evidence-based practice, but is intrinsically related to it. Best Practice is the structured integration of the best available evidence into clinical practice within an organisational construct.⁽¹⁹¹⁾ This process of integration seeks to embed the best evidence into the local organisational culture and preferences using a continuous quality assessment process. Where available, Best Practice guidelines should inform this process. The goal of such guidelines is to inform practitioners and consumers of the recommended approaches to managing a particular health issue and the potential for these recommendations to be extrapolated to different clinical contexts and environments.⁽¹⁹⁰⁾ Such guidelines are developed by multi-disciplinary field experts utilising the best available evidence, in particular systematic reviews where available. The results from the current review will be able to be considered for appraisal as part of evidence-based practice and Best Practice approaches of health care provision.

1.16 Previous systematic reviews in fields of research related to physiotherapy and intellectual disability

Systematic reviews about topics related to but not however specifically addressing the current review's focus were identified.^(5, 149, 150, 192, 193) These reviews varied in regard to whether specific or broad-based inclusion criteria were used for the PICO (population, intervention, comparator, outcome) domains, particularly for the domains of population and intervention. For example, previous systematic reviews have focused either on specific interventions (such as PBWSTT) for a broader range of disability diagnoses (including intellectual disability),^(149, 150) or on more general physiotherapy interventions⁽¹⁹³⁾ for study populations with a specific disability diagnosis such as cerebral palsy.⁽¹⁹³⁾ As well, reviews have been conducted which included more focused criteria for both the population and intervention, for example strength training for participants with cerebral palsy,⁽¹⁹⁴⁾ and for Prader-Willi syndrome.⁽⁵⁾ The

ability to extrapolate evidence from systematic reviews with more focused inclusion criteria for population and intervention domains to physiotherapy practice can be limited. This is because a clinician may be considering intervention planning for clients with a different diagnosis or for clients who do not tolerate certain types of interventions. The current review sought to overcome these issues by using broad-based inclusion criteria for the population and intervention PICO domains.

1.17 Need for this systematic review

There is an imperative for clients with an intellectual disability that physiotherapy interventions are effective, safe and evidence-based.⁽¹⁴⁵⁾ Evidence is required to support effective clinical practice in order to enable these clients to best meet the impacts of their lifelong disabilities. To date, no systematic review has been identified that appraises physiotherapy interventions used to improve GMSs in people with an intellectual disability.

The objectives of the current systematic review were to identify the best available evidence regarding the effectiveness of physiotherapy to improve GMSs in people with an intellectual disability aged six years and older. A secondary outcome was also to assess improvements in levels of physical activity, given the clear relationship between GMSs and physical activity in this population (section 1.12.1).

1.17.1 Use of overarching search strategies in this review

The search strategies used in this review were designed to reflect international trends in support of disability health research, and to fit within the ICF framework (sections 1.8 and 1.9). Additionally, the search strategies were structured to be specific to intellectual disability rather than developmental disability in general, and investigate the less resourced clinical field of therapy for clients with intellectual disability aged six years and over.

1.17.1.1 *Wide age spectrum*

Physiotherapists can assist clients with an intellectual disability of any age to overcome GMS deficits, using a habilitative approach to attaining more mature GMSs. There are well researched multi-disciplinary team interventions incorporating physiotherapy for children aged under six years with a developmental disability including an intellectual disability^(17, 174, 177) (section 1.14.3.1). There is comparatively less therapy follow-up beyond this age for individuals with an intellectual disability, as well as less research in this clinical area. It is therefore necessary to systematically review the available literature for persons aged six and over with intellectual disability in order to identify the best evidence to support physiotherapy practice for this population.

There is precedence in the reported literature to applying research findings across different age ranges for people with an intellectual disability. A previous study reported the development of a pain assessment tool for non-communicating adults with intellectual developmental disabilities being based upon a previously validated pain assessment tool developed for non-communicating children with comparable disabilities.⁽¹⁹⁵⁾ This approach of extrapolating clinical research findings to different age-groups may also be applicable in the field of physiotherapy for the improvement of GMSs in people with intellectual disability.

The relevance of conducting a systematic review researching across a wide age-range is also relevant to current clinical issues dilemmas identified by and advocated for by the Australian Physiotherapy Association (APA). These issues include difficulties experienced by individuals with disabilities when transitioning from one age-bracketed disability service to the next, for example between early intervention, school-age, adolescent and adult services.⁽¹⁹⁶⁾ The APA has also highlighted a lack of pragmatic assessment and therapeutic inputs for children with mild to moderate physical disabilities within Australian schools.⁽¹⁹⁷⁾

1.17.1.2 *Addressing specific learning needs*

The search strategies were also designed to capture as wide a field of physiotherapy clinical practice as possible, due to the paucity of research within this field. As well, it was thought that this approach could help to elicit common approaches used to address specific learning and motivation needs for clients with an intellectual disability (section 1.6). This has been reflected in the related clinical field of stroke rehabilitation, in which it has been found that rehabilitation following stroke is more efficacious when clients are mentally well engaged.⁽⁴¹⁾ The value of optimising the level of mental engagement for people with an intellectual disability when undertaking new activities has been previously reported: for example when participating in early schooling⁽¹⁹⁸⁾ or when increasing general activity levels.⁽¹⁹⁹⁾

1.17.1 Objectives of this systematic review

This review aimed to identify the best available evidence of physiotherapy interventions to improve GMSs in people with an intellectual disability aged six years and over. A broad-ranging search strategy was chosen as it was expected that the field of available research would be small. Broad inclusion criteria were applied for; the types of studies considered, the age-range of participants, allowing for any diagnosed intellectual disability to be considered, the types of interventions and the assessment outcomes.

2 Methods

2.1 Published *a priori* systematic review protocol

This review was conducted in accordance with the published *a priori* systematic review research protocol,⁽²⁰⁰⁾ with one point of change only: this review used only primary sourcing databases. Accordingly two of the databases stated in the *a priori* research protocol⁽²⁰⁰⁾ (PEDro and Scopus) were not utilised.

2.2 Review Question

What is the best available evidence for the effectiveness of physiotherapy to improve gross motor skills in people with an intellectual disability aged six years and older?

2.3 Objectives of this review

2.3.1 Primary objective

The primary objective of this systematic review was to identify the best available evidence regarding the types, as well as the effectiveness, of physiotherapy interventions used to improve gross motor skills in people with an intellectual disability (children aged six years and over, and adults of all ages).

2.3.2 Secondary objective

The secondary objective of this systematic review was to identify, from studies already included in this review in accordance with the primary review objective, the best available evidence regarding the types, as well as the effectiveness, of physiotherapy interventions used to improve levels of physical activity in people with an intellectual disability (children aged six years and over, and adults of all ages).

2.4 Inclusion criteria: PICO

The search strategy followed the PICO inclusion criteria format. Broad inclusion criteria were chosen for this review to optimise the identification of studies in what was determined to be a small field of research. The population of interest was people aged six years and older with an intellectual disability, with or

without any other type of disability including any physical disability. The types of interventions eligible for inclusion were any type of physiotherapy intervention. The inclusion criteria for the comparator domain encompassed study participants receiving either no physiotherapy intervention, or their usual care where this did not include physiotherapy. The criteria used for outcome assessment was the objective measurement of any GMS using any validated assessment tool. Additionally, the objective measure of levels of physical activity was also considered, where this outcome was reported in any included study as per the secondary research objective above (section 2.3.2).

2.4.1 Population

This review considered studies in which 50% or more of participants were aged six years old or older, and in which 50% or more of participants had an intellectual disability (mild through to profound) with or without GMS deficits (mild through to profound).

2.4.1.1 *Participants with an intellectual disability*

For the purposes of this systematic review, the term ‘intellectual disability’ referred to a person having a non-progressive impairment in their academic ability and decreased skills in adaptive behaviour and learning, with the onset of the intellectual disability occurring during the developmental years.^(1, 201) The overarching definition – intellectual disability – was chosen as it allowed for studies with participants who had intellectual disabilities arising from any diagnosis, or with no known cause, to be considered for inclusion, rather than selecting studies based on diagnoses of specific conditions from which intellectual disability may arise. Previous systematic reviews have similarly used the overarching term of intellectual disability as the inclusion criteria for the study population of interest.^(124, 149)

For studies which included participants with conditions which may or may not result in an intellectual disability, such as Autism Spectrum Disorder or cerebral palsy, only those papers which specifically reported the number or percentage of participants with an intellectual disability were considered for

inclusion. Additionally, these studies were only included if the reported prevalence of intellectual disability was at least 50%.

2.4.1.2 Age range of participants

The considered age range was six years and older without an upper limit. The rationale for using this age range was to exclude early intervention studies focusing on children with developmental disabilities under six years of age. Children in this younger age range undergo rapid developmental change, and for this age group there are more therapy interventions available. In contrast, the availability of therapies for people with an intellectual disability diminishes through the school years, young adulthood, and particularly in mid-later adulthood. Additionally, interventions for individuals aged six and older which result in improved GMSs are likely to be subsequently advantageous in improving the ability to participate in school and recreational and vocational activities.

The broad age range chosen for consideration for this review also reflects previous clinical research in the field of intellectual disability in which a pain assessment tool previously validated for assessing pain in non-communicating children was successfully extrapolated and applied for use with adults who were unable to communicate.⁽¹⁹⁵⁾ It was felt for the current review that identifying data for study populations covering broad age ranges could provide a platform for using a similar approach of extrapolating findings for effective physiotherapy interventions across different age ranges.

2.4.1.3 50 percent prevalence for key characteristics of study participants

The threshold of 50% prevalence for two key participant characteristics of age and intellectual disability was chosen due to some primary studies in the current field of research including participants with and without an intellectual disability and/or with varied age ranges. The 50% thresholds used in this review are comparable to the approach used in a systematic review of outcomes for children aged nine years or younger with physical disabilities.⁽¹⁷⁵⁾ In that review, studies were considered for inclusion if 30% or more

of participants had a physical disability, and at least 45% of participants were aged under 10 years. For the current review, the higher threshold of 50% was chosen to optimise the likelihood of including primary studies in which intellectual disability was a prominent consideration when developing the study design and choosing interventions and outcome assessments to be used in the study.

2.4.1.4 *Aetiologies for GMS deficits*

For the purposes of this systematic review, GMS deficits were defined as a person having activity limitations resulting in impaired movement and function arising from biomedical and /or psychosocial causes as outlined in the WHO ICF framework (figure 1).⁽³³⁾ The GMS deficits needed to have been assessed using validated outcome assessment tools.

The aetiologies for GMS deficits that were considered included acquired reasons such as an acute health condition (for example orthopaedic trauma, cerebro-vascular accident), developmental causes (for example Down syndrome, cerebral palsy, developmental delay), or a combination of both acquired and developmental reasons, and any other cause/s.

2.4.2 Interventions

Physiotherapy interventions could be either habilitative (focusing on the attainment of new or improved GMSs not previously able to be performed by the individual to a more age-specific or typical level) or rehabilitative (focusing the regaining of GMSs lost following an acute health insult or impairment of health). Such interventions could include, but not be limited to; exercise therapy, task-specific practice, group programmes, individual intervention sessions, and peer-facilitated contexts.

Studies that reported outcome assessments for interventions lasting eight weeks or longer were eligible to be included. Previous reviews have reported physical interventions for people with cerebral palsy with varied durations ranging from half a week to 20 weeks for stretching programmes, and three to ten weeks for strength training for individuals with cerebral palsy.⁽¹²⁾ For individuals with Prader-Willi syndrome,

physical training programmes along with pharmaceutical interventions have been studied;^(5, 6) the duration of the physical training programmes were found to vary between two weeks and one year.

2.4.2.1 Registered physiotherapist status

In this review, a physiotherapist was considered to be an individual who had successfully completed a recognised tertiary qualification enabling formal professional registration as a physiotherapist with an accredited registering body. Physiotherapy, whilst being represented through a World Confederation, is a nationally controlled profession.⁽²⁰²⁾ As such, there are different practice guidelines and requirements for physiotherapists in each nation that has the profession of physiotherapy practicing within it.⁽²⁰²⁾

This systematic review accepted on face value the statement within study reports that the intervention and assessment was provided, or prescribed and supervised, by a physiotherapist. Studies in which interventions for improving GMSs were administered solely by other professions, such as occupational therapy or physical education, were not included in this review (appendix III). This was done due to the physiotherapy profession having its own unique and specific professional registration and ethical requirements,⁽⁴⁰⁾ and standards of tertiary education.⁽³⁹⁾

2.4.2.2 Oversight of study interventions provided by physiotherapist

For the purposes of this systematic review, the interventions and predetermined assessments included in the studies needed to have been instigated by a qualified and registered physiotherapist, working either as an independent therapist or within a multi-disciplinary team⁽⁴¹⁾ context. The interventions could be conducted with a physiotherapist providing either direct and/or supervised care. Studies in which a physiotherapist provided supervised interventions (for example when a carer, family member, or other appropriate individual was trained to assist the participant/s to complete the intervention) were considered for inclusion only if a physiotherapist supervised the entire intervention programme and provided appropriately timed re-assessment.

2.4.2.3 *Settings for interventions*

Settings for interventions could be any usual physiotherapy clinical setting including acute hospital inpatient wards, hospital based rehabilitation centres, and outpatient clinics, or community-based settings, such as residential care facilities, schools, or the client's home (section 1.5.2.1).

2.4.2.4 *Study design considerations to meet participants' cognitive and learning needs*

For this review, particular attention was given to study design elements which aimed to meet the specific cognitive and learning needs of participants. Examples of these could include inputs to enhance participant engagement in the intervention, the use of extrinsic cues to help lessen the need for participants to use intrinsic foci of attention, or facilitation to support the carry-over of skills learnt during the intervention into the participant's everyday life.

2.4.3 Comparators

Types of comparators considered for inclusion in this review were having no physiotherapy intervention, or receiving usual care which did not include any physiotherapy, but which could include other more general activities such as a school-based physical activity programme led by a physical education teacher.

2.4.4 Outcomes

The outcome criteria used in this review encompassed any type of GMS which was assessed using a validated outcome assessment tool. The tools for measuring GMS outcomes were considered to have been "validated" if they had been previously assessed for reliability and validity in a clinical cohort the same or similar to the study population.

2.4.4.1 *Primary outcomes*

The primary outcome for this review was the quantitative assessment of the level of attainment of any

GMS using a validated measurement instrument. A similarly broad inclusion criteria was used in a Cochrane review of effectiveness of PBWSTT for children at risk of neuromotor delay, for assessing the outcome of motor function.⁽¹⁵⁰⁾

This review's primary outcome was designed to be broadly inclusive of both the type of GMS outcome being assessed, for example gait or balance, as well as of the assessment of any sub-classification of that GMS. For example, for gait, sub-classifications could include cadence and velocity, and for balance, sub-classification may include measuring postural sway in different directions.

2.4.4.2 *Secondary outcomes*

The secondary outcome for this review was the level of engagement in physical activity achieved by study participants. Physical activity can be measured in different ways, including level of endurance⁽³²⁾ and time spent in non-sedentary activity.⁽³¹⁾ This outcome was considered if it was reported in studies which had been primarily sourced based on GMSs as the primary outcome.

2.5 Types of study designs

This review considered a range of experimental study designs including randomised and pseudo-randomised control trials, quasi-experimental, before and after studies and case control studies. This review also considered descriptive epidemiological studies including case series and individual case reports. A previous review that reported the effectiveness of PBWSTT similarly considered case study reports due to the field of research being small.⁽²⁰³⁾

Any quantitative systematic reviews identified in the searching process, which reported on topics related to physiotherapy for improving GMSs in people with an intellectual disability, were retrieved for the purpose of hand-searching the reference lists for relevant articles. Of note, none of these systematic reviews were directly related to the systematic review presented in this thesis.

2.5.1 Time frame for date of publication

Studies published from 1-1-2008 to 22-10-2014 were considered for inclusion in this review. The start date was chosen to ensure that recent models of physiotherapy practice and theory were represented in the review's findings. It was also chosen to optimize the likelihood that the studies would reflect the tenets of the 2006 UN CRPD which was assigned formal status as a Human Rights Treaty in 2008.⁽²⁰⁴⁾ The UN CRPD has received ongoing review and monitoring.⁽¹⁰⁶⁾ The overarching clinical paradigms of the WHO ICF (section 1.8), which provides a comprehensive framework for assessing and appraising disability,⁽³³⁾ have been incorporated into this monitoring process.⁽²⁰⁵⁾

2.5.1.1 *Optional extended timeframe*

As outlined in the *a priori* protocol,⁽²⁰⁰⁾ in the case of an insufficient number of papers being identified within the timeframe for publication to 2008, a broader timeframe extended to 2001 would be used. This earlier date was chosen to coincide with the publication of the initial version of the WHO ICF⁽²⁰⁵⁾ which provides clinical paradigms useful to physiotherapy. However, a sufficient number of papers were identified and the extended timeframe did not need to be used.

2.5.2 Language of publication

Only studies published in English were considered for inclusion in this review. This was required due to the researchers only being fluent in the English language, and not having resources available for accessing translation services.

2.6 Reporting of results and types of statistical analyses

Numerical reporting of results was required using descriptive statistics such as mean and standard deviation (SD). Clear reporting of participant demographics was also required, including number of participants, the chronicity of their condition, and characteristics of the participants (including the similarities and differences between groups where applicable), as these factors impact upon the choice of

statistical analysis.

Appropriate statistical analyses needed to have been performed in order for the study to be included.

Note was made of any power analysis undertaken, or of any statistical comparison to normative data, or the use of z-scores for case report studies.

2.6.1 Head-to-head analyses

Head-to-head analyses are studies in which two interventions are compared, generally with one being the standard or default intervention, and the other being hypothesised to have superior performance in some regard. For this review's population of interest there are no known interventions which have been determined to be comparably effective in improving any GMS, and accordingly studies which undertook a head-to-head analysis were not eligible for inclusion in this review. However, in studies comparing two interventions of any previously determined efficacy, where before and after data were available, these studies were considered. In such studies, only the pre- and post- intervention data would be extracted for the primary intervention of interest.

2.7 Search strategy

The search strategies were designed to be broadly inclusive of the three domains of this review: intellectual disability, physiotherapy and GMSs. A broad-reaching approach was needed in order to increase the likelihood of identifying papers in a small and novel field of research.

General terms for 'intellectual disability' were used in the search strategies, including similes of the more historical term of mental retardation.⁽¹⁾ Diagnostic terms for specific conditions (diseases or syndromes) resulting in intellectual disability were not incorporated into the search strategies, except for Down syndrome as this is the most common condition causing intellectual disability.^(82, 206)

Diagnostic terms for different types of developmental disabilities for which a high proportion, but, importantly, not all, of affected individuals have an intellectual disability were not included. This was due

to the possibility that studies investigating outcomes for participants with such conditions would not necessarily include a study population of 50% or more of the participants having an intellectual disability. Accordingly, the diagnostic term of cerebral palsy was not included in the search strategy, as only approximately 50% of individuals with cerebral palsy have an intellectual disability.⁽⁸⁴⁾ Similarly, search terms for autism spectrum disorders were not included in this review as only approximately 75% of persons with autism spectrum disorders will have an intellectual disability.⁽⁸⁵⁾ Nonetheless, studies reporting results for participants with these and other developmental disabilities who also had an intellectual disability could still be identified by the broad-based search strategy and therefore considered for inclusion in this review.

