

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

Judith Alexandra Hocking

The Joanna Briggs Institute, University of Adelaide

February 2016

Table of Contents

Abstract.....	11
Declaration.....	13
Acknowledgements.....	15
List of abbreviations.....	17
1 Introduction.....	19
1.1 Overview.....	19
1.2 Gross motor skills (GMSs).....	21
1.3 Physical activity.....	21
1.4 Defining GMS deficits.....	22
1.4.1 GMS deficits and reduced Quality of Life (QOL).....	22
1.5 Physiotherapy.....	22
1.5.1 Physiotherapy clinical assessment.....	23
1.5.2 Types of physiotherapy interventions.....	23
1.5.2.1 Settings for physiotherapy interventions.....	24
1.5.2.2 Optimising engagement in physiotherapy interventions.....	24
1.5.3 Physiotherapy to improve GMS deficits.....	24
1.5.4 Task specific practice.....	24
1.6 Intellectual disability.....	25
1.6.1 Prevalence of intellectual disability: internationally; nationally.....	26
1.6.1.1 Prevalence of intellectual disability in Australian children.....	26
1.6.2 Accommodation and education support.....	27
1.6.3 Lower health status and reduced QOL.....	27
1.6.3.1 Difficulty accessing appropriate health care.....	27

1.7	International and national documentary support for improving levels of health and health care delivery for people with an intellectual disability.....	28
1.8	The World Health Organization's International Classification of Functioning, Disability and Health (WHO ICF).29	

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

paradigms.....30

1.9	Use of the ICF when researching the needs of individuals with complex disabilities	30
1.9.1	The ICF and how it relates to this review	31
1.9.1.1	Defining GMS deficits with respect to the ICF.....	31
1.9.1.2	Relating physiotherapy assessments to the ICF	31
1.9.1.3	Relating physiotherapy interventions to the ICF	32
1.10	GMS deficits in people with an intellectual disability across the lifespan	32
1.10.1	Obesity and GMS deficits	33
1.11	The impact of GMS deficits in decreasing functional independence for people with an intellectual disability	34
1.12	Physical activity in people with an intellectual disability.....	34
1.12.1	Physical activity and GMSs in people with an intellectual disability	35
1.13	Assessment of GMSs in people with an intellectual disability	35
1.13.1	GMS outcome assessments for people with an intellectual disability.....	36
1.14	Therapy interventions for improving GMSs in people with an intellectual disability.....	36
1.14.1	Teaching and learning approaches to optimise client engagement	38
1.14.2	Consideration of specific health issues	39
1.14.3	Multi-disciplinary team models of care	40
1.14.3.1	Multi-disciplinary team programmes for children with intellectual disabilities	40
1.15	Systematic reviews: overview	41
1.15.1	Secondary research incorporating a range of study designs	42
1.15.2	Evidence-based practice, Best Practice and systematic reviews.....	42
1.16	Previous systematic reviews in fields of research related to physiotherapy and intellectual disability	43
1.17	Need for this systematic review	44

	5
1.17.1	Use of overarching search strategies in this review44
1.17.1.1	Wide age spectrum45
1.17.1.2	Addressing specific learning needs46
1.17.1	Objectives of this systematic review46
2	Methods.....47
2.1	Published <i>a priori</i> systematic review protocol47
2.2	Review Question.....47
2.3	Objectives of this review47
2.3.1	Primary objective47
2.3.2	Secondary objective.....47
2.4	Inclusion criteria: PICO47
2.4.1	Population.....48
2.4.1.1	Participants with an intellectual disability48
2.4.1.2	Age range of participants49
2.4.1.3	50 percent prevalence for key characteristics of study participants49
2.4.1.4	Aetiologies for GMS deficits50
2.4.2	Interventions50
2.4.2.1	Registered physiotherapist status51
2.4.2.2	Oversight of study interventions provided by physiotherapist51
2.4.2.3	Settings for interventions52
2.4.2.4	Study design considerations to meet participants' cognitive and learning needs52
2.4.3	Comparators52
2.4.4	Outcomes52
2.4.4.1	Primary outcomes52
2.4.4.2	Secondary outcomes.....53
2.5	Types of study designs53
2.5.1	Time frame for date of publication54