The inclusion criteria of 'physiotherapy' was represented in the search strategy using terms used to formally describe it; this included its equivalent term of 'physical therapy', a term widely used in many areas of the world including the United States of America. The search strategy was developed to enable identification of studies in which a physiotherapist provided the intervention either as an independent practitioner or as part of a multi-disciplinary team; this was to reflect that people with an intellectual disability commonly benefit from health interventions within a multi-disciplinary context, from a range of health professionals. Terms to represent specific, narrower fields of practice in which physiotherapists may work, such as aquatic therapy or hippotherapy, were not included as these fields can also employ other health professionals; however any such studies identified by the database searches could be considered for inclusion.

The search domain of 'gross motor skills' was represented using derivatives of this specific phrase as well as other terminology with equivalent meaning. The use of such overarching terms was chosen to optimise the broad-reaching nature of the review whilst still enabling the identification of studies reporting on specific GMSs such as gait or balance; as such, terms to describe discreet types of GMSs were not utilised.

2.7.1 Three-step search strategy

The search strategy aimed to find published and unpublished (grey literature) studies. A three-step search strategy was utilised in this review. The search strategy was formulated following an initial limited search of all the primary-searching databases used: PubMed, CINAHL, Embase, and ProQuest, which were considered to be likely to provide comprehensive coverage of the field. This initial search aimed to identify relevant search terms which reflected the review's PICO inclusion criteria. The keywords used during initial database searches included terms relating to the age-range of six years and older (child, adolescent, adult, older adult, geriatric, very old adult), to the diagnosis of intellectual disability (intellectual disability, intellectual developmental disability, mental retardation, developmental disability, learning disability), to physiotherapy intervention (physiotherapy, physical therapy, rehabilitation, multi-disciplinary therapy, habilitation, exercise, hydrotherapy), clinical assessment (assessment, treatment, intervention, therapy, group, individual), and GMSs (gross motor skill/ function/ delay/ development, functional in/dependence, developmental milestones, balance, falls). Comprehensive search strategies were then developed for each of the four included databases. Full details of the database search strategies are presented below.

A second, full search was then undertaken across the four databases using the comprehensive search strategies.

Finally, the reference lists of all identified articles and relevant systematic reviews were searched for additional studies.

2.7.2 Database search strategies

Four primary sourcing databases were searched for this review: three databases comprising published peer-reviewed articles (PubMed, CINAHL, and Embase) and one database for grey literature searching of theses (ProQuest).

2.7.2.1 CINAHL

(MH Mental retardation+ OR MH Down syndrome+ OR TX mental retardation OR TX mentally retard* OR TX intellectual disab* OR TX intellectually disab* OR TX 'Down syndrome' OR TX 'Down's syndrome' OR TX 'Downs syndrome') AND (MH Motor skills+ OR TX Motor skill* OR TX Gross motor) AND (MH Physical Therapy+ OR MH Home Physical therapy+ OR MH Pediatric physical therapy+ OR TX physical therap* OR TX physiotherap*)

2.7.2.2 Embase

('intellectual impairment'/syn OR 'intellectual disability':ti,ab OR 'intellectual disabilities':ti,ab OR 'Intellectually disabled':ti,ab OR 'mentally retarded':ti,ab OR 'mental retardation':ti,ab OR 'Down syndrome'/syn OR 'Downs syndrome':ti,ab OR (Down next/1 's syndrome'):ti,ab) AND ('Motor performance'/syn OR 'motor development'/syn OR 'Motor skill':ti,ab OR 'motor skills':ti,ab OR 'gross motor':ti,ab) AND (physiotherapy/syn OR physiotherapist/syn OR 'home physiotherapy'/syn OR 'pediatric physiotherapy'/syn OR 'physiotherapy practice'/syn OR Physiotherap*:ti,ab OR 'Physical therapy':ti,ab OR 'Physical therapies':ti,ab OR 'Physical therapist':ti,ab OR 'Physical therapists':ti,ab)

2.7.2.3 ProQuest

(SU.exact("Downs syndrome") OR TI,AB(Downs syndrome) OR TI,AB(Down syndrome) OR TI,AB(Down's syndrome) OR TI,AB(Intellectually disabled) OR TI,AB(Intellectual impairment*) OR TI,AB(Intellectually impaired) OR SU.exact("Mental retardation") OR TI,AB(Mental retardation) OR TI,AB(Mentally retard*)) AND (SU.exact("motor ability") OR TI,AB(motor ability*) OR TI,AB(motor skill*) OR TI,AB(Gross motor*)) AND (SU.exact("physical therapy") OR TI,AB(Physical therap*) OR TI,AB(Physiotherap*))

2.7.2.4 PubMed

(physiotherapy/syn OR physiotherapist/syn OR 'home physiotherapy'/syn OR 'pediatric physiotherapy'/syn OR 'physiotherapy practice'/syn OR Physiotherap*:ti,ab OR 'Physical therapy':ti,ab OR 'Physical therapies':ti,ab OR 'Physical therapist':ti,ab OR 'Physical therapists':ti,ab) AND (Motor skills[mh] OR Motor skill*[tw] OR Gross motor[tw]) AND (Motor skills[mh] OR Motor skill*[tw] OR Gross motor[tw])

2.8 Study selection

Studies identified from the database searches were screened by title and abstract to assess whether they related to the review topic. Assessment of eligibility was then undertaken using full-text review, to determine whether the studies met the inclusion criteria; reasons for exclusion of studies were recorded. Study selection was performed by a single assessor (JH).

2.9 Critical appraisal

2.9.1 Critical appraisal instruments

Papers selected for retrieval were assessed at the study level by two independent reviewers for methodological validity prior to inclusion in the review; standardised critical appraisal instruments from the Joanna Briggs Institute Meta-Analysis of Statistics Assessment and Review Instrument (JBI-MAStARI) (appendix I)⁽²⁰⁷⁾ were utilised.

The studies included in this review reflected a variety of study designs. Accordingly, different MAStARI critical appraisal instruments which best matched each study design were utilised. For descriptive experimental studies, the appraisal instrument with best fit was the instrument for Randomised Control Trial/Pseudo-randomised Trial studies. For repeated measures studies, the Descriptive Experimental Studies appraisal instrument was used (appendix I).

2.9.2 Thresholds for inclusion

A threshold of four yes responses was required for a study to be included. This tolerance for a lower threshold score was chosen due to this field of research being small and novel, and one which is overlaid with various ethical and resource issues which create barriers to the ease of recruiting participants to studies. Specifically, 'Yes' responses were required for questions regarding: whether objective outcomes were reported; whether valid outcome measurement tools were utilised; if appropriate statistical analysis was undertaken; and whether follow-up assessment was done over a sufficient time-frame, or, for RCTs, whether the groups were treated identically.

2.9.3 Agreement between co-reviewers

It was pre-determined that if there were disagreements between the reviewers which could not be resolved by discussion, a third reviewer would be consulted to decide the matter. All disagreements, however, were resolved through discussion, and consultation with a third reviewer was therefore not required.

2.10 Data extraction

Data was extracted from papers included in the review using the standardised data extraction tool from JBI-MAStARI (appendix II).⁽²⁰⁷⁾

The data extracted included specific details about the interventions, populations, study methods and outcomes relevant to the review question and specific objectives. Attention was given to the reporting of study design elements which addressed participants' cognitive, behavioural and/or learning needs.

2.10.1 Outcomes results data

Data was only extracted if exact numerical results were reported, irrespective of whether exact results for statistical significance were reported. Usual choices for extraction of data were made for experimental studies in which the control group received no physiotherapy intervention. For experimental studies in

which the control group received an alternative physiotherapy intervention, comparator results were obtained by extracting base-line data for the intervention group participants, and control group data were not extracted. For repeated measures studies, pre-intervention baseline data were extracted.

2.11 Data synthesis

Two approaches for data synthesis were considered due to the possibility that a wide variety of PICO criteria could be represented in the included studies resulting from the use of a broad-based search strategy. These two approaches were meta-analysis and narrative review. Meta-analysis could be considered for any results for which there was a sufficient degree of homogeneity between the studies regarding population and intervention characteristics; additionally, meta-analysis of sub-groups could be performed. For data results with a greater level of heterogeneity present within the PICO criteria, a narrative synthesis could be completed.

3 Results

3.1 Primary and secondary objectives of this review

This review successfully met its primary objective by identifying the best evidence regarding physiotherapy interventions for improving GMSs in people with an intellectual disability aged six years and older, and the effectiveness of these interventions. In contrast, the secondary objective for this review could not be addressed as none of the papers selected for this review measured levels of physical activity in study participants.

3.2 Selection of studies

The search strategy identified 42 papers for full text review (figure 2). Thirty-five were subsequently excluded (appendix III). Critical appraisal was conducted for the remaining seven papers.

3.2.1 Included studies

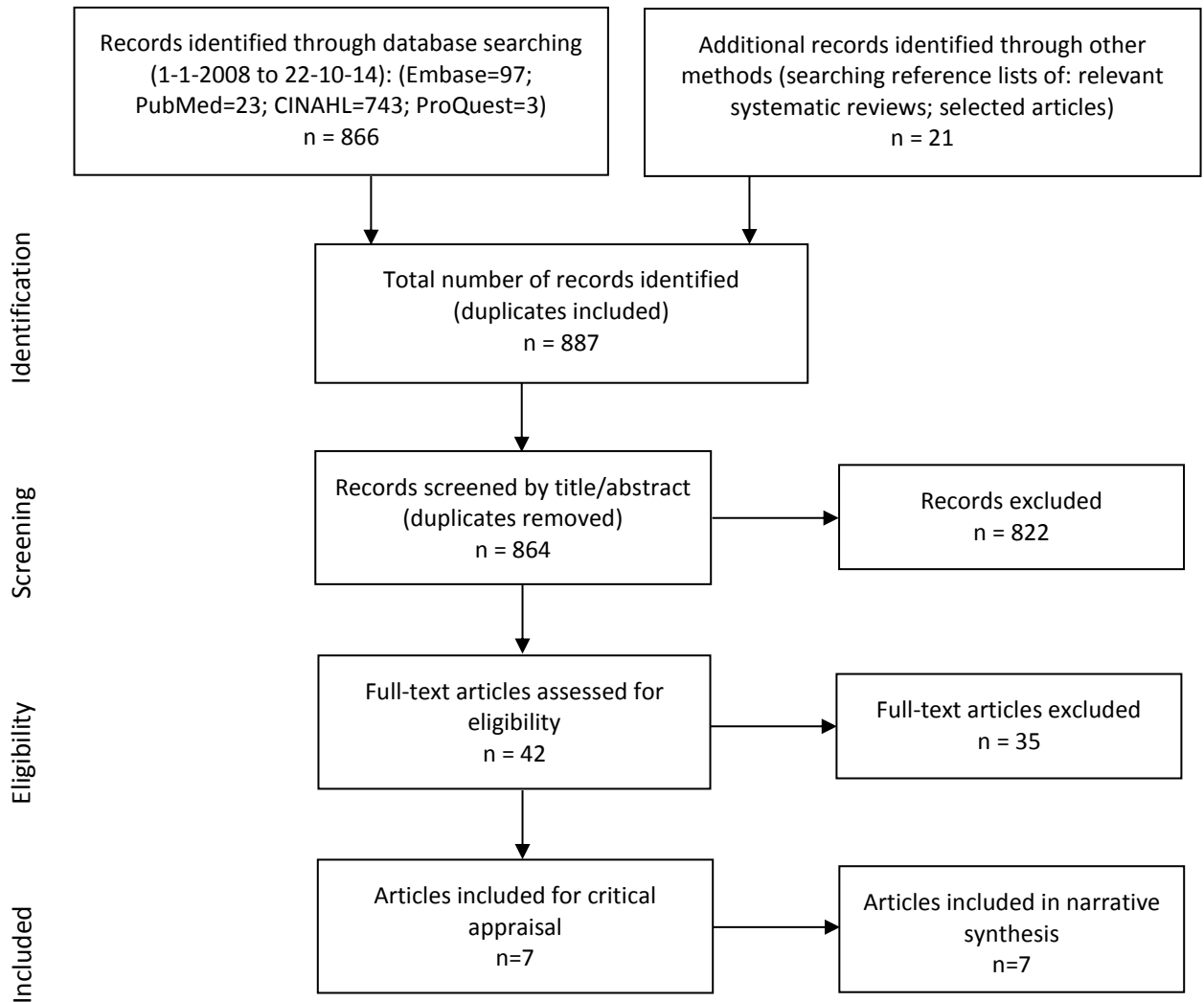
Following critical appraisal, all seven articles were selected for inclusion: two RCTs, two pseudo-RCTs, two pre-post studies and one case report study (table 4).

3.2.2 Excluded studies

Following full-text review of 42 papers (figure 2), 35 out of the 42 papers were excluded from this review because they did not meet the inclusion criteria (appendix III). The most common reasons for studies being excluded were a lack of reporting of whether the participants had any intellectual disability (nine studies), or there being less than 50% of participants with an intellectual disability in a particular study (eight studies) (appendix III).

Figure 2: Flow diagram of selection process

Based on Moher D, Liberati A, Tetzlaff J, Altman DG, The PRISMA Group. Preferred reporting items for systematic reviews and meta-analyses: the PRISMA statement.⁽²⁰⁸⁾



3.3 Results of critical appraisal

All included studies achieved a 'yes' response to the four required categories (section 2.9.2). Most questions for critical appraisal were able to be answered conclusively, with only a small number being rated as 'unclear' due to a lack of reporting detail in the study (tables 1 and 2). Appropriate statistical analysis, where relevant, was performed in all included studies (tables 1 and 2). Recruitment methods were described in most (six) studies; the use of convenience sampling was a common source of bias (five studies). In one study, the equipment used for assessments within the study was provided by the equipment manufacturer (Berg et al⁽²⁰⁹⁾). Approaches to allocation and blinding utilised in the RCTs and pseudo-RCTs were variably reported (table 4); in all cases the blinding of participants was recorded as 'no' due to the obvious nature of the interventions (tables 1 and 2). None of the studies reported whether the assessor was blinded to the allocation of participants; it was therefore presumed that the assessors were not blinded.

Table 1: Results of Critical Appraisal for Randomised Control Trial/Pseudo-randomised Trial Studies

| Study | Q1 | Q2 | Q3 | Q4 | Q5 | Q6 | Q7* | Q8* | Q9* | Q10* |
|--|-------|------|-------|------|-------|-------|--------|--------|--------|--------|
| RCTs | | | | | | | | | | |
| Shields et al ⁽²¹⁰⁾ | Y | N | Y | N/A | Y | N | Y | Y | Y | Y |
| Su et al ⁽¹⁶⁵⁾ (Cross-over RCT) | U | N | U | N | U | N | Y | Y | Y | Y |
| Pseudo-RCTs | | | | | | | | | | |
| Capodaglio et al ⁽²¹¹⁾ | U | N | U | N/A | U | Y | Y | Y | Y | Y |
| Vismara et al ⁽¹⁸⁷⁾ | U | N | U | N/A | U | Y | Y | Y | Y | Y |
| % yes | 25.00 | 0.00 | 25.00 | 0.00 | 25.00 | 50.00 | 100.00 | 100.00 | 100.00 | 100.00 |

Legend:

* = needed to score a 'yes' score for these question to enable inclusion in the review. Y = Yes, N = No, N/A = Not Applicable, U = Unclear, % yes = percentage of studies with 'yes' answer for particular question.

Questions:

Q1: Was assignment to treatment groups truly random? Q2: Were participants blinded to treatment allocation? Q3: Was allocation to treatment groups concealed from the allocator? Q4: Were the outcomes of people who withdrew described and included in the analysis? Q5: Were those assessing outcomes blind to the treatment allocation? Q6: Were the control and treatment groups comparable at entry? Q7: Were groups treated identically other than for the named interventions? Q8: Were outcomes measured in the same way for all groups? Q9: Were outcomes measured in a reliable way? Q10: Was appropriate statistical analysis used?

Table 2: Results of Critical Appraisal for Descriptive Experimental Studies

| Study | Q1 | Q2 | Q3 | Q4* | Q5 | Q6* | Q7 | Q8* | Q9* |
|----------------------------------|------|-------|-------|--------|-----|--------|-----|--------|--------|
| Repeated measures studies | | | | | | | | | |
| Aguar et al ⁽¹⁶⁹⁾ | U | N | N | Y | N/A | Y | N/A | Y | Y |
| Kurz et al ⁽²¹²⁾ | N | Y | Y | Y | N/A | Y | N/A | Y | Y |
| Case report | | | | | | | | | |
| Berg et al ⁽²⁰⁹⁾ | U | Y | Y | Y | N/A | Y | N/A | Y | Y |
| % yes | 0.00 | 66.67 | 66.67 | 100.00 | N/A | 100.00 | N/A | 100.00 | 100.00 |

Legend:

* = needed to score a 'yes' score for these question to enable inclusion in the review. U = Unclear, N = No, Y = Yes, N/A = not applicable, % yes = percentage of studies with 'yes' answer for particular question.

Questions:

Q1: Was study based on a random or pseudo-random sample? Q2: Were the criteria for inclusion in the sample clearly defined? Q3: Were confounding factors identified and strategies to deal with them stated? Q4: Were outcomes assessed using objective criteria? Q5: If comparisons are being made, was there sufficient descriptions of the groups? Q6: Was follow up carried out over a sufficient time period? Q7: Were the outcomes of people who withdrew described and included in the analysis? Q8: Were outcomes measured in a reliable way? Q9: Was appropriate statistical analysis used?

3.4 The JBI Grades for Levels of Evidence

The new grading approach developed by the JBI for classifying the levels of published research evidence⁽²¹³⁾ provides a useful framework for evaluating the quality of evidence included in this review. Through using this grading approach, the levels of evidence of the papers included in this review have been classified as: 1.c (two studies); 1.d (two studies); 3.e (two studies); and 4.d (one study) (table 3). This indicates that the overall level of evidence in this review was low.

Table 3: JBI Levels of evidence for Effectiveness Reviews

Based upon Joanna Briggs Institute, School of Translational Health Science. New JBI Levels of Evidence; The University of Adelaide; 2014.⁽²¹³⁾

| JBI Levels of Evidence | Sub-categorisation of levels; descriptions | No. of studies | Citations |
|---|--|----------------|---|
| Level 1 Experimental Designs | 1.a - SR of RCTs | | |
| | 1.b - SR of RCT & other study designs | | |
| | 1.c - RCT | 2 | Shields N, Taylor NF, Dodd KJ. Effects of a community-based progressive resistance training program on muscle performance and physical function in adults with Down syndrome: a randomized controlled trial. Arch Phys Med Rehabil 2008;89:1215-20. Su IYW, Chung KKY, Chow DHK. Treadmill training with partial body weight support compared with conventional gait training for low-functioning children and adolescents with nonspastic cerebral palsy: A two-period crossover study. Prosthet Orthot Int 2013;37:445-53. |
| | 1.d - Pseudo-RCTs | 2 | Capodaglio P, Cimolin V, Vismara L, Grugni G, Parisio C, Sibilia O et al. Postural adaptations to long-term training in Prader-Willi patients. J Neuroeng Rehabil 2011;8:26. Vismara L, Cimolin V, Grugni G, Galli M, Parisio C, Sibilia O et al. Effectiveness of a 6-month home-based training program in Prader-Willi patients. Res Dev Disabil 2010;31:1373-9. |
| Level 2 Quasi- experimental Designs | 2.a - SR of quasi-experimental studies | | |
| | 2.b - SR of quasi-experimental & other lower study designs | | |
| | 2.c - Quasi-experimental prospectively controlled study | | |
| | 2.d - Pre-test – post-test or historic/retrospective control group study | | |
| Level 3 Observational – Analytic Designs | 3.a - SR of comparable cohort studies | | |
| | 3.b - SR of comparable cohort & other lower study designs | | |
| | 3.c - Cohort study with control group | | |

| | | | |
|---|--|----------|--|
| | 3.d - Case-controlled study | | |
| | 3.e - Observational study without a control group | 2 | <p>Aguiar Jr AS, Tuon T, Albuquerque MM, Rocha GS, Speck AE, Araujo JC et al. The exercise redox paradigm in the Down's syndrome: Improvements in motor function and increases in blood oxidative status in young adults. J Neural Transm 2008;115:1643-50.</p> <p>Kurz MJ, Stuberger W, DeJong S, Arpin DJ. Overground body-weight-supported gait training for children and youth with neuromuscular impairments. Phys Occup Ther Pediatr 2013;33:353-65.</p> |
| Level 4 Observational- Descriptive Studies | 4.a - SR of descriptive studies | | |
| | 4.b - Cross-sectional study | | |
| | 4.c - Case series | | |
| | 4.d - Case study | 1 | Berg P, Becker T, Martian A, Primrose KD, Wingen J. Motor control outcomes following Nintendo Wii use by a child with Down syndrome. Pediatr Phys Ther 2012;24:78-84. |
| Level 5 Expert Opinion and Bench Research | 5.a - SR of expert opinion | | |
| | 5.b - Expert consensus | | |
| | 5.c - Bench research/single expert opinion | | |

Legend:

JBI = The Joanna Briggs Institute; RCT = randomised controlled trial; SR = systematic review;

3.5 Narrative synthesis

The use of broad-based inclusion criteria in this review resulted in there being a high level of heterogeneity within the included studies in regard to the PICO domains of sample populations, therapy interventions, and outcome measurement tools. Only one gross motor outcome was reported in more than one study (cadence was reported in Kurz et al,⁽²¹²⁾ and Vismara et al⁽¹⁸⁷⁾); however it could not be considered for aggregate synthesis due to the sample populations being too heterogeneous (participants were diagnosed with neuromuscular impairments resulting from various developmental conditions, and Prader-Willi syndrome, respectively). It was therefore concluded that meta-analysis would not be possible for this review, and that the appropriate method for presenting the results would be narrative synthesis.

3.6 Study designs and characteristics

A range of study designs were represented in this review including RCTs, pseudo-RCTs, repeated measures studies and a case report (table 4). In the experimental studies, participant blinding was not attempted (all interventions were of an obvious nature to the participants), and no mock-up programmes were used for control participants. No head-to-head analyses were included. One of the RCTs (Su et al⁽¹⁶⁵⁾) reported on the comparative effectiveness of two physiotherapy interventions as part of a two-period cross-over study: it was not inferred, however, that the interventions had comparable efficacy, and so this study was not considered to be a head-to-head analysis.

Two pseudo-RCT studies were included in this review (Capodaglio et al;⁽²¹¹⁾ Vismara et al⁽¹⁸⁷⁾). It was noted that the summary demographic data describing the intervention groups in both studies (participants with Prader-Willi syndrome) were identical, whereas the demographic data for the control group participants in each study (healthy, non-disabled participants) differed slightly. It was confirmed (through email communication to the corresponding author, (who was the same person for both studies) that the intervention group participants in both of these studies received the same intervention at the same time, and that there were minimal differences in the study populations between the studies. For each of these

studies, different GMS outcomes were reported (balance and gait respectively). Both studies reported the results of statistical analyses comparing the baseline and interim results of the intervention groups (participants with Prader-Willi syndrome) to the baseline data for the control group (only base-line results were reported for the control group participants). These analyses revealed that the participants with Prader-Willi syndrome scored significantly poorer in cadence and velocity⁽¹⁸⁷⁾ and postural sway balance measures⁽²¹¹⁾ compared to the control group (table 4). These analyses, although not addressing the primary purposes of this review, do however illustrate that the intervention group participants had significant GMS impairments. Further discussion of results from these studies is presented below (sections 3.9.1 and 3.9.2).

3.6.1 Ethics

All studies reported full ethics approval, and all studies gained consent from participants or care-givers where appropriate.

3.6.2 Recruitment

Only one study utilised random sampling: by advertising on a limited number of disability listservs (computer database for sending information to registered individuals) to invite participation in the study (Berg et al⁽²⁰⁹⁾). One study did not report on the method used for recruiting participants (Aguiar et al⁽¹⁶⁹⁾). The remaining studies utilised convenience sampling.

3.6.3 Studies originating from research centres in industrialised nations

The studies included in this review arose primarily from developed countries, a factor which has been noted in a previous systematic review of mobility issues in adults with an intellectual disability.⁽¹²⁴⁾ These countries were Australia (Shields et al⁽²¹⁰⁾), Hong Kong (Su et al⁽¹⁶⁵⁾), Italy (Capodaglio et al⁽²¹¹⁾; Vismara et al⁽¹⁸⁷⁾), and the United States of America (Berg et al⁽²⁰⁹⁾; Kurz et al⁽²¹²⁾). Only one developing nation was represented which was Brazil (Aguiar et al⁽¹⁶⁹⁾).

3.6.4 Outcome results data

Numerical outcome data (table 4) were able to be extracted directly from the published papers except in three studies. For both pseudo-RCTs (Capodaglio et al;⁽²¹¹⁾ Vismara et al⁽¹⁸⁷⁾), a small number of summary statistical results were obtained from the corresponding author. In the study by Aguiar et al,⁽¹⁶⁹⁾ the summary results for the GMS outcome assessments were reported in a graphical format only. Consequently, raw data were obtained from the author, and numerical median results were subsequently calculated from this data.

3.6.5 Statistical analyses reported in the included studies

All of the included studies utilised a threshold value of $p \leq 0.05$ to determine whether a result was statistically significant and the null hypothesis was to be rejected. The numerical data for outcome assessment results were primarily presented as mean and SD data. Median results were presented in one repeated measures study (Aguiar et al⁽¹⁶⁹⁾), and, for a case report, individual results with z scores were reported (Berg et al⁽²⁰⁹⁾). A range of appropriate statistical designs were reported for analysing the GMS outcome results of the included studies (table 4).

In one RCT, the independent t test and the Fisher exact probability test were used as appropriate (Shields et al⁽²¹⁰⁾). The Wilcoxon matched pair test was used in two pseudo-RCTs (interim versus final results for intervention sub-group participants) (Capodaglio et al;⁽²¹¹⁾ Vismara et al⁽¹⁸⁷⁾), and in a repeated measures study for pre- versus post-intervention results (Aguiar et al⁽¹⁶⁹⁾). Effect sizes were calculated for RCTs (Shields et al;⁽²¹⁰⁾ Su et al⁽¹⁶⁵⁾) and repeated measures studies (Kurz et al;⁽²¹²⁾ Aguiar et al⁽¹⁶⁹⁾).

3.6.5.1 *Intention-to-treat analysis*

Intention-to-treat analysis (carry forward technique) was used for missing data for non-attendance of participants in one study (Shields et al⁽²¹⁰⁾). Only one study reported some attrition of participants (Su et al⁽¹⁶⁵⁾); intention-to-treat analysis was not reported in this study.

3.6.5.2 *Assessing demographic data and outcome results*

The impact of demographic data on GMS outcome results was assessed in only three studies (table 4). In two studies results data were normalised: results for standing balance were normalised to participants' height;⁽²¹¹⁾ and results for gait velocity were normalised to participants' leg length.⁽²¹²⁾ In one study, age stratification was used for the reporting of GMS outcome results (specifically for gait and the Gross Motor Function Measure (GMFM)⁽²¹²⁾); however, assessment of significance was not reported for these stratified results.

3.6.5.3 *Between-group differences in experimental studies*

Assessment of between-group differences regarding demographic details was only reported in one study (Aguar et al⁽¹⁶⁹⁾); this analysis revealed a statistical difference for participants' body weight between the two groups, but not for body mass index (BMI).

3.6.5.4 *Power analyses*

One study reported a pre-hoc power analysis to obtain 80% power (Shields et al⁽²¹⁰⁾); this analysis identified that a sample size of 10 was required for both the intervention and control groups. Ultimately, a sample size of 11 for the control group and nine for the intervention group was included; no explanation for the unequal numbers of participants in the two groups was given by the authors. None of the other studies reported prospective power analysis calculations prior to recruiting and undertaking the clinical study. However, a post-hoc power analysis was appropriately calculated in repeated a measures study (stated by the authors to be a pilot study) (Kurz et al⁽²¹²⁾), for the purpose of determining the required sample size for further clinical research. Su et al⁽¹⁶⁵⁾ discussed the usefulness of the two period cross-over design used in their RCT for increasing the power of the results by removing variability between participants who acted as their own controls; however they did not report the power of the results data. These authors also noted that a limitation of the cross-over design was the potential for a carryover effect,

and, as such, they recommended that future studies should have larger sample sizes and that a conventional RCT design be considered.⁽¹⁶⁵⁾

3.6.6 Confounding variables

Potential confounding variables were only discussed in one study in which participants were recruited from a special school (Kurz et al⁽²¹²⁾). In this study the researchers reported their informal observations of the school staff – that the staff were giving more support to students in practicing walking while the study was in progress. No other study reported either formally or informally on any potential confounding factors.

3.6.7 Safety issues addressed in the studies

Safety issues, with regard to avoiding known clinical risks, were necessarily addressed in one study of adults with Down syndrome. Potential participants for an intervention of an adapted Judo training⁽¹⁶⁹⁾ were screened for atlanto-axial instability, and were excluded if this spinal condition was present.⁽¹⁶⁸⁾ There was no other reporting of screening for known clinical risks in the remaining studies.

3.7 Participant characteristics

3.7.1 Participants' diagnosis of intellectual disability

A range of diagnoses for developmental disabilities resulting in intellectual disability were reported in the studies. These diagnoses included Down syndrome, Prader-Willi syndrome, cerebral palsy and various aetiologies arising in early childhood (table 4). Information regarding how the diagnosis of participants' intellectual disabilities were made was variably reported in the studies. For example, of the three studies which included participants with Down syndrome (a condition which always results in some degree of intellectual disability),^(169, 209, 210) only one study described how the diagnosis of Down syndrome had been confirmed for each participant (physical assessment of participant and review of karyotype reported in their case-notes) (Aguilar et al⁽¹⁶⁹⁾). In the second study, a default indication of how the diagnosis of Down

syndrome was determined was provided by stating that the participant had been recruited through Down syndrome awareness group listservs.⁽²⁰⁹⁾ In the third study,⁽²¹⁰⁾ no direct or indirect indication was provided of how the Down syndrome diagnosis had been made; instead it was simply stated that participants were recruited through vocational agencies that supported adults with an intellectual disability. For the two studies with participants with Prader-Willi syndrome,^(187, 211) (a condition which always results in intellectual disability (often mild)), it was reported that the diagnosis of Prader-Willi syndrome was made through clinical assessment of participants' phenotype and cytogenetic analysis of each participant.

In the remaining two studies, participants were diagnosed with developmental disabilities (childhood onset neuromuscular impairments (Kurz et al⁽²¹²⁾) and cerebral palsy (Su et al⁽¹⁶⁵⁾)) which do not always result in having an intellectual disability. For these studies, the participants' diagnosis of having an intellectual disability was inferred from the fact that the participants were recruited from special schools for youth with severe-grade mental handicaps (Su et al⁽¹⁶⁵⁾) or severe or profound cognitive disabilities (Kurz et al⁽²¹²⁾).

3.7.1.1 *Description of the severity of intellectual disability*

The severity of the intellectual disabilities present in the study participants was described in the articles as either mild, moderate, severe, or profound (table 4). There were varied criteria used for determining the severity of the intellectual disability. These included informal criteria such as the description of the special school at which participants attended, and the opinion of the participants' family regarding the level of severity; and formal criteria such as the use of the Mini-Mental State Examination (MMSE) Italian version. All participants in all studies were able to follow simple cues, which accorded with the study selection criteria reported in the studies.

3.7.2 Physical impairments of study participants

The physical disabilities present in the study participants were described in each study by reporting either the diagnosis, for example non-spastic cerebral palsy (Su et al⁽¹⁶⁵⁾), and/or the participants' key physical impairments, such as decreased balance (Capodaglio et al⁽²¹¹⁾). The functional impact of the physical disability for study participants was described using either a criterion classification system, for example the Gross Motor Classification Scale (GMFCS), or by describing the level of independence and use of assistive devices for gait (table 4).

3.7.3 Participants' levels of physical activity

None of the included studies reported any baseline or follow-up assessment of the levels of physical activity of the participants.

3.7.4 Age of participants

All studies had 100% of participants aged over six years (table 4). Studies mainly included children, youths and young adults (age range for all studies: 10.8 - 33.8 years). No studies included older adult or geriatric participants.

3.7.5 Reporting of body weight and Body Mass Index (BMI)

Body weight and BMI data were reported as baseline results with or without post-intervention results in most (five) studies. Pre- and post-intervention results were reported for: body weight in Berg et al⁽²⁰⁹⁾ (slightly underweight weight, remained stable); BMI in Aguiar et al⁽¹⁶⁹⁾ (non-significant reduction in BMI, participants remained within healthy weight range) and BMI in Capodaglio et al⁽²¹¹⁾ (overweight; slight reduction post-intervention). In two studies, only baseline measures were reported: and BMI in Shields et al⁽²¹⁰⁾ and Vismara et al.⁽¹⁸⁷⁾ No results for body weight or BMI were reported in Su et al⁽¹⁶⁵⁾ and Kurz et al.⁽²¹²⁾ None of the studies measured or discussed the potential impact body weight or BMI could have upon the GMS outcome results.

3.8 Interventions

3.8.1 Clinical oversight and settings for interventions

All studies had physiotherapist-led assessments and interventions which were conducted within a sole profession mode of practice. In four studies, a physiotherapist provided indirect supervision of the intervention programmes: three of these studies incorporated home-based exercise programmes,^(187, 209, 211) and one study included exercise programmes supervised by a fitness instructor within a community gym setting⁽²¹⁰⁾ (table 4).

A multi-disciplinary team model encompassing physiotherapy was utilised in only two studies: (for an initial hospital-based rehabilitation programme); in these studies a subsequent home exercise programme was supervised by a physiotherapist.^(187, 211)

3.8.2 Types of interventions

A range of physiotherapy interventions were reported in the studies (table 4). These included gait training and exercise programmes. Gait training was delivered in two modes: partial body-weight supported treadmill training⁽¹⁶⁵⁾ (PBWSTT), and overground body-weight supported (BWS) gait training.⁽²¹²⁾ The exercise programmes were provided as closed-chain leg strengthening exercises,^(187, 211) upper and lower limb gym-based resistance exercises;⁽²¹⁰⁾ and practice of Nintendo Wii games.⁽²⁰⁹⁾ Of the exercise programmes, three studies administered a home exercise programme intervention (Berg et al,⁽²⁰⁹⁾ Capodaglio et al;⁽²¹¹⁾ Vismara et al⁽¹⁸⁷⁾). The therapies which provided most physical support for participants were the gait training interventions using PBWSTT and BWS modalities; these were used in studies in which participants had a high severity of both their physical and intellectual disabilities^(165, 212) (table 4).

3.8.2.1 *Habilitative approaches*

All studies investigated interventions designed to provide a habilitative approach in therapy for improving GMS deficits arising from developmental disabilities. No studies included rehabilitation therapy interventions for recovery of and improvement in GMSs following acute injury, illness or surgery.

3.8.2.2 *Task specificity*

Task specific practice is the therapeutic practice of the same skill which the intervention is designed to improve; for example, to practice gait with or without modifications, to improve gait. The level of task specificity within the study interventions varied. Examples of interventions with high task specificity relevant to the type of GMS outcome being assessed included BWS gait training for improvements in walking (Kurz et al⁽²¹²⁾), and an adapted Judo training intervention for improvements in general GMSs (Judo is a sport which trains a wide array of GMSs for the whole body. Accordingly, an appropriate outcome tool for assessing the effectiveness of a Judo intervention is the GMFM-88 which measures multiple GMSs) (Aguiar et al⁽¹⁶⁹⁾). Studies in which the interventions showed low task specificity included leg strengthening exercises for improving balance (Capodaglio et al⁽²¹¹⁾) and gait (Vismara et al⁽¹⁸⁷⁾) and gym-based limb strengthening for improving stair-climbing (Shields et al⁽²¹⁰⁾).

In the case report study by Berg et al,⁽²⁰⁹⁾ the GMS outcome assessments were chosen to match whichever Wii games were chosen by the participant, so as to optimise relevance to the interventions. This pre-determined but somewhat open-ended approach to determining which GMS assessments to use, decreased the purposeful focus of which GMSs to improve in the study, but improved the relevance between the types of intervention undertaken, and the GMSs being assessed.

3.8.2.3 *Progression of interventions*

The approaches used for progressing interventions varied between studies. Types of approaches for progression included having no progression (Berg et al;⁽²⁰⁹⁾ Capodaglio et al;⁽²¹¹⁾ Vismara et al⁽¹⁸⁷⁾), or

progressing according to participant tolerance or ability (Shields et al⁽²¹⁰⁾; Su et al⁽¹⁶⁵⁾) which was progressively appraised at interim time-points and results were compared to pre-defined thresholds (table 4).

3.8.2.4 *Optimising participant engagement*

Approaches which were used to enhance participant engagement and motivation during the interventions included the use of a daily journal of adherence to the intervention programme,^(187, 211) enabling participant involvement in the choice of interventions,⁽²⁰⁹⁾ prescribing simple exercises, and ensuring the psychological needs of participants were considered when allocating them to a study intervention.⁽¹⁸⁷⁾

None of the studies reported on any specific or general study design considerations in regard to meeting the learning needs of the participants - either in regard to the types of interventions implemented, or the types of outcome assessment tools utilised. However, it was noted that all of the interventions incorporated a high degree of repetition in practice; this factor inherently allowed for less cognitive demands upon the participants, and so could be considered an informal approach to meeting participants' learning needs.

3.8.3 Attrition; adverse events

Only one study (Su et al⁽¹⁶⁵⁾) reported some attrition with two participants dropping-out due to medical reasons. High adherence rates were reported in all studies (table 4).

There were no or negligible adverse events reported in four studies. Two studies reported that the participants experienced no adverse events,^(165, 212) and one study commented that the participants did not suffer any severe injuries.⁽¹⁶⁹⁾ One study reported that the participants experienced mild muscle soreness following participation in the muscle strengthening intervention; this symptom resolved quickly.⁽²¹⁰⁾ Three studies did not include any reporting of whether there were adverse events.^(187, 209, 211)

3.9 Results extracted

All studies presented base-line results for all participants for all of the GMS outcomes reported. All studies reported post-intervention results for assessments conducted at the completion of the intervention for all participants who received an intervention. Interim results were reported in only two studies^(187, 211) (table 4).

3.9.1 Extraction of post-intervention data from experimental studies

Specific decisions regarding the extraction of post-intervention data were made for three of the experimental studies: one RCT⁽¹⁶⁵⁾ and two pseudo-RCTs.^(187, 211)

One RCT (Su et al⁽¹⁶⁵⁾) utilised a two-period cross-over design, and reported post-intervention results not only at the completion of the primary intervention of interest, but also following completion of an alternative physiotherapy intervention. For the purposes of the current review, only the post-intervention results measured at the completion of the primary intervention (PBWSTT) for the intervention group which received this intervention first were able to be extracted from this RCT.

In both pseudo-RCTs (Capodaglio et al;⁽²¹¹⁾ Vismara et al⁽¹⁸⁷⁾), post-intervention results data for participants with an intellectual disability was presented for two time-points. These time-points were; after a two-week preliminary inpatient rehabilitation programme delivered to all participants with an intellectual disability, and then after a subsequent six-month home programme intervention delivered to a sub-group of participants with an intellectual disability only. The interim results reported in both of these studies revealed no significant differences between the two sub-groups and accordingly the final post-intervention results were the main interest of this review.

3.9.2 Extraction of comparator data from experimental studies

All comparator results extracted for this review were for participants who had had no physiotherapy intervention. Specific decisions were made for this review regarding the extraction of comparator data from the experimental studies.

For the crossover RCT by Su et al,⁽¹⁶⁵⁾ the comparator data extracted were the base-line results measured prior to the administration of the primary intervention of interest for the first group receiving this intervention. For both of the pseudo-RCT studies (Capodaglio et al;⁽²¹¹⁾ Vismara et al⁽¹⁸⁷⁾), control group data could not be extracted because the control group was comprised of healthy, non-disabled participants, and only baseline results were reported. In both studies, all intervention group participants (individuals with Prader-Willi syndrome) received an initial two-week inpatient rehabilitation programme. Following this programme, the participants were divided into two sub-groups: group 1 who received a six-month home exercise programme; and group 2 who received no further intervention. The outcome data that was reported for each sub-group were results measured at three time-points: at baseline; at an interim time-point (following completion of the two-week inpatient programme); and post-intervention final results (following the six-month period for the home exercise programme). Statistical analyses of results for the intervention sub-groups compared the final and interim results for each sub-group separately; however, the final outcome results from each sub-group were not compared to each other (table 4). Accordingly, comparator data for both studies was considered to be the interim results for each sub-group.

3.9.3 Data extracted from repeated measures studies

There were no complicating factors with regards to the comparator results for the before and after studies included in this review (Aguilar et al;⁽¹⁶⁹⁾ Berg et al;⁽²⁰⁹⁾ Kurz et al). Base-line data as well as post-intervention results for measurements undertaken at the completion of the intervention were extracted for all participants in these studies.

3.10 Reporting of GMS outcome assessments

All studies measured one or more GMSs. Some studies also measured other clinical outcomes which were not specifically a type of GMS. In keeping with the review protocol, results for other outcome measures, even if they were the primary focus of the study, were not extracted; only outcome data for GMS domains were extracted.

3.10.1 Validity of GMS outcome assessment tools

For each of the GMS outcome assessments, prior reporting of validity testing carried out in a clinical population that was the same as or similar to the one represented in the study was confirmed, before data was extracted for this review. The GMS data extracted from the selected studies were measured using a range of validated outcome measurement tools specific for the clinical population, or for a clinical group with comparable physical characteristics (table 5).

The validated outcome assessment tools used in the studies included visual assessment check-lists, complex video-analysis, and force platform assessment tools (table 5). Only one GMS outcome assessment was excluded from this review: the Grocery Shelving Task (Shields et al⁽²¹⁰⁾), due to this test only being previously validated for a population of participants with respiratory disorders⁽²³⁶⁾ and not in a population with any intellectual disability and/or primary GMS impairment.

3.10.2 Length of follow-up

All studies delivered the interventions over a sufficient time-period. The duration of interventions in the studies varied, and ranged from eight weeks to six months (table 4). All post-intervention data were results measured at the completion of the study interventions; no study measured longer term follow-up results at a time following completion of the intervention.

Table 4: Characteristics of studies

| Study | Ss (sample size); (gender); Age (yrs) (mean (SD)); BMI (kg/m ²) or weight (lbs) | ID: diagnosis; severity. GMSs | Recruitment / Allocation | Intervention details: Setting; Type; Duration; Support; Progression | Adherence; Adverse events; Attrition | Data extracted | | |
|-------------------------------------|---|--|--|---|---|---|---|---|
| | | | | | | GMS Outcomes | Results: mean (SD) unless otherwise stated; italicized if $p \leq 0.05$ or results exceed MID | |
| RCTs | | | | | | | | |
| Shields et al 2008 ⁽²¹⁰⁾ | <p>Ss: Total = 20 (13 males); power analysis required 10 participants in each gp for $d = 80\%$ (per previous DS study⁽²³⁷⁾) 26.8 (7.8) yrs. IG = 9 (7 male) 25.8 (5.4) yrs BMI: 31.2 (6) CG = 11 (6 male) 27.6 (9.5) yrs ($p = 0.61$); BMI: 26.8 (5.2) ($p = 0.09$)</p> | <p>ID: DS; Moderate - severe ID (per carer report). GMS: Sufficiently fit to tolerate intervention (Physical Activity Readiness Questionnaire (PAR-Q) completed).</p> | <p>Recruitment: From two disability support agencies. Flyer inviting participation sent to clients' families. Allocation: Random assignment after recruitment completed; concealed allocation; block randomization.</p> | <p>Setting: Community gym. Type: Progressive resistance training using weight machines: strength exercises for upper limbs (3 exercises) & lower limbs (3 exercises). Completion of log book. Intensity: 10-12 reps (to fatigue), 2-3 sets, rest 2 min between sets. Duration: 2x/wk, 10 wks (20 sessions). Support: Small gp exercise training led by fitness trainer; log book completed by trainer. Progression: Resistance \uparrow when 2 x 12 reps achieved.</p> | <p>Adherence: 167 training sessions attended (92.8%). Non-attendance due to illness unrelated to the intervention. All participants tolerated increased weight resistance loading by $\geq 90\%$. Adverse events: Mild muscle soreness in 4 participants, resolved fully. Attrition: Nil.</p> | <p>Data extracted: Difference between groups, Week 10-Week 0, Intervention Gp-Control Gp, p value, effect size (95% CI)</p> | <p>Timed up and down stairs test (sec)</p> | <p>-3.4 (-7.7 to 1.0) [$p = .12$; effect size = 0.16 (-0.73 to 1.04)]</p> |
| Su et al | Ss: | ID: Non-spastic CP; | Recruitment: From | Primary intervention: | Adherence: 14-18 | Data extracted: Pre-and post-intervention | | |

| | | | | | | | | | | | | | | | | |
|---|--|--|---|--|---|---|----------------|--|---|--|-------------------|-----------|-------------------|-----------|------|-----------|
| 2013 ⁽¹⁶⁵⁾ | <p>Initial = 10 Gp I = 5 (4 males) Age 10.8 (1.6). Gp II = 5 (4 males) Age 11 (2.4). BMI not reported Following attrition = 8 (Gender not stated. Study results reported following attrition for Ss=8 only) Gp I = 4; Gp II = 4.</p> | <p>severe ID. GMS: GMFCS level (N): Level II (1); level III (1); level IV (5); level V (1). Types of CP (N): Dystonia (1); Choreoathetosis (6); Hypotonia without ataxia (1).</p> | <p>Special School for youth with severe ID Allocation: Stated that random allocation done; however randomisation method not described.</p> | <p>PBWSTT. Secondary intervention: conventional gait training. Timing of interventions: Two-period crossover study (groups I & II) with two arms of study. Gps received opposite interventions during each intervention period. Gp I received PBWSTT as initial intervention. Setting: not reported Type: PBWSTT. Intensity: 10 min, 5 min rest, further 10 min. Sessions 2x/wk. Duration: 2 training periods: each arm of the study comprised initial intervention (12 wks), 10-wk washout period, followed by other intervention (12 wks). Progression: Initial % BWS = 30%; (% BWS ↓ according to participant comfort) Final BWS = 27.5 +/- 5%. Treadmill speed (m/sec): Initial: 0.36 m/sec; (all participants reached pre-determined capped speed of 0.8 m/sec in 1st session; cap released if no adverse events; speed</p> | <p>intervention sessions attended, non-attendance due to health or emotional problems. Adverse events: Nil. Attrition: Attrition of 2 participants (1 per group) due to medical reasons. No ITT analysis.</p> | <p>results for Gp I for initial intervention (PBWSTT)</p> <table border="1" data-bbox="1520 334 1955 618"> <tr> <td colspan="2" data-bbox="1520 334 1955 375">GMFM-66</td> </tr> <tr> <td colspan="2" data-bbox="1520 375 1955 467">Mean (SD) of change scores: final score - baseline score for each participant; significance not reported for this data.</td> </tr> <tr> <td data-bbox="1520 467 1734 545">GMFM-66 section D</td> <td data-bbox="1740 467 1955 545">3.5 (3.4)</td> </tr> <tr> <td data-bbox="1520 545 1734 618">GMFM-66 section E</td> <td data-bbox="1740 545 1955 618">3.8 (3.5)</td> </tr> </table> <table border="1" data-bbox="1520 618 1955 1471"> <tr> <td data-bbox="1520 618 1734 1471">GMAE</td> <td data-bbox="1740 618 1955 1471">2.5 (1.1)</td> </tr> </table> | GMFM-66 | | Mean (SD) of change scores: final score - baseline score for each participant; significance not reported for this data. | | GMFM-66 section D | 3.5 (3.4) | GMFM-66 section E | 3.8 (3.5) | GMAE | 2.5 (1.1) |
| GMFM-66 | | | | | | | | | | | | | | | | |
| Mean (SD) of change scores: final score - baseline score for each participant; significance not reported for this data. | | | | | | | | | | | | | | | | |
| GMFM-66 section D | 3.5 (3.4) | | | | | | | | | | | | | | | |
| GMFM-66 section E | 3.8 (3.5) | | | | | | | | | | | | | | | |
| GMAE | 2.5 (1.1) | | | | | | | | | | | | | | | |

| | | | | ↑ as tolerated;) Final = 1.10 (0.38) m/sec. | | | | |
|---|---|---|--|---|--|--|---|--|
| Pseudo-RCTs | | | | | | | | |
| †Capodaglio et al 2011 ⁽²¹¹⁾ | <p>Ss: Participants with PWS = 11 (gender not reported) 33.8 (4.3) yrs. BMI 43.3 (5.9)</p> <p>Ss of sub-gps of participants with PWS: Gp1 = 6 BMI: Pre: 40.38 (3.46) Post: 42.57 (4.92); p > 0.05 Gp 2 = 5 BMI: 42.54 (7.69) vs Post: 38.35 (2.13); p > 0.05 Control gp (age-matched healthy non-obese participants) Ss=20 (10 males); 30.5 (5.3)yrs BMI 21.6 (1.6)</p> | <p>ID: PWS; mild ID (24/30 cut-off for MMSE Italian version). GMS: Not described.</p> | <p>Recruitment: Clients admitted to authors' rehabilitation hospital enrolled in study. Allocation: not described.</p> | <p>Interventions: Initial 2/52 hospital rehabilitation programme (rehab) for all PWS; Subsequent 6/12 HEP for Gp 1 (Gp 2 had no intervention). Setting: rehabilitation hospital; home Type: Rehab: exercises (same as HEP below), education. HEP: Heel walking 4m x10 reps; closed chain lower limb strength exercises 3 x 15 reps. Duration: Rehab: exercises 4x/wk, for 2 wks; HEP: 3x/wk for 6 mths; Support: Educational talk at start of rehab programme explaining clinical issues and rehabilitation. Completion of daily journal of adherence. Progression: Nil</p> | <p>Adherence: Rehab: not reported. HEP: 90% sessions completed. Adverse events: Not reported. Attrition: Nil.</p> | <p>Data extracted: repeated measures results for Standing Balance measures Data reported: Base-line data: for CG; for all participants with PWS (All PWS); separate data for sub-groups of participants with PWS not reported. Interim results data: results post 2/52 rehab; for PWS participants only (All, Gp 1 & Gp 2); results compared to CG baseline results. Final results data: results following 6/12 HEP; for PWS participants only (Gp 1 & Gp 2).</p> | | |
| | | | | | | | Standing balance Range: Medial-Lateral | Base-line CG = 9.36 (3.53) All PWS = 14.79 (9.53) [p<0.05 vs CG] Interim All PWS = 12.59 (5.21) [p<0.05 vs CG] Gp 1 = 15.83 (3.13) Gp 2 = 13.97 (3.14) Final Gp 1 = 16.58 (4.1) Gp 2 = 18.24 (5.6) |
| | | | | | | | Standing balance Range: Anterior-Posterior | Base-line CG = 5.03 (2.65) All PWS = 19.04 (6.76) [p<0.05 vs |

| | | | | | | | |
|--------------------------------------|---|---|--|--|---|--|--|
| | | | | | | | CG] Interim All PWS = 17.67 (5.24) [p<0.05 vs CG] Gp 1 = 14.9 (3.2) Gp 2 = 18.43 (3.9) Final Gp 1 = 14.75 (4.48) Gp 2 = 17.68 (3.98) |
| | | | | | | Standing balance Sway Path | Base-line: CG = 201.33 (45.86) All PWS = 573.58 (86.19) [p<0.05 vs CG] Interim All PWS = 513.03 (80.90) [p<0.05 vs CG] Gp 1 = 498.74 (70.26) Gp 2 = 527.32 (91.18) Final Gp 1 = 469.53 (58.67) Gp 2 = 506.63 (90.92) |
| †Vismara et al 2010 ⁽¹⁸⁷⁾ | Ss: Participants with PWS = 11 (gender not reported) 33.8 (4.3) yrs. BMI: 43.3 (5.9) | ID: PWS; mild ID (24/30 cut-off for MMSE Italian version). GMS: Independent walking; no gait aides used. | Recruitment: Clients admitted to authors' rehabilitation hospital enrolled in study. Allocation: not | Interventions: Initial 2/52 hospital rehabilitation programme (rehab) for all PWS; Subsequent 6/12 HEP for Gp 1 (Gp 2 had no intervention). | Adherence: Not reported. Adverse events: Not reported. Attrition: Nil. | Data extracted: repeated measures results for Gait parameters Base-line data: as above Interim data: as above Final data: as above; compared to Interim results, non-significant improvements found (exact significance not reported) | |

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|----------------------------------|---|--|------------|--|--|--|--|
| | <p>Ss of sub-groups of participants with PWS: (demographic data not reported) Gp1 = 6 Gp 2 = 5</p> <p>Ss of sub-gps of participants with PWS: Gp1 = 6 Gp 2 = 5</p> <p>Results reported: Control gp (age-matched healthy non-obese participants) Ss=20 (gender not reported); 28.4 (7.8) yrs BMI 21.6 (2.7).</p> | | described. | <p>Setting: rehabilitation hospital; home</p> <p>Type: Rehab: exercises (same as HEP below), education. HEP: Heel walking 4m x10 reps; closed chain lower limb strength exercises 3 x 15 reps.</p> <p>Duration: Rehab: exercises 4x/wk, for 2 wks; HEP: 3x/wk for 6 mths;</p> <p>Support: Educational talk at start of rehab programme explaining clinical issues and rehabilitation. Completion of daily journal of adherence.</p> <p>Progression: Nil</p> | | <p>Gait Cadence (steps/min)</p> <p>Gait Velocity (m/sec)</p> | <p>Base-line CG = 129.8 (4.8) All PWS = 113.89 (9.30) [p<0.05 vs CG]</p> <p>Interim All PWS =112.78 (10.24) [p<0.05 vs CG] Gp1: 111.8 (7.5); Gp 2: 113.88 (13.73)</p> <p>Final Gp 1: 117.0 (65) [p=0.02] Gp 2: 118.0 (10.18)</p> <p>Baseline CG = 1.2 (0.2) All PWS = 1.03 (0.12) [p<0.05 vs CG]</p> <p>Interim All PWS =1.03 (0.14) [p<0.05 vs CG] Gp 1 = 1.04 (0.16) Gp 2 = 1.01 (0.12)</p> <p>Final Gp 1 = 1.08 (0.16) Gp 2 = 1.03 (0.13)</p> |
| Repeated measures studies | | | | | | | |

| | | | | | | | | | | | | | |
|---|---|---|---|--|--|---|---|---|---|---|---|--|--|
| <p>‡Aguiar et al 2008⁽¹⁶⁹⁾</p> | <p>Ss = 21 (21 male) 23.3 (2.1) yrs. BMI: Pre: 23.0 (1.2); Post: 22.0 (2.8) (normal range)</p> | <p>ID: DS; severity not described. GMSs: Not described. Other: No atlanto-axial instability on radiological assessment.</p> | <p>Recruitment: not described. Allocation: N/A.</p> | <p>Setting: not stated Type: Adapted Judo training; aerobic training (monitored on lactate threshold) 50 min/session, 3x/wk Duration: 16 wks (total 2400 minutes) Support: Adapted Judo training taught by a physiotherapist. Progression: Not described</p> | <p>Adherence: ≥ 80% by all participants. Adverse events: Participants did not sustain any 'severe mechanical injuries'. Attrition: Nil.</p> | <p>GMFM-88</p> | <p>Results (median, range) reported as box and whisker diagram without exact numerical data; [<i>p</i><0.05]</p> <p>Pre: 65 (median) Post: 81 (median)</p> | | | | | | |
| <p>Kurz et al 2013⁽²¹²⁾</p> | <p>Ss = 8 (2 males) 16.3 (5) yrs. BMI: not reported.</p> | <p>ID: diagnosis (no. of participants) CP (3), chromosome disorders (2), Rett syndrome (2), brain injury in infancy (1); severe-profound ID. GMS: GMFCS levels II or III. Able to walk ≥ 10 m +/- gait aide. Other: Aides used (no. of participants): anterior walker (3); posterior walker (3); ankle-foot orthoses (6); thoraco-lumbo-sacral orthosis (1); shoe inserts (1).</p> | <p>Recruitment: From Special School for youth with severe-profound ID. Allocation: N/A.</p> | <p>Setting: Special school Type: BWS gait training (using a mobility frame with body weight-supporting harness on it, allowing participant to traverse overground) along 27m hallway; BWS system pushed along with participant. 20 min walking with 1-2 rests, 2 days/wk (1 day rest between). Duration: 12 wks. Support: Verbal encouragement given for BWS training; overground gait training integrated into usual school routines. Progression: Initial BWS = 40%; BWS ↓ by 5% fortnightly.</p> | <p>Adherence: 94% +/- 0.03% sessions attended Adverse events: Nil Attrition: Nil.</p> | <p>Data extracted for all participants (All) [significance reported (or MID) for results] Stratified results extracted: Youth (aged 16-21yrs; Ss=5); Children (aged 9-10yrs; Ss=3) [significance not reported]:</p> <table border="1" data-bbox="1522 917 1957 1468"> <tr> <td data-bbox="1522 917 1734 1128"> <p>Preferred Walk Speed: (m/sec)</p> </td> <td data-bbox="1743 917 1957 1128"> <p>Pre = 0.51 (0.21) Post = 0.67 (0.28) [33% improvement; exceeds MID⁽²²⁷⁾] Youth = 45% ↑ Children = 16% ↑</p> </td> </tr> <tr> <td data-bbox="1522 1135 1734 1281"> <p>Non-dimensionalized velocity (gait)</p> </td> <td data-bbox="1743 1135 1957 1281"> <p>All: Pre = 0.19 (0.7) Post = 0.25 (0.10) [<i>p</i><0.01]</p> </td> </tr> <tr> <td data-bbox="1522 1287 1734 1468"> <p>Cadence (gait) (steps/min)</p> </td> <td data-bbox="1743 1287 1957 1468"> <p>All: Pre = 37.8 (7.2); Post = 43.2 (8.4); [<i>p</i>=0.04, <i>d</i>=0.94]. Youth = 15% ↑</p> </td> </tr> </table> | | <p>Preferred Walk Speed: (m/sec)</p> | <p>Pre = 0.51 (0.21) Post = 0.67 (0.28) [33% improvement; exceeds MID⁽²²⁷⁾] Youth = 45% ↑ Children = 16% ↑</p> | <p>Non-dimensionalized velocity (gait)</p> | <p>All: Pre = 0.19 (0.7) Post = 0.25 (0.10) [<i>p</i><0.01]</p> | <p>Cadence (gait) (steps/min)</p> | <p>All: Pre = 37.8 (7.2); Post = 43.2 (8.4); [<i>p</i>=0.04, <i>d</i>=0.94]. Youth = 15% ↑</p> |
| <p>Preferred Walk Speed: (m/sec)</p> | <p>Pre = 0.51 (0.21) Post = 0.67 (0.28) [33% improvement; exceeds MID⁽²²⁷⁾] Youth = 45% ↑ Children = 16% ↑</p> | | | | | | | | | | | | |
| <p>Non-dimensionalized velocity (gait)</p> | <p>All: Pre = 0.19 (0.7) Post = 0.25 (0.10) [<i>p</i><0.01]</p> | | | | | | | | | | | | |
| <p>Cadence (gait) (steps/min)</p> | <p>All: Pre = 37.8 (7.2); Post = 43.2 (8.4); [<i>p</i>=0.04, <i>d</i>=0.94]. Youth = 15% ↑</p> | | | | | | | | | | | | |

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| | | | | <p>Training speed (m/sec): Initial (average speed of first 4 sessions): 0.3 +/- 0.04; Final (average speed of last 4 sessions) 0.54 +/- 0.03 (80% ↑ vs initial speed [p<0.001, d=0.80]).</p> | | | <p>Children = 9% ↑</p> |
| | | | | | | <p>OGS (mean pre-post test change)</p> | <p>All: Pre = 11.4 (3.4); Post = 11.8 (3.5); [p=0.16; Cohen's d=0.28]. Youth = 13% ↑ Children = 12% ↓</p> |
| | | | | | | <p>SWAPS (mean pre-post test change)</p> | <p>All: Pre = 69.1 (11.1) Post = 71.2 (17.2) [p=0.28; Cohen's d=0.36] Youth = 11% ↑ Children = 12% ↓</p> |
| | | | | | | <p>GMFM-88 Section E (mean pre-post test change)</p> | <p>All: Pre = 18.1 (14.3); Post = 18.5 (14.7); [p=0.15; Cohen's d=0.09]. Youth = 2% ↑ Children = 2% ↑</p> |
| Case report | | | | | | | |
| Berg et al 2012 ⁽²⁰⁹⁾ | <p>Ss = 1 (male) 12yo. Weight (lbs) Pre: 108.0 Post: 108.6 (normal range)</p> | <p>ID: DS; mild ID. GMS: Not described.</p> | <p>Recruitment: Via advertising on listservs for two DS awareness groups. Attending public school. Allocation: N/A</p> | <p>Setting: Home. Type: Nintendo Wii games, participant's choice of games, ≥20 min/session, ≥4x/wk. Participant chose 4 games (bowling, baseball, rhythm boxing, snowboarding) Parents kept log of</p> | <p>Adherence: Wii games chosen = 4 Achieved total practice time over 4 wks = 547 min (average 68 min/week) Adverse events: Nil reporting regarding any adverse events.</p> | <p>Standing Balance (Biodex Portable BioSway Balance System) [improvement if score ↓]</p> | |
| | | | | | | <p>Overall Stability Index</p> | <p>Pre 1.39, Post 1.00</p> |
| | | | | | | <p>Anterior / Posterior Index</p> | <p>Pre 1.39, Post 0.64</p> |
| | | | | | | <p>Medial / Lateral Index</p> | <p>Pre 0.53, Post 0.94</p> |

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|--|--|--|--|--|------------------------|--|---|
| | | | | therapy engagement. Duration: 8 wks (total 640min). Support: Physiotherapist made fortnightly phone or email contact with parents. Progression: Nil. | Attrition: Nil. | BOT-2 (z score) | |
| | | | | | | Subtest Scaled Score: Balance | Pre 5 (-2.0), Post 6 (-1.8) <i>Exceeded MID⁽²³⁴⁾</i> |
| | | | | | | Subtest Scaled Score: Running speed and agility | Pre 7 (-1.6), Post 8 (-1.4) <i>Exceeded MID⁽²³⁴⁾</i> |
| | | | | | | Composite Standard Score: Body Coordination | Pre 36 (-1.4), Post 33 (-1.7) <i>Exceeded MID⁽²³⁴⁾</i> |

Legend:

Ss = sample size; BMI = body mass index; GMS = gross motor skills; ID = intellectual disability; DS = Down syndrome; CP = cerebral palsy; PBWSTT = partial body-weight supported treadmill training; PWS = Prader-Willi syndrome; MMSE = Mini-Mental State Examination; HEP = home exercise programme; BWS = body-weight supported; MID = minimum important difference; ITT = intention to treat; GMFCS = Gross Motor Function Classification Scale; GMFM = Gross Motor Function Measure; GMAE = Gross Motor Ability Estimator; IG = intervention group; CG = control group; OGS = Observational Gait Scale; SWAPS = Supported Walker Ambulation Scale; BOT-2 = Bruininks-Oseretsky Test of Motor Proficiency, 2nd edition

* = author communication confirmed the Capodaglio et al⁽²¹¹⁾ and Vismara et al⁽¹⁸⁷⁾ studies were for the same interventions conducted at the same time (section 3.6);

† = lack of exact numerical data in study; exact numerical data (mean (SD)) for interim and final results provided by corresponding author;

‡ = lack of exact numerical data in study; numerical median results calculated from raw data provided by corresponding author.

Table 5: Validated GMS outcome assessment tools used in studies

| Study | ID Diagnosis; Mean age (yrs) | GMS outcome assessment tools | Prior reported validity testing (type of testing; clinical population) |
|---|---------------------------------|--|--|
| RCTs | | | |
| Shields et al 2008 ⁽²¹⁰⁾ | DS 26.8 yrs. | Timed up and down stairs test (sec) | Validated for typically developing children & children with CP, ⁽²¹⁴⁾ and for adults with MS. ⁽²¹⁵⁾ Systematic review of normative values in different clinical populations; DS included in calculations for neurological conditions. ⁽²¹⁶⁾ |
| Su et al 2013 ⁽¹⁶⁵⁾ | Non-spastic CP 10.8 yrs | GMFM-66 section D : dimension % score of 39 possible points | GMFM-66 criterion standard ⁽²¹⁷⁾ against which other paediatric assessments are compared for validity. ⁽²¹⁸⁾ Use of a limited number of items from the GMFM-66 previously established as valid for predicting overall GMFM-66 scores; ⁽²¹⁹⁾ however this study did not assess sections D and E specifically. [However, in for the longer GMFM-88 sections D and E used as criterion standard when testing other assessment tools in children with CP. ⁽²²⁰⁾ |
| | | GMFM-66 section E : dimension % score of 72 possible points | |
| | | GMFM-66 GMAE : composite score calculated from GMFM-66 D & E results | |
| Pseudo-RCTs | | | |
| Capodaglio et al 2011 ⁽²¹¹⁾ | PWS 33.8 yrs | Balance Standing for 60 seconds on force platform (Kistler, CH; acquisition frequency: 500 Hz) with integrated video, measuring Component of Displacement (CoP) in mm. Results normalised to participant's height. CoP measures : uni-directional measures - Medial-Lateral - Anterior-Posterior : two-dimensional measure - total CoP Sway Path | Force platform technique described ⁽²²¹⁾ & assessed for its correlation against alternative commonly used assessment tool. ⁽²²²⁾ Normalisation of results to participant's height previously reported; additionally, Sway Path found to be a more robust measure, however uni-directional measures can identify specific clinical issues. ⁽²²³⁾ |
| Vismara et al 2010 ⁽¹⁸⁷⁾ | PWS 33.8 yrs | Cadence (steps/min) Video analysis: participant walking at comfortable pace. Opto-electronic video, and force platforms embedded centrally in walkway. Distance of | 3-d video considered as gold standard for gait analysis. ⁽²²⁴⁾ Similar assessment approach incorporating video analysis and |

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| | | walkway & distance walked for assessment not reported. | embedded force meter platform used with DMD. ⁽²²⁵⁾ |
| | | Gait velocity (m/sec) (Video analysis: as above) | |
| Repeated measures studies | | | |
| Aguiar et al 2008 ⁽¹⁶⁹⁾ | DS 23.3 yrs | GMFM-88 | Validity tested for children with DS in regard to: motor function; ⁽¹⁴¹⁾ motor development. ⁽²²⁶⁾ |
| Kurz et al 2013 ⁽²¹²⁾ | Neuromuscular impairments (50% with spastic CP). 16.3 yrs | Cadence (steps/min) Video analysis: participant walking along 16m walk-way; result calculated from no. of steps taken over mid 6m of walk way. | 3-d video considered as gold standard for gait analysis; validity tested. ⁽²²⁴⁾ MID values for children with CP. ⁽²²⁷⁾ |
| | | Preferred Walk Speed (m/s) Video analysis: participant walking along 16m walk-way; result calculated from time taken to traverse middle 6m of walk way. Results compared to MID values. | |
| | | Non-dimensionalized gait velocity : calculated from individual results for Preferred Walk Speed normalised to leg length. | Rationale for normalisation conversion. ^(228, 229) Gait biomechanics in youth aged 7 to 18 (mean 12.9) yrs. ⁽²³⁰⁾ |
| | | OGS : visual assessment of various components of lower limb gait biomechanics a : max score of 22 for each leg indicates normal gait. | Children with spastic diplegic CP: ⁽²³¹⁾ validity and inter- and intra-rater reliability acceptable for knee and foot position, lower for hind-foot and base of support. |
| | | SWAPS : 4-point Likert scale for gait posture, support and lower limb stepping biomechanics. | Initial development and preliminary testing of SWAPS in toddlers with spastic CP: ⁽²³²⁾ high interrater reliability; good construct validity when compared to GMFM. |
| | | GMFM-88 Section E : expressed as % score of 72 possible points | GMFM-88 (D and E) used as criterion standard when testing other assessment tools in children with CP. ⁽²²⁰⁾ |
| Case report | | | |
| Berg et al | DS | Balance (Biodex BioSway Balance System) (computerised force platform providing varying degrees of stability/ perturbations of the | Test-retest reliability assessed as acceptable for clinical use. ⁽²³³⁾ |

| | | | |
|-----------------------|--------|---|---|
| 2012 ⁽²⁰⁹⁾ | 12 yrs | supporting surface): Overall Stability Index; Anterior/Posterior Index; Medial/Lateral Index | |
| | | BOT-2 Subset Scaled Scores: - Balance - Running speed and agility | Reliability and responsiveness tested, and MID and MDC established. ⁽²³⁴⁾ Adapted for 4-18yo with an ID. ⁽²³⁵⁾ |
| | | BOT-2 Composite Standard Score: - Body Coordination | |

Legend:

ID = intellectual disability; DS = Down syndrome; CP = cerebral palsy; PWS = Prader-Willi syndrome; MS = multiple sclerosis; DMD = Duchenne muscular dystrophy; GMFM = Gross Motor Function Measure; GMAE = Gross Motor Ability Estimator; MID = minimum important difference; MDC = minimum detectable change; OGS = Observational Gait Scale; SWAPS = Supported Walker Ambulation Scale; BOT-2 = Bruininks-Oseretsky Test of Motor Proficiency, 2nd edition.

3.11 **GMS outcome assessment results**

All of the results extracted in this study showed some amount of improvement in the GMS outcomes measured; however, results varied in whether they were statistically significant or not.

A range of GMSs, predominantly representative of gait, standing balance, as well as composite assessment of general gross motor function (tables 4 and 5), were reported in the included studies.

3.11.1 Gross Motor Function Measure (GMFM)

General gross motor function was assessed in three studies using the GMFM, a multi-item visual assessment checklist which assesses a wide array of GMSs. Two of the subsidiary sections of the GMFM, section D (which assesses standing abilities), and/or section E (which assesses walking, running and jumping), have previously been analysed against levels of physical activity in youth with spastic diplegic cerebral palsy.⁽²³⁸⁾ It was found that section E is a better predictor of physical activity levels, whereas section D better indicates prevalent levels of physical activity in this population.⁽²³⁸⁾

Results for two versions of the GMFM were reported in the studies: either its full version using the larger 88-item form (GMFM-88)^(169, 212) or its abridged version with 66 items (GMFM-66).⁽¹⁶⁵⁾ Subsidiary sections of the GMFM were used^(165, 212) (specifically section D for standing, and/or section E for walking, running and jumping) (tables 4 and 5).

Improvements in GMFM scores were reported in three studies. Aguiar et al⁽¹⁶⁹⁾ reported significantly improved results for the GMFM-88 following an adapted Judo training intervention. Su et al⁽¹⁶⁵⁾ found improvement in standing ability (GMFM-66 Section D) following PBWSTT; this result was reported as a percentage change score within a cross-over design study and so statistical significance was not assessed. Results for GMFM section E (locomotion) scores were reported as percentage change scores in two studies: Su et al⁽¹⁶⁵⁾ (GMFM-66 tool) and Kurz et al⁽²¹²⁾ (GMFM-88 tool). Both of these studies utilised similar gait training interventions (either PBWSTT⁽¹⁶⁵⁾ or BWS overground training⁽²¹²⁾

respectively), for similar study populations (table 4). Both studies reported improvements: however, in the study by Kurz et al⁽²¹²⁾ the results were not significant, and assessment of significance was not reported in the study by Su et al.⁽¹⁶⁵⁾

3.11.2 Gait

Different parameters of gait were considered in the studies by Kurz et al⁽²¹²⁾ and Vismara et al.⁽¹⁸⁷⁾ Cadence was shown to improve significantly following lower limb strengthening (Vismara et al⁽¹⁸⁷⁾) and BWS gait training (Kurz et al⁽²¹²⁾). The study by Kurz et al⁽²¹²⁾ also showed a 33% improvement in preferred walking speed which exceeded the minimum important difference (MID). Statistical significance was not assessed for this finding; however, non-dimensionalized velocity, which was calculated from this result, was found to be significantly improved. In this study, lower limb gait biomechanics measured using two observational checklist scales also improved, but results did not achieve statistical significance.⁽²¹²⁾ The study by Vismara et al⁽¹⁸⁷⁾ reported on a number of measures for different components of gait; only data which reflected gait in its entirety⁽¹⁸⁷⁾ were extracted (table 4). In this study, gait velocity showed only minimal improvement and significance was not reported for this result.

3.11.3 Balance

Improvements in standing balance were reported in two studies. In Capodaglio et al,⁽²¹¹⁾ changes in static standing postural sway were assessed by measuring the path distance of postural sway in different directions; non-significant improvements were identified. In Berg et al⁽²⁰⁹⁾, the effect of Wii games on standing balance was assessed. Improved results were found for the Bruininks-Oseretsky Test of Motor Proficiency measure, second edition (BOT-2),⁽²³⁴⁾ which exceeded the MID, and for static standing balance measured using the Biodex Balance system.

3.11.4 Other weight-bearing skills

The effect of Wii games on motor skill proficiency, specifically running (speed and agility) and body coordination, were measured by the BOT-2 in Berg et al.⁽²⁰⁹⁾ Improved scores were reported for these skills and results exceeded MID thresholds indicating an effective response for their case report study.⁽²⁰⁹⁾ Stair climbing was reported in an RCT that investigated the effectiveness of strength resistance training, and found a non-significant improvement for the intervention group when the between-group difference was analysed (Aguar et al⁽²¹⁰⁾).

4 Discussion

To date, this is the first systematic review which has investigated the effectiveness of physiotherapy interventions for improving gross motor skills in people with an intellectual disability aged six years and older. This review identified a range of physiotherapy interventions which were provided to adolescent and adult participants. The interventions were administered in various settings including an inpatient rehabilitation hospital, a community gym, and participants' homes. A number of the interventions did not require either complex or expensive equipment. The studies reported high rates of participant retention and adherence to the interventions, and negligible adverse events. Such factors indicate the potential for these interventions to be considered for physiotherapy practice.

Three of the interventions resulted in statistically significant improvements in GMS outcomes: physiotherapist-led adapted Judo training (improvement in the GMFM-88);⁽¹⁶⁹⁾ lower limb weight-bearing strengthening exercises (improvement in cadence);⁽¹⁸⁷⁾ and BWS overground gait training (improvement in cadence and non-dimensionalized gait velocity).⁽²¹²⁾ These major findings can be considered in physiotherapy clinical practice. Although the other studies included in this review also reported improvements in GMSs, these other results did not reach statistical significance and, accordingly, the capacity for extrapolating these findings to physiotherapy practice is therefore limited.

This review's broad inclusion criteria enabled identification of all relevant studies in what is a sparse field of clinical research. However, despite this, strong clinical recommendations could not be formulated. This was mainly due to the high level of heterogeneity of the interventions and outcomes reported in the studies, which precluded the use of statistical pooling for meta-analysis, and the low quality of evidence represented by the included studies, of which only two were RCTs. A previous systematic review of early intervention programmes for children with physical disabilities⁽¹⁷⁵⁾ also utilised broad-ranging PICO criteria. That review similarly identified high heterogeneity between their selected papers in regard to the range of study designs used, levels of study quality, and types of results reported.

4.1 Narrative review

As the high heterogeneity between studies in the current review prevented meta-analysis, a narrative approach was instead utilised to synthesize the extracted results. This approach reflects the nature of a broad-ranging systematic review in coalescing a wide sphere of research and contextualizing a new field. A high rate of narrative syntheses have been found amongst public health reviews, which seek to more broadly appraise the influencing factors on a given health issue in a population.⁽²³⁹⁾ Although narrative syntheses are not able to clearly establish the existence of effects as is possible in meta-analyses, they do play an important role in clarifying research evidence in emerging fields of study, and can provide an initial evidence platform from which more specifically structured primary or secondary studies can be framed and developed.

4.2 Overview of the research field

4.2.1 Interventions to improve GMSs well tolerated

Studies included in this review reported negligible adverse events and excellent participant retention and adherence. This is a particularly positive finding of this review as concentration, motivation, and learning ability all impact on tolerance and engagement and can often be impaired in people with an intellectual disability.^(240, 241) Finding interventions which are safe, effective and optimise participant engagement is important when working with clients with an intellectual disability. Additionally, safety is a key consideration as people with an intellectual disability more often have difficulty reporting discomfort or pain arising during an intervention.⁽¹⁹⁵⁾

4.2.2 Level of engagement in physical activity (secondary outcome) not assessed

It is of considerable interest that study participants' level of engagement in physical activity, the secondary outcome for this review, was not reported in any of the included studies. This is particularly so given that increased levels of physical activity have been shown to help stave off secondary deterioration and

progressive disability⁽¹⁶⁵⁾ which may result from inactivity (table 6). Interestingly however, some of the studies informally discussed the importance of improving particular GMSs in order to increase general physical activity, and the need for longer term follow-up of participants' integration of improved GMSs into daily function and activities (table 6).

4.2.3 Lack of reporting of rehabilitation measures following acute medical condition

None of the studies investigated rehabilitative physiotherapy interventions for regaining GMSs lost due to acute medical or surgical conditions. This was surprising given that individuals with an intellectual disability, as with the general population, experience acute clinical needs relating to orthopaedic surgery, cardiac failure or arrest, infective and/or obstructive respiratory disease, and cerebro-vascular accident and other central nervous system diseases.⁽²⁴²⁻²⁴⁴⁾ All of these conditions can result in acquired GMS deficits and physical impairments such as impaired balance, gait, strength, or coordination. There is much physiotherapy and multi-disciplinary research reporting on rehabilitative interventions for promoting recovery after acute injury or illness in study populations recruited from the general population. These studies report the effectiveness of interventions provided in acute and sub-acute healthcare settings.⁽²⁴⁵⁻²⁴⁸⁾ People with an intellectual disability may similarly require health care within an acute hospital for such conditions. During these periods, sensitive management of the communication between the client and the health care providers is needed in order to ensure optimal care for a person with an intellectual disability.⁽²⁴⁹⁾ The dearth of primary research evidence could result in considerable short-falls in the quality of care for people with an intellectual disability.

4.2.4 Study design considerations

The overall lack of research identified by this review may be due to the difficulty in obtaining large enough sample sizes to undertake studies with adequate statistical power. Challenges can exist when planning primary research intervention studies for participants with an intellectual disability in regard to study design. These include the difficulty of conducting randomized allocation when it is preferable that

participants' specific learning or behavioural needs are matched carefully with specific interventions rather than randomly.⁽¹⁸⁷⁾ As well, there can be difficulty in recruiting large numbers of participants. These challenges may in part explain the limitations of the types of study designs found in the present review (small sample sizes, a majority of observational study designs, and convenience sampling), which resulted in a reduced ability to draw strong conclusions.

A pragmatic short-term solution to the current lack of sufficient quality studies could be addressed through the reporting of findings from well-designed case reports, similar to the Berg et al⁽²⁰⁹⁾ paper which reports clear inclusion criteria, intervention, and outcome measurements as well as statistical analyses.

4.2.4.1 Use of convenience sampling

Convenience sampling, which was utilised in five of the seven studies, contains an inherent risk of bias which weakens the reliability of the results. Convenience sampling may include the study population being recruited from a single facility or a limited geographical area. Additionally, the study authors may have some rapport with the study population. These relational factors may affect the interactions between the investigators and the participant, and even how the study is designed. As such, the effectiveness of the intervention may be influenced compared to if the investigators had no prior knowledge of the study participants. All of these factors contribute to the likelihood that a study population recruited using convenience sampling will not be representative of the general clinical population. It is this lack that can skew the study's results, positively or negatively, depending on the circumstances.

Nonetheless, for newer fields of clinical research for people with an intellectual disability, convenience sampling may still provide a reasonable platform from which to develop a study project. Reasons for this may include there being a greater number of potential study participants in a specialised facility or clinical catchment area, the investigators having prior rapport with the potential participants and their caregivers, or the local ethics committee having knowledge of the participants and their needs. As such, although convenience sampling weakens the results of a study, while the current field of research regarding

physiotherapy for people with an intellectual disability remains small, there is an apparent need to consider these studies as important sources of research evidence.

4.2.4.2 *Possible reasons for the use of convenience sampling*

The frequent use of convenience sampling is likely due to a range of barriers that exist when recruiting persons with an intellectual disability into research studies⁽²⁵⁰⁾ (section 4.2.4). These barriers include difficulty recruiting large numbers of participants,⁽²⁵⁰⁾ with additional challenges relating to careful consent processes for recruitment.⁽²⁵¹⁾ As well, there can be a need for a strong sense of trust by the participant and/or care-giver that the research is for the study population's best interest in order to overcome any sense of taking advantage of persons in a disadvantaged position.⁽²⁵⁰⁾ These factors can be accommodated where possible by the use of appropriately designed consent processes which are inclusive of participant involvement.⁽²⁵¹⁾ Recruitment processes which supportively engage participants and their carers in learning about the study may facilitate a degree of random sampling. Additionally, Modern Ethics Committee approval processes include a rigorous appraisal of the study design by requiring researchers to submit a thorough ethics application, such as the Australian National Ethics Application Form,⁽²⁵²⁾ in support of the research proposal.

4.2.4.3 *Small sample sizes*

Despite all of the studies having small sample sizes (the number of participants in the studies ranging from one to 21), which contributed to reduced statistical power in the results, none of the included studies addressed or explained this limitation. In future research it would be advantageous for authors to briefly explain the difficulties of recruiting larger numbers of people with an intellectual disability for clinical studies, in order to help establish the challenges of undertaking high quality research in this population,⁽²⁵³⁾ and spur efforts to overcome these challenges. Additionally, referral to normative data where appropriate would assist with judging clinical applicability.

4.2.4.4 *Reporting on statistical significance*

Appropriate statistical significance analysis was carried out and reported for the extracted data in the majority of included studies. For two of the studies that did not report on the statistical significance of the extracted findings, there were clear justifications for their absence. In the first of these studies, a two-period cross-over RCT (Su et al⁽¹⁶⁵⁾), findings were reported for two groups of participants receiving two different physiotherapy interventions. Statistical assessment appropriately compared the effectiveness of the interventions and the carry-over effects due to time sequencing, but understandably did not analyse differences between repeated measures within each group. For the purposes of the current review, in accordance with the requirements of the *a priori* protocol, only pre- and post-intervention results for the first group for the primary intervention of interest (PBWSTT) were of interest. As such, analyses of statistical significance were not available for the extracted results. In the other study, a case report (Berg et al⁽²⁰⁹⁾), results for a range of outcome assessments results (BOT-2 scores for balance, body coordination and running speed and agility) were reported with z-scores and compared to minimum detectable change (MDC) and MID normative values.⁽²⁰⁹⁾ These analyses were appropriate for a case report. However, the results for the Biodex BioSway balance assessments in this study were presented simply without statistical analysis. Caution should therefore be applied if extrapolating these findings to clinical practice.

Some of the studies reported non-specific results for significance.^(169, 187, 211) Future research should ensure exact statistical reporting, as, even if the results were non-significant and/or were findings for a small sample size, such reporting would facilitate data extraction in systematic reviews and the potential inclusion of results into meta-analysis.

4.2.5 International representation in included studies

Representation of a number of developed countries from where the studies originated was found in this review. A previous systematic review of mobility in adults with an intellectual disability⁽¹²⁴⁾ similarly found

that all studies arose from developed nations. Despite the comparative lack of representation of research from developing nations in the current review, the results can be considered in developing world settings. Some of the simpler assessment and intervention approaches which did not require computerised or expensive equipment could be adopted in less developed nations in which access to digital assessment devices or expensive assistive mobility aids may not be possible due to limited resources. This further supports the relevance of the current review's topic across the globe.

4.3 Consideration of participants' learning needs reported within studies

People with intellectual disabilities experience reduced independence in general life activities due to difficulties with learning and adaptive behaviour. These limitations in functional independence can be further exacerbated by the individual experiencing GMS deficits.⁽²⁵⁴⁾ Additionally, the person's learning ability to overcome these motor deficits is likely to be hampered by having an intellectual disability. Nonetheless, the findings of this review demonstrate that physiotherapy interventions for improving GMSs can be safely implemented for individuals with varying levels of severity of intellectual disability, including profound cognitive impairment, with some degree of improvement in GMSs resulting.

Unfortunately, none of the included studies reported on specific study design considerations which were implemented to meet the learning needs of the participants in regard to their intellectual disabilities. Instead, only general comments were made by the authors regarding participant's learning needs (table 6). A possible reason that these factors were not clearly considered and stated is that the included studies focused predominantly on the participants' physical impairments and activity limitations rather than upon appraising and addressing learning needs of participants. This potential issue may also underscore why there is a lack of reporting of levels of intellectual disability in physiotherapy studies of participants with developmental disabilities, such as cerebral palsy (appendix III); this was a main reason for studies being excluded following full-text review. In future research in which study participants have a developmental disability which commonly but not always results in having an intellectual disability (such as cerebral

palsy⁽⁸⁴⁾ or Autism Spectrum Disorder⁽⁸⁵⁾) authors should clearly report not only the type and severity of intellectual disability of participants but also the impact these disabilities have upon the ability of participants to participate in the intervention and assessments of the study. Study authors should also clearly explain the ways in which the study design addressed the cognitive needs of the participants, for example in regard to specific approaches used in delivering the interventions, how randomisation was conducted or even considered, and the types of assessments used. This level of reporting would support the development of more robust research paradigms for physiotherapy for people with an intellectual disability.

None of the studies found by this review tested a mainstream intervention against a modified version of the same intervention incorporating specific communication or teaching approaches designed to better meet the particular learning style need of study participants. Such intervention modifications could have included increased repetition when practising a GMS, greater simplicity in the type of verbal cues given for general teaching and coaching, decreasing the amount of verbal cues given and increasing the amount of non-verbal (visual and tactile) cues provided for feedback on performance.

4.3.1 Informal consideration of learning needs of participants

Despite the lack of explicit study design considerations regarding the learning needs of participants with intellectual disabilities, it can be concluded that the apparent needs of these participants in regards to their intellectual disabilities were informally reflected within the studies. Authors' general considerations (table 6) regarding the cognitive needs of participants were informally discussed in the studies. These considerations included the need for simple instructions, greater repetition of practice, predictability in exercise regimes, for manual handling issues to be addressed, and the need to consider participants' psychological factors (which may preclude being able to randomly allocate participants within an experimental study).

Although the general considerations discussed by the studies' authors were not formally assessed or measured within the study designs, they were reflected more generally in the types of interventions included. In two studies, the intervention included practice of an exercise programme over a lengthy period of six months, with no integrated progressions of the exercises.^(187, 211) This contrasts to usual physiotherapy practice of exercises being progressed in regard to one or more predetermined assessment criteria, particularly over such a lengthy timeframe. In the same two studies, participant engagement was supported by participants completing a daily journal of their adherence to the prescribed home exercise programme,^(187, 211) which has previously been shown to be an effective intervention for increasing client engagement.⁽²⁵⁵⁾ In another study, participant engagement was facilitated by allowing the participant to choose the type of therapy they performed, (in this case, which Wii games they played⁽²⁰⁹⁾) and then tailoring the outcome assessment to match the participant's choices of intervention. Future primary studies could offer participants a range of interventions from which they could choose which they would like to do. As well, keeping a record of adherence to the program, either in a journal, or via an alternative means such as a wall chart showing progress with reward features, could be utilised and measured for efficacy.

Table 6: Authors' considerations regarding impact of participants' intellectual disability on study design

| Authors / date | Author rationale for intervention with respect to GMS deficits | Author perspectives: Teaching and learning and ethical considerations for ID | Author conclusions following completion of study |
|--|--|---|--|
| RCTs | | | |
| Shields N, et al. (2008) | <p>Decreased muscle strength results in decreased function in adults with DS</p> <p>Improved strength and function expedites vocation and employment options.</p> <p>Strengthening exercises are needed to improve physical function</p> | <p>Exercises could be modified by trainer if participant found exercise difficult</p> <p>Close supervision of smaller sub-groups of 2-3 participants by trainers</p> <p>Transport to/from programme provided; control group offered strengthening programme after completion of the study</p> | <p>High compliance due in part to supportive training environment</p> <p>Improvements in stair climbing not significant; this may be in part due to minimal impairment in baseline muscle endurance in lower limbs</p> |
| Su IYW, et al. (2013) ⁽¹⁶⁵⁾ | <p>Risk of secondary functional deterioration in individuals with complex disabilities from CP needs to be minimised.</p> <p>Using assistive gait training devices for more physically dependant persons enables stepping practice whilst maintaining safe manual handling for therapists</p> <p>A trained assistant helped during interventions for participants with GMFCS levels IV and V</p> | <p>Instrument-supported gait practice enables children with severe complex disabilities opportunity to practice gait for extended periods.</p> <p>This repetition enables improved central neurological learning.</p> | <p>Lack of homogeneity and low sample size despite using a cross-over design precluded being able to determine significant results.</p> |
| Pseudo-RCTs | | | |
| Capodaglio P, et al. (2011) ⁽²¹¹⁾ | <p>Decreased balance in PWS may improve if leg strength is improved</p> <p>There is need for targeted lower limb exercises for improving postural balance, as discussed in their earlier research (Capodaglio et al 2011)⁽¹²⁸⁾</p> | <p>Completion of daily journal of adherence to programme</p> <p>Participants with PWS need predictability in interventions, therefore the exercise programme was not progressed</p> | <p>Improvement not significant. Insufficient intensity and task-specificity of exercises.</p> |
| Vismara L, et al. (2013) ⁽¹⁸⁷⁾ | <p>Relationship between walking ability and level of spontaneous activity in individuals with PWS</p> <p>Improvements in gait results in increased</p> | <p>Use of a daily adherence journal and simplicity of prescribed exercises to enhance long-term compliance</p> | <p>Significantly improved cadence;</p> <p>feasible that individuals with PWS, even those with specific psychological needs, can do long-term HEP</p> |

| | | | |
|--|---|--|---|
| | independence in daily activities Improving the level of walking can be addressed in rehabilitation | Random allocation not possible because individuals' psychological profile should be appraised and managed appropriately when choosing an appropriate intervention for participants with PWS | at low intensity with gains made; Non-randomized allocation and small Ss may be limitations for extrapolation of results |
| Repeated measures studies | | | |
| Aguiar Jr AS, et al. (2008) ⁽¹⁶⁹⁾ | Physical exercise can improve motor function in people with DS | Not discussed. | Significant improvement in motor function is of clinical importance; however, small sample size so caution with extrapolation of results |
| Kurz MJ, et al. (2013) ⁽²¹²⁾ | BWS gait training is task specific practice; it facilitates cortical neuroplasticity learning and improvement of gait due to high repetition of stepping practiced: Providing BWS gait training overground provides additional facilitation to this motor learning | Intervention done at school within usual daily program Pre- and post-intervention preferred walking speed was measured by taking the average result from the results for the initial and final four training sessions; this approach was done to overcome the inherent sources of variability in gait due to individuals in this population varying in regard to their motivation and physical function | Stratified results according to age not significant as small Ss, but further research still needed to determine what age best to do overground gait training Large effect sizes for some outcomes, but due to there being no long-term follow-up assessment nor assessment of community participation, it is unable to be determined whether there was any beneficial carry-over into daily function |
| Case report | | | |
| Berg P, et al. (2012) ⁽²⁰⁹⁾ | Increased physical fitness results in improved weight-bearing and functional abilities for people with DS Higher frequency of exercise practice enhances positive neural plasticity changes | Participant chose which Wii games he practised Parents and siblings encouraged to also participate Authors conducted motor control analysis of the Wii games and determined the motor skills which were likely to be influenced by practice of specific Wii games Outcome assessments were matched to the choice of Wii games practised by the participant | Outcomes showing improvement were related specifically to the types Wii games practised Extrapolation of results to clinical practise is limited due to this being a case report |

Legend:

ID = intellectual disability; GMS = gross motor skill; DS = Down syndrome; CP = cerebral palsy; PWS = Prader-Willi syndrome; GMFCS = Gross Motor Function Classification System; BWS = body-weight supported; HEP = home exercise programme; Ss = sample size.

4.4 Limitations of this review

Limitations of this systematic review included only considering studies published in English, and the potential for reporting bias. However, the presence of a number of papers which reported non-significant findings and small sample sizes suggest that reporting bias may not be in effect. Other limitations of the current review were the use of broad-ranging inclusion criteria and a low threshold score following critical appraisal, which resulted in greater heterogeneity in the selected studies. The low number of papers finally selected, and the varied sources of bias within these studies underscores the need to be cautious extrapolating the studies' findings to clinical practice even where there are statistically significant results.

4.4.1 Broad inclusion criteria

In the current review, the first known for this field of enquiry, the use of broad inclusion criteria enabled the identification of all relevant studies in what is a sparse area of clinical research. However, it not surprisingly also resulted in a high level of heterogeneity between the included studies which precluded the use of statistical pooling for meta-analysis. As such, the formulation of strong clinical recommendations from this review was impeded and would not support the creation of a best practice guideline for definitive therapy interventions and/or styles and approaches for delivering physiotherapy interventions in this clinical area. However, the low number of papers that were ultimately included demonstrates that the use of broad-ranging inclusion criteria was warranted.

4.4.2 Low threshold for inclusion of a study following critical appraisal

The current review had a cut-off threshold of four 'yes' responses; this inherently allowed a greater potential for bias to be present within the selected studies. However, a low critical appraisal threshold was chosen in order to optimise inclusion of studies from the small field of research, and to accommodate the expected limitations of this field (purposeful rather than random allocation of participants due to psychological needs, the need to use convenience sampling, and there being difficulty recruiting large

numbers of participants). The low inclusion threshold applied for critical appraisal, whilst contributing to weakening the overall strength of the statistical findings of this review, was clearly justified in light of the low numbers of papers which were ultimately selected.

4.5 Limited evidence base for supporting physiotherapy clinical interventions

The low number of papers and high level of heterogeneity of the studies in the current review demonstrated a serious lack of research evidence investigating physiotherapy interventions for this population for improving GMSs. Physiotherapy resources in this clinical field are usually limited, which can potentiate the tendency to provide adaptive rather than restorative or rehabilitative therapy to overcome the client's GMS deficits. A lack of research evidence undermines the confidence with which physiotherapists and consumers may choose and plan needed therapy.

This is of concern not only for physiotherapists when deciding on optimal interventions to use in therapy, but also for consumers. This is particularly so in the current context of limited overall resourcing of therapy services for people with an intellectual disability, and in regard to newer funding models for disability services. For example, funding for disability services in Australia has received considerable restructuring with the staged implementation of consumer-controlled funding,⁽¹⁰⁸⁾ under the NDIS⁽¹⁰⁷⁾ (section 1.7). Prior to the implementation of the NDIS, the government provided free therapy services to people with an intellectual disability for eligible clients. The NDIS funding scheme aims to improve the sector's efficiencies and expertise whilst enabling consumers and/or their carers to determine the amount and type of care and therapy services utilised.⁽¹⁰⁸⁾ This has resulted in increased numbers of private therapist services for clients with disabilities including intellectual disabilities. While this enables improved access to seeing a physiotherapist, the current lack of evidence supporting the effectiveness of physiotherapy interventions confounds the confidence in which therapy interventions can be chosen and implemented.

4.6 Recommendations for future research

A lack of research evidence jeopardizes the effective use of limited funding for therapy resources, and/or contributes to ineffective physiotherapy interventions being provided. Further primary and secondary research is warranted in order to identify statistically significant and clinically important evidence which will inform physiotherapists, consumers and service providers regarding effective interventions to improve GMSs in clients with an intellectual disability. A greater body of quality research evidence navigating effective models of physiotherapy interventions for habilitating and also rehabilitating GMSs in people with an intellectual disability would better support clinical practice, and physiotherapists' ability to choose effective interventions for optimising their clients' physical function. As well, it would ameliorate the risk of this relatively vulnerable group of consumers receiving lower quality care, or using scarce resources on minimally effective interventions.

4.6.1 Reporting of participants' characteristics

Generally, in this review the participants' physical impairments were more thoroughly documented than their intellectual impairments. For example, muscle tone was commonly described; including hypotonia (in Down syndrome and Prader-Willi syndrome), and dystonia or hypertonia (in cerebral palsy). This skew in the focus of reporting towards detail of physical issues was possibly due to the emphasis being on assessing physical change resulting from an intervention. It could also be that it is assumed that the authors expect the reader to be broadly aware of how intellectual disability impacts on clinical work and therefore do not explicitly explain this in their article. However, due to cognitive attention and perceptual-motor function being additional factors in the uptake of new motor skills for people with an intellectual disability,⁽²⁵⁶⁻²⁵⁸⁾ future research for this population should describe the cognitive abilities and impairments of participants, as well as how these factors were considered in the study design.

For the purposes of better evaluating and improving the effectiveness of physiotherapy interventions it is imperative that participants' intellectual disabilities are clearly described in studies. This will enable

improved ability to assess the potential impact that participants' intellectual disabilities have upon the choices for interventions. Reporting of participants' intellectual disabilities in future studies should include details of how the intellectual disability and its severity was diagnosed, details of any concomitant psychiatric disease, and how these conditions may impact upon motivation and learning. In a meta-analysis of prevalence of intellectual disability world-wide, all of the included primary studies reported prevalence based upon the diagnosis of intellectual disability being made by a 'mental health expert' or a paediatrician, and adopted the definition proposed by the WHO ICD Working Group on the Classification of Intellectual Disabilities of intellectual developmental disability.⁽⁷⁹⁾ More explicit reporting in primary research of the participants' intellectual disabilities could enhance the identification and development of effective study designs which meet participants' cognitive and psychological needs, and also expedite the extraction of data for systematic reviews.

4.7 Clinical topic areas for future quantitative primary research studies

There is broad scope from which to choose research topics for future primary quantitative research related to the field of physiotherapy to improve GMSs in people with an intellectual disability.

4.7.1 Participants with comparable physical deficits

The papers in the current review reported findings for study populations in which the participants had comparable physical impairments resulting from having the same clinical diagnosis (these diagnoses included Down syndrome, Prader-Willi syndrome and cerebral palsy). Only one study included participants with a range of clinical diagnoses (for childhood onset motor impairments). It is recommended that future primary research could include study populations based on persons with any intellectual disability but with specific physical impairments, such as low muscle tone, or diplegic or hemiplegic hypertonia. A greater body of evidence for specific types of physical impairments would support the development of physiotherapy recommendations for specific clinical conditions.

4.7.2 Modifying physiotherapy interventions to meet participants' learning needs

It is of note that none of the included studies identified in this review tested a mainstream intervention against the same intervention with modifications incorporating specific communication or teaching approaches. Future research assessing the potential impact of specifically designed teaching models within mainstream physiotherapy approaches could be of benefit in refining recommendations for clinical practice.

4.7.3 Consideration of intervention approaches used in related clinical fields

In the current review, the lack of specific attention given to the participants' learning needs in the studies was surprising. This may reflect a paucity of discipline specific evidence for this clinical need. However, teaching and learning frameworks appropriate for use by physiotherapists for clients with an intellectual disability could be sourced from other clinical fields of research, such as special education or psychology, and adapted to fit within physiotherapy research studies. Examples of relevant teaching and learning approaches and clinical reasoning paradigms could include consideration of psychological motivation and physiological neural plasticity to optimise rehabilitation, as has been reported for clients with an acquired brain injury.⁽²⁵⁹⁾ There are parallels between this model of rehabilitation for brain injury and physiotherapy for people with an intellectual disability: both areas must consider motivation and the need for central integration of improved or new GMSs. These considerations should be contextualised to the degree of functional impairment of the central nervous system and the impact this has upon the client's learning.^{(260,}
²⁶¹⁾ Similarly, effective intervention approaches identified for clients with cerebral palsy, for which there is a wide body of physiotherapy research evidence, can be considered when working with clients with an intellectual disability. Both of these conditions are types of developmental disabilities, and additionally cerebral palsy can result in varying degrees of intellectual impairment for the individual. Accordingly, these ideas could be incorporated in future primary research investigating effective physiotherapy interventions for people with an intellectual disability.

4.7.4 Investigation of other GMSs

All of the GMS outcomes reported in the included studies for the current review assessed weight-bearing skills: standing balance, gait, and stair climbing. This focus on weight-bearing skills reflects the clinical relevance for improvement of these skills for individuals: greater freedom with everyday mobility is experienced, and the requirements for carer support are reduced.⁽²¹²⁾ Additionally, improved balance can decrease the risk and frequency of falls and of injury from falls.

Other more complex GMSs such as ball skills or climbing were not investigated, nor were more basic skills such as independent sitting or head control. This is despite the fact that many individuals with an intellectual disability can struggle with any of these skill domains,⁽²⁶²⁻²⁶⁴⁾ whilst also having potential to improve within these skill domains.⁽²⁶⁵⁾ The lack of research for habilitating the more severely disabled person may reflect a lack of resources for habilitative inputs and a tendency for therapy inputs to use supportive seating equipment and other adaptive interventions to manage rather than overcome more extreme physical impairments. More research regarding benefits in seeking to improve GMSs, in particular weight-bearing skills, in these individuals is warranted. Given the significant improvements noted from studies included in the current review for participants with complex multiple disabilities following BWS gait training,^(165, 212) it is worthwhile to consider individuals with a greater degree of functional limitation in future research studies.

4.7.5 Reporting of results: improving clinical relevance

Future primary research studies should, where appropriate, consider reporting not only the exact statistical significance of the study results, but also the comparison of the results to normative data. This would optimise the ability for readers to appraise the clinical importance of the study's findings. Normative results such as population means, MID and MDC thresholds are clearly clinically relevant, whereas percentage improvement and statistically significant results may or may not be clinically important depending on the amount of change recorded. In future research, it may be useful to compare the

baseline results of participants with an intellectual disability to a comparator group of healthy, non-disabled individuals; this was conducted in two of the included studies in this review.^(187, 211) Such comparisons would further illustrate the severity of motor deficits present in the study population. This approach could be particularly useful when MID scores or normative data are not available for the given outcome assessments being used. Standardising participant results, for example for walking speed (standardised to leg length⁽²¹²⁾) or for balance (standardised to height⁽²¹¹⁾) would be also be useful for determining the overall effectiveness of an intervention whilst controlling for known confounding factors.

4.7.6 Use of outcome assessments validated for persons with intellectual disability

Ideally, the use of relevant outcome assessment tools which have been validated for the distinct clinical profile of the client group or population should be utilised in physiotherapy clinical practice. The GMS data extracted from the included studies in this review were measured using validated outcome measurement tools relevant for use with the study's population (table 5). Some of these tools were specific to the study participant's diagnoses, for example Down Syndrome or cerebral palsy. For the tools validated for use with individuals with cerebral palsy - a developmental disability condition in which intellectual disability is not always present⁽⁸⁴⁾ - there was no delineation for consideration of how an individual's level of cognition or the presence of any intellectual disability in the individual may have impacted upon the individual's performance. It is understandable that disease specific assessment tools would be chosen for a given population (such as for individuals with cerebral palsy); but for study cohorts comprised entirely of participants with an intellectual disability arising from any cause, it could be relevant to use additional outcome measures which are sensitive to the cognitive needs of this cohort. A range of motor assessment tools have been developed specifically for use with clients with an intellectual disability (section 1.13.1). Interestingly, minimal learning effect has been shown for outcome assessments for physical fitness in older adults with an intellectual disability despite the sample population having various levels of severity of intellectual disability.⁽¹³⁹⁾ This study also showed good test-retest reliability for the assessments, indicating

that despite variability in the cognitive presentations of the participants, repeated assessment processes are reliable over time. For future research assessing interventions for improving GMSs in this population, it is recommended that an explanation regarding the choice of outcome measures chosen should be provided when measures specifically developed for assessing people with an intellectual disability are not utilised. In future research, the use of assessment tools which have been specifically tested as reliable to use with individuals with an intellectual disability⁽²⁶⁶⁾ will strengthen the evidence base for this field.

4.7.7 Assessment approaches

In the retrieved studies in this review, some GMS outcomes were measured with complex and expensive specialised equipment; for example balance measured on computer integrated force platforms,⁽²¹¹⁾ digital video mixer to assess gait;⁽²¹²⁾ whereas other tests utilised simpler approaches such as standardised clinical tests including the GMFM.^(169, 212) Expensive equipment is not always available for clinicians to use, and simpler outcome assessments instead have to be used. Such assessment tools have been previously researched for this population (section 1.13.1). It would be beneficial if future studies of effectiveness utilised simpler outcome assessments in order to facilitate developing a body of evidence that can be more easily extrapolated to clinical practice.

In the current review, the included studies did not report results for any assessment conducted beyond the completion of the intervention programme. Future quantitative research may focus on measuring longer-term outcomes of the intervention beyond the completion of the programme, including the effect of the outcomes achieved on related domains such as levels of physical activity and participation in family life.⁽²⁶⁷⁾

4.7.8 All study participants to receive an intervention

Controlled trials often carry the potential ethical issue of denying a group of participants (the control group) the opportunity for intervention. This is particularly problematic when there is an obvious need for

it, or when it is the participant's usual therapy. However, due to there being no physiotherapy interventions with reliably proven efficacy for improving GMSs in participants with intellectual disability, head-to-head analyses are not appropriate to perform.

In the absence of any intervention with reliably proven efficacy (or even proven lack of harm), the results from any study comparing two interventions, although they may be statistically significant, should be treated with caution when being extrapolated to clinical practice. For one RCT⁽¹⁶⁵⁾ included in this review in which two interventions were compared (PBWSTT and conventional gait training), there was no comment by the authors that the interventions had comparable proven efficacy or that it was a head-to-head analysis. Accordingly, only data for the primary intervention (PBWSTT) was extracted. Interestingly, this approach has been previously reported in an RCT investigating the same two interventions, for a population of youth with cerebral palsy recruited from a special school for youth with severe cognitive and physical disabilities. However, in this study there also was no discussion regarding whether the two interventions had comparable efficacy or whether the study was/not a head-to-head analysis,⁽²⁶⁸⁾ (in this study, the participants' level of cognitive function was not described, and therefore it could not be included in the current review).

An alternative approach to designing a study which meets the ethical requirement of ensuring that all participants receive a therapeutic input would be to offer control group participants the opportunity to receive the same therapy as the intervention group following the completion of the study (and after ensuring the intervention was safe and somewhat beneficial for the intervention group). An example of this study design has been reported by Fowler et al.⁽²⁶⁹⁾ This study incorporated a modified-bicycle training programme for youth with cerebral palsy (only 13% of participants had an intellectual disability) for the primary intervention; the same intervention was then offered to the control group following the study's completion.

4.7.9 Incorporate the ICF structure

None of the studies included in the current review referred to the WHO ICF. Utilisation of the ICF⁽¹¹⁾ in future primary research would provide a common structural paradigm for developing study designs and for formulating recommendations of how best to extrapolate study results into clinical practice (section 1.9; figure 1). Previous studies reporting on interventions for individuals with cerebral palsy and other conditions have incorporated the ICF into the study design.^(115, 270)

4.7.10 Considerations for service delivery and evidence-based practice

The dearth of research investigating physiotherapy interventions for people with an intellectual disability is of considerable concern. It is possible that this lack may indicate the presence of barriers within health care settings for providing and researching physiotherapy interventions for people with an intellectual disability. A better understanding of these possible barriers and ways to optimise evidence-based practice in this clinical field is needed.⁽¹⁴⁾

4.8 Future qualitative research

Future qualitative research elucidating perspectives on improving the accessibility and appropriateness of physiotherapy for people with an intellectual disability could help to inform clinical practice, resulting in improved learning responses in clients.

4.8.1 Workplace factors affecting Best Practice

Future qualitative research investigating factors affecting the delivery of therapy services by health care providers could include ethnographic assessment of contextual workplace factors which can either support or inhibit uptake of evidence-based approaches to best practice.⁽²⁷¹⁾ A previous mixed methods study that assessed the feasibility, acceptability and contextual factors related to the implementation of a community centre-based exercise programme for older adults with an intellectual disability found good acceptance and practical feasibility.⁽²⁷²⁾ Where such research evidence is available, organisational clinical

governance systems can be embedded into practice to improve quality of health care delivery for adults with an intellectual disability.⁽²⁷³⁾

4.9 Future systematic reviews

4.9.1 Quantitative reviews to manage broad outcomes and sparse data

The dearth of studies on physiotherapy found by this systematic review suggests that in order to provide clients, carers and practitioners with the information that they need to make informed decisions regarding care, it will be necessary to go beyond just one area of clinical practice and investigate all interventions for improving GMSs in this population. A first step towards this would be to conduct a scoping review of this topic that included a wide selection of health and education professionals and a broader array of additional outcomes such as muscle strength and endurance. The results from such a review could be used to inform the conduct of more focused systematic reviews that have been designed to provide clear answers to more complex clinical questions.⁽²⁷⁴⁾ It has been previously recommended that systematic reviews that investigate complex questions and interventions should define specific domains in their research question.⁽²⁷⁵⁾ These domains should address the 'what', 'how', 'when' and 'where' of the clinical question. For the clinical area of intellectual disability, with there being varied issues present relating to equity, self-efficacy, and lower health status, such detail in the review questions is clearly warranted.

4.9.2 Search strategies

Given the limited number of papers in this field, it would be worth expanding the search strategy for a future systematic review to allow for a wider sphere of literature to be considered. For example, using a lower inclusion threshold of 40% for participant characteristics may be more appropriate, similar to the threshold levels (30% and 45%) utilised in a previous review of interventions for children with physical disabilities.⁽¹⁷⁵⁾ As well, inclusion criteria could also allow for inclusion of studies conducted by researchers from related healthcare professions, for example occupational therapy and physical education. Of note,

these two professions were represented in the list of excluded studies (appendix III). Gathering data from different healthcare disciplines may enable identification of effective approaches to be used in physiotherapy interventions to support clients' learning needs (for example, communication, repetition of interventions, simplicity of cues, and careful grading of progression of tasks). Applying such approaches to physiotherapy practice could influence the prescription of exercise programmes and how teaching is conducted with the learning of new GMS skills.

4.9.3 Qualitative systematic reviews

Disability is an interaction between an individual's intrinsic physical impairments and their medical diagnoses, their ability to cope with and overcome these issues, and the environment (for example, how inclusive versus poorly accessible it is).⁽²⁷⁶⁾ Due to considerations of learning and communication difficulties, limited finances, and poorer health status, questions relating to tolerance, and acceptance, feasibility and accessibility are important to the clinical population of people with an intellectual disability. Future research to address these issues could be best structured using a qualitative review approach.⁽¹⁸⁹⁾

4.10 Considerations for physiotherapy clinical practice

As results were limited and highly heterogeneous no strong clinical recommendations could be developed from the findings of this systematic review. However, a number of factors of high clinical interest were noted, and warrant consideration by physiotherapists.

4.10.1 Clinical assessment

Despite some of the selected studies utilising expensive and complex assessment equipment, a number of simpler assessment measures were also successfully used. As well, there are a range of accessible clinical tests which have been validated for assessing GMSs in people with an intellectual disability^(73, 142) (section 1.13.1). The use of validated outcome assessments for people with an intellectual disability is achievable.

4.10.2 Considerations for meeting learning needs of clients

For the study interventions, the level of participation required varied, and reflected the intellectual abilities of the participants. Interventions involving a greater degree of active participation by the participants were chosen for participants with mild levels of intellectual disability.^(187, 209-211) In contrast, for participants with severe intellectual disability, more assisted interventions were implemented, such as PBWS gait training therapies.^(165, 212) Physiotherapists may consider a similar approach of modifying the level of participation required during an intervention.

A number of informal approaches for improving engagement in the interventions were demonstrated in the studies. These approaches included the use of a daily journal,^(187, 211) conducting group sessions in local community settings such as gyms,⁽²¹⁰⁾ providing check/support phone calls,⁽²⁰⁹⁾ enabling participant-involvement in choice of interventions,⁽²⁰⁹⁾ and prescribing longer term home exercise programmes.^(187, 211) A recent mixed methods study found that longer term interventions have been shown to be well tolerated and feasible for people with an intellectual disability within community centre settings.⁽²⁶⁶⁾ Physiotherapists could consider similar approaches when working with clients with intellectual disabilities.

4.10.3 Task specific practice

The results from this review suggest that task specific training is clinically important (tables 4 and 6) and should be considered by physiotherapists. Task-specific modes of training were used in two of the studies with statistically significant improvements: adapted Judo training (a whole body activity training a broad array of GMSs, resulting in improved GMFM-88 scores);⁽¹⁶⁹⁾ and BWS gait training (which enables high repetitions of stepping practice for gait, with improvements in gait cadence and non-dimensionalized velocity⁽²¹²⁾). However consideration should be given to carefully matching clients to the type of intervention. For example, Judo is a more rigorous activity which is suited to only some clients, and most physiotherapists will not be skilled in this area. Alternative simpler and gentler therapies may need to be chosen to achieve the similar goal of training a broad array of GMSs; for example referring to gymnastics

classes or dancing⁽²⁷⁷⁾ could be advantageous for improving GMSs in some individuals. The use of BWS training devices were reported in two studies. BWS gait education provides a high level of task-specific training for improvements in gait. It offers the opportunity to walk at a faster cadence, and to practice a greater number of stepping actions, which increases the likelihood of central nervous system learning and adaptation.⁽²¹²⁾ BWS modalities offer safe manual handling (section 4.10.4.2) and can be practiced at lower frequencies than conventional overground gait practice.⁽¹⁶⁵⁾ Both BWS overground gait training⁽²¹²⁾ and PBWSTT⁽¹⁶⁵⁾ have been shown to be effective at a low frequency of twice weekly.⁽²¹²⁾ These positive factors support consideration of BWS modalities in physiotherapy practice, however the cost of such equipment can be prohibitive for some clinical settings.

4.10.4 Safety considerations in choice of clinical approaches

Even though statistical analyses of safety and number needed to harm were not undertaken in the included studies, the overall safety of study interventions was encouraging. This supports the consideration by physiotherapists of similar interventions for improving GMSs in people with intellectual disabilities.

4.10.4.1 *Approaches for improving safety*

The safety of clients with an intellectual disability can be optimised through a range of approaches. These include the simplification of any instructions and the overall style of the intervention, the avoidance of potentially painful or exhausting interventions particularly for participants with limited communication,⁽¹³⁸⁾ involvement of a carer or trainer to support and supervise ongoing practice of exercises,^(209, 210) and the use of assistive aides in therapy^(147, 165, 212) or modified training equipment.⁽²⁷⁸⁾ In this review, all of these approaches were reflected. Additional approaches which were implemented in the studies to ensure participants' safety and comfort included the reporting of base-line assessments and interventions only being progressed when a participant had reached a pre-defined threshold in their ability; and most studies reporting any adverse events, which, when reported, were nil or negligible (table 4). There was also

evidence in the studies of screening for known clinical safety issues, specifically prior screening of potential study participants with Down syndrome for atlanto-axial instability (section 1.14.2). If this condition is poorly managed it can lead to spinal cord impingement, particularly with an increase in level of physical activity.⁽¹⁶⁸⁾ Physiotherapists should assess for and implement appropriate safety approaches for clients with an intellectual disability.

4.10.4.2 *Manual handling*

Another safety aspect reported in the studies was with regard to manual handling⁽²¹²⁾ through the use of assistive BWS equipment. This equipment may not be an easily available resource, however, due to financial cost and bulky physical dimensions. Physiotherapists' work includes assessment of the safe manual handling needs of clients and staff, and accordingly should seek to optimise both the comfort and safety of clients during any interventions. Increased comfort can also enhance the performance of adaptive and motor skills.⁽²⁷⁹⁾ As such, where available, the use of assistive BWS equipment should be considered for use for clients with greater manual handling support needs, such as for clients with multiple and profound disabilities for whom achieving safe manual handling and gait practice can be challenging.⁽¹⁶⁵⁾

When access to expensive supportive aides is impossible, the practice of weight-bearing skills may still be achieved through the use of basic gait aides^(165, 268) and, where applicable, orthotic devices.⁽¹⁴⁷⁾ The planning of such interventions may also incorporate the help of additional staff, such as physiotherapy assistants where needed to ensure safety.⁽¹⁶⁵⁾ Safe manual handling and comfort for the client and therapists is essential.

4.11 **Conclusion**

People with an intellectual disability suffer poorer health outcomes and lower levels of independence than the general population, particularly when a concurrent physical disability related to GMS deficits is

present. International guidelines support endeavors to assist in overcoming discrepancies in health needs for people with any disability.^(106, 110, 112) As such, the current lack of research investigating physiotherapy interventions for improving GMSs in people with an intellectual disability is concerning. Based on the results of this review there is considerable indication for further primary and secondary research in this and related fields.

There is limited research evidence supporting physiotherapy for improving GMSs in people with an intellectual disability. This is surprising given the known health disparities for people with a disability, and the key role that physiotherapy plays in addressing GMS deficits. However, specific ethical and study design challenges exist when planning primary research intervention studies for participants with an intellectual disability. It can be difficult to conduct RCTs,^(187, 257) and convenience sampling can be difficult to avoid. Additionally, the inherent difficulties in recruiting large numbers of participants contribute to reduced statistical power. These factors may in part explain study design issues observed in this review of small sample sizes, convenience sampling, with only two studies being RCTs.

In the current review, a range of physiotherapy interventions to improve weight-bearing types of GMSs were identified. Significant findings were found in three of the included studies (for cadence, non-dimensionalized gait velocity and GMFM-88 scores). The studies reported excellent retention of participants and demonstrated a range of approaches for optimising clinical safety and engagement. Finding interventions that are not only safe and effective but also well tolerated by individuals with an intellectual disability is important, as concentration, motivation, and learning ability all impact on tolerance and engagement, and are known to often be impaired in this population.^(256, 280) Currently the available evidence supports the use of task-specific training, as this approach was utilized in two of the studies that showed statistically significant improvements (adapted Judo training for improved GMFM-88 scores,⁽¹⁶⁹⁾ and BWS gait training for improved cadence and non-dimensionalized gait velocity.⁽²¹²⁾)

The high level of heterogeneity of the studies in the current review demonstrated that there is a limited body of evidence in this field. This is of concern both clinically and from a resource perspective in regard to newer funding models for disability services which aim to improve the sector's efficiencies and expertise,⁽¹⁰⁷⁾ whilst enabling consumers and/or their carers to determine the amount and type of care and therapy services utilised.⁽¹⁰⁸⁾ A comparative lack of research evidence for the use of physiotherapy interventions to improve GMSs in people with an intellectual disability undermines the confidence with which physiotherapists and consumers may plan their therapy inputs. Further research is required to support physiotherapists' choice of interventions, and improve the impact of therapy on clients' GMSs. This research should focus on validating the initial findings of effectiveness identified in the current review, and also on the generalizability of physiotherapy interventions for improving GMSs in people with an intellectual disability. Due to the lack of direct primary research found in this review for the field of physiotherapy, a future systematic review could examine effective approaches for people with an intellectual disability across different health care disciplines.

This review successfully identified seven quantitative research papers which reflect the best available evidence for physiotherapy interventions to improve GMSs in people with an intellectual disability aged six years or older. Strong clinical recommendations cannot be made based upon the results of this review, however, by detailing the best available evidence for clinical practice, this systematic review should help to enable both physiotherapists and consumers to make informed choices for therapy interventions.

5 Appendices

5.1 Appendix I: MASTARI Appraisal instruments

From the Joanna Briggs Institute Meta-Analysis of Statistics Assessment and Review Instrument (JBI-MAStARI).⁽²⁰⁷⁾

JBI Critical Appraisal Checklist for Randomised Control / Pseudo-randomised Trial

Reviewer Date

Author Year Record Number

| | Yes | No | Unclear | Not Applicable |
|---|--------------------------|--------------------------|--------------------------|--------------------------|
| 1. Was the assignment to treatment groups truly random? | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> |
| 2. Were participants blinded to treatment allocation? | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> |
| 3. Was allocation to treatment groups concealed from the allocator? | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> |
| 4. Were the outcomes of people who withdrew described and included in the analysis? | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> |
| 5. Were those assessing outcomes blind to the treatment allocation? | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> |
| 6. Were the control and treatment groups comparable at entry? | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> |
| 7. Were groups treated identically other than for the named interventions | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> |
| 8. Were outcomes measured in the same way for all groups? | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> |
| 9. Were outcomes measured in a reliable way? | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> |
| 10. Was appropriate statistical analysis used? | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> |
| | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> |

Overall appraisal: Include Exclude Seek further info.

Comments (Including reason for exclusion)

JBI Critical Appraisal Checklist for Descriptive / Case Series

Reviewer Date

Author Year Record Number

| | Yes | No | Unclear | Not Applicable |
|---|--------------------------|--------------------------|--------------------------|--------------------------|
| 1. Was study based on a random or pseudo-random sample? | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> |
| 2. Were the criteria for inclusion in the sample clearly defined? | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> |
| 3. Were confounding factors identified and strategies to deal with them stated? | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> |
| 4. Were outcomes assessed using objective criteria? | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> |
| 5. If comparisons are being made, was there sufficient descriptions of the groups? | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> |
| 6. Was follow up carried out over a sufficient time period? | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> |
| 7. Were the outcomes of people who withdrew described and included in the analysis? | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> |
| 8. Were outcomes measured in a reliable way? | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> |
| 9. Was appropriate statistical analysis used? | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> | <input type="checkbox"/> |

Overall appraisal: Include Exclude Seek further info

Comments (Including reason for exclusion)

5.2 Appendix II: Data extraction instruments

From the Joanna Briggs Institute Meta-Analysis of Statistics Assessment and Review Instrument (JBI-MASARI).⁽²⁰⁷⁾

JBI Data Extraction Form for Experimental / Observational Studies

Reviewer Date

Author Year

Journal Record Number

Study Method

RCT Quasi-RCT Longitudinal

Retrospective Observational Other

Participants

Setting

Population

Sample size

Group A _____ Group B _____

Interventions

Intervention A

Intervention B

Authors Conclusions:

Reviewers Conclusions:

Study results**Dichotomous data**

| Outcome | Intervention () number / total number | Intervention () number / total number |
|----------------|---|---|
| | | |
| | | |
| | | |
| | | |

Continuous data

| Outcome | Intervention () number / total number | Intervention () number / total number |
|----------------|---|---|
| | | |
| | | |
| | | |
| | | |

5.3 Appendix III: Studies excluded after full-text review, with reasons

5.3.1 Summary of reasons for exclusion (number of studies)

Intellectual disability: No reporting of intellectual disability (9); <50% participants had an intellectual disability (8).

Physiotherapy: Physiotherapist input for some but not all participants (1); physical education study (5); occupational therapy study (2); other clinician led study (2); participant receiving usual therapy care whilst also participating in study intervention (1); unclear if physical therapist involved in study (1).

GMSs: study did not assess a GMS (3).

Age: <50% participants aged under 6yo (3).

5.3.2 List of excluded studies (citations) with reasons

1. Borggraefe I, Meyer-Heim A, Kumar A, Schaefer JS, Berweck S, Heinen F. Improved gait parameters after robotic-assisted locomotor treadmill therapy in a 6-year-old child with cerebral palsy. *Mov Disord*. [Internet]. 2008;23(2):280-3. DOI: 10.1002/mds.21802.

Reason: No reporting of intellectual disability in participants.

2. Brehm MA, Harlaar J, Schwartz M. Effect of ankle-foot orthoses on walking efficiency and gait in children with cerebral palsy. *J Rehabil Med*. 2008 Jul;40(7):529-34. DOI: 10.2340/16501977-0209.

Reason: Orthoses (intervention) done by prosthetist-orthotists.

3. Capio CM, Poolton JM, Sit CH, Eguia KF, Masters RS. Reduction of errors during practice facilitates fundamental movement skill learning in children with intellectual disabilities. *J Intellect Disabil Res*. 2013 Apr;57(4):295-305. DOI: 10.1111/j.1365-2788.2012.01535.x.

Reason: Unclear whether a physical therapist was involved in the study; unable to clarify this via author (contact email no longer active).

4. Cernak K, Stevens V, Price R, Shumway-Cook A. Locomotor training using body-weight support on a treadmill in conjunction with ongoing physical therapy in a child with severe cerebellar ataxia. *Phys Ther.* 2008 Jan;88(1):88-97. DOI: 10.2522/ptj.20070134.

Reason: Participant receiving ongoing home-based multi-disciplinary team care which included physical therapy, whilst also having the intervention training.

5. Chiviacowsky S, Wulf G, Ávila LTG. An external focus of attention enhances motor learning in children with intellectual disabilities. *J Intellect Disabil Res.* 2013 Jul;57(7):627-34. DOI: 10.1111/j.1365-2788.2012.01569.x.

Reason: A physical education study.

6. Damiano DL, Arnold AS, Steele KM, Delp SL. Can strength training predictably improve gait kinematics? A pilot study on the effects of hip and knee extensor strengthening on lower-extremity alignment in cerebral palsy. *Phys Ther.* 2010 Feb;90(2):269-79. DOI: 10.2522/ptj.20090062.

Reason: No reporting of intellectual disability in participants.

7. Deutsch JE, Borbely M, Filler J, Huhn K, Guarrera-Bowlby P. Use of a low-cost, commercially available gaming console (Wii) for rehabilitation of an adolescent with cerebral palsy. *Phys Ther.* 2008 Oct;88(10):1196-207. DOI: 10.2522/ptj.20080062.

Reason: Participants did not have an intellectual disability (IQ 79).

8. Eek MN, Tranberg R, Zugner R, Alkema K, Beckung E. Muscle strength training to improve gait function in children with cerebral palsy. *Dev Med Child Neurol.* 2008 Oct;50(10):759-64. DOI: 10.1111/j.1469-8749.2008.03045.x.

Reason: No reporting of intellectual disability in participants.

9. Fowler EG, Knutson LM, DeMuth SK, Siebert KL, Simms VD, Sugi MH, et al. Pediatric Endurance [Development] and Limb Strengthening (PEDALS) for children with cerebral palsy using stationary cycling: a randomized controlled trial. *Phys Ther.* 2010 Mar;90(3):367-81. DOI: 10.2522/ptj.20080364.

Reason: Insufficient proportion of participants with ID: only 13% of participants in both the control group and the intervention group had an ID.
10. Giagazoglou P, Arabatzi F, Kellis E, Liga M, Karra C, Amiridis I. Muscle reaction function of individuals with intellectual disabilities may be improved through therapeutic use of a horse. *Res Dev Disabil.* 2013 Sep;34(9):2442-8. DOI: 10.1016/j.ridd.2013.04.015.

Reason: A physical education study.
11. Giagazoglou P, Kokaridas D, Sidiropoulou M, Patsiaouras A, Karra C, Neofotistou K. Effects of a trampoline exercise intervention on motor performance and balance ability of children with intellectual disabilities. *Res Dev Disabil.* 2013 Sep;34(9):2701-7. DOI: 10.1016/j.ridd.2013.05.034.

Reason: A physical education study.
12. Hayakawa K, Kobayashi K. Physical and motor skill training for children with intellectual disabilities. *Percept Mot Skills.* 2011 Apr;112(2):573-80. DOI: 10.2466/06.13.15.PMS.112.2.573-580.

Reason: A physical education study.
13. Jankowicz-Szymanska A, Mikolajczyk E, Wojtanowski W. The effect of physical training on static balance in young people with intellectual disability. *Res Dev Disabil.* 2012 Mar-Apr;33(2):675-81. DOI: 10.1016/j.ridd.2011.11.015.

Reason: A physical education study.
14. Johnston TE, Wainwright SF. Cycling with functional electrical stimulation in an adult with spastic diplegic cerebral palsy. *Phys Ther.* 2011 June;91(6):970-82. DOI: 10.2522/ptj.20100286.

Reason: The single participant did not have an ID.

15. Kotte EMW, de Groot JF, Winkler AMF, Huijgen BCH, Takken T. Effects of the Fitkids exercise therapy program on health-related fitness, walking capacity, and health-related quality of life. *Phys Ther*. 2014 Sept;94(9):1306-18. DOI: 10.2522/ptj.20130315.

Reason: Less than 50% of participants had ID.

16. Kurz MJ, Corr B, Stuberg W, Volkman KG, Smith N. Evaluation of lower body positive pressure supported treadmill training for children with cerebral palsy. *Pediatr Phys Ther*. 2011 Fall;23(3):232-9. DOI: 10.1097/PEP.0b013e318227b737.

Reason: No reporting of intellectual disability in participants.

17. Kurz MJ, Stuberg W, Dejong SL. Body weight supported treadmill training improves the regularity of the stepping kinematics in children with Cerebral Palsy. *Dev Neurorehabil*. 2011 April;14(2):87-93.

Reason: No reporting of intellectual disability in participants.

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Reason: A biomedical engineering study. Only the clinical goals for improved head/neck movement were set by the participants' usual physical therapist; there was no other physical therapist input into the study.

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Reason: Age of participants too young: mean age of intervention group 6.3 +/- 2.1 and control group 6.3 +/- 2.9.

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Reason: An occupational therapy study. Emailed contact author for confirmation whether there was any physiotherapist involvement in study: no reply.

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Reason: No reporting of intellectual disability in participants.

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Reason: No reporting of intellectual disability in participants.

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Reason: All participants with developmental coordination disorder and not ID.

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Reason: Author communication clarified that participants were without any significant ID.

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Reason: Did not measure GMS outcome/s; instead measured changes in gait on a treadmill in response to changes in the upward inclination of the treadmill supporting surface; no actual intervention and reassessment undertaken.

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Reason: More than 50% of participants aged under 6yo.

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Reason: No reporting of intellectual disability in participants.

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Reason: An occupational therapy study.

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Reason: Less than 50% of participants with ID: only 7/16 participants had intellectual disability (as per author communication).

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Reason: Did not measure GMS outcome/s; instead measured strength only.

31. Tamminga JS, Martin KS, Miller EW. Single-subject design study of 2 types of supramalleolar orthoses for young children with Down syndrome. *Pediatr Phys Ther*. 2012 Fall;24(3):278-84. DOI: 10.1097/PEP.0b013e31825c8257.

Reason: Age-range too young.

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Reason: Not all participants received input by a physical therapist (as per author communication).

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Reason: No reporting of intellectual disability in participants.

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Reason: Did not measure GMS outcome/s; instead measured muscle effort and recruitment used for driving wheelchairs with different types of controls.

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Reason: Did not measure GMS outcome/s; instead measured reaction time.

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