2.5.1.1	Optional extended timeframe.....	54
2.5.2	Language of publication.....	54
2.6	Reporting of results and types of statistical analyses.....	54
2.6.1	Head-to-head analyses	55
2.7	Search strategy.....	55
2.7.1	Three-step search strategy	57
2.7.2	Database search strategies	57
2.7.2.1	CINAHL	58
2.7.2.2	Embase	58
2.7.2.3	ProQuest.....	58
2.7.2.4	PubMed	59
2.8	Study selection	59
2.9	Critical appraisal	59
2.9.1	Critical appraisal instruments	59
2.9.2	Thresholds for inclusion.....	60
2.9.3	Agreement between co-reviewers.....	60
2.10	Data extraction.....	60
2.10.1	Outcomes results data	60
2.11	Data synthesis	61
3	Results	62
3.1	Primary and secondary objectives of this review	62
3.2	Selection of studies	62
3.2.1	Included studies.....	62
3.2.2	Excluded studies	62
	Figure 2: Flow diagram of selection process.....	63
3.3	Results of critical appraisal.....	64

Table 1: Results of Critical Appraisal for Randomised Control Trial/Pseudo-randomised Trial Studies	65
Table 2: Results of Critical Appraisal for Descriptive Experimental Studies.....	66
3.4 The JBI Grades for Levels of Evidence.....	67
Table 3: JBI Levels of evidence for Effectiveness Reviews	68
3.5 Narrative synthesis.....	70
3.6 Study designs and characteristics	70
3.6.1 Ethics	71
3.6.2 Recruitment	71
3.6.3 Studies originating from research centres in industrialised nations	71
3.6.4 Outcome results data	72
3.6.5 Statistical analyses reported in the included studies	72
3.6.5.1 Intention-to-treat analysis.....	72
3.6.5.2 Assessing demographic data and outcome results	73
3.6.5.3 Between-group differences in experimental studies	73
3.6.5.4 Power analyses	73
3.6.6 Confounding variables.....	74
3.6.7 Safety issues addressed in the studies	74
3.7 Participant characteristics	74
3.7.1 Participants' diagnosis of intellectual disability	74
3.7.1.1 Description of the severity of intellectual disability.....	75
3.7.2 Physical impairments of study participants	76
3.7.3 Participants' levels of physical activity	76
3.7.4 Age of participants	76
3.7.5 Reporting of body weight and Body Mass Index (BMI).....	76
3.8 Interventions.....	77
3.8.1 Clinical oversight and settings for interventions	77

3.8.2	Types of interventions	77
3.8.2.1	Habilitative approaches.....	78
3.8.2.2	Task specificity	78
3.8.2.3	Progression of interventions	78
3.8.2.4	Optimising participant engagement	79
3.8.3	Attrition; adverse events	79
3.9	Results extracted	80
3.9.1	Extraction of post-intervention data from experimental studies.....	80
3.9.2	Extraction of comparator data from experimental studies	81
3.9.3	Data extracted from repeated measures studies	81
3.10	Reporting of GMS outcome assessments	83
3.10.1	Validity of GMS outcome assessment tools.....	83
3.10.2	Length of follow-up	83

Table 4: Characteristics of studies.....84

Table 5: Validated GMS outcome assessment tools used in studies.....92

3.11	GMS outcome assessment results	95
3.11.1	Gross Motor Function Measure (GMFM).....	95
3.11.2	Gait.....	96
3.11.3	Balance	96
3.11.4	Other weight-bearing skills	97
4	Discussion.....	98
4.1	Narrative review	99
4.2	Overview of the research field	99
4.2.1	Interventions to improve GMSs well tolerated	99
4.2.2	Level of engagement in physical activity (secondary outcome) not assessed	99
4.2.3	Lack of reporting of rehabilitation measures following acute medical condition	100

4.2.4	Study design considerations	100
4.2.4.1	Use of convenience sampling.....	101
4.2.4.2	Possible reasons for the use of convenience sampling	102
4.2.4.3	Small sample sizes	102
4.2.4.4	Reporting on statistical significance	103
4.2.5	International representation in included studies.....	103
4.3	Consideration of participants' learning needs reported within studies.....	104
4.3.1	Informal consideration of learning needs of participants	105

Table 6: Authors' considerations regarding impact of participants' intellectual disability on study design 107

4.4	Limitations of this review.....	109
4.4.1	Broad inclusion criteria	109
4.4.2	Low threshold for inclusion of a study following critical appraisal.....	109
4.5	Limited evidence base for supporting physiotherapy clinical interventions	110
4.6	Recommendations for future research.....	111
4.6.1	Reporting of participants' characteristics	111
4.7	Clinical topic areas for future quantitative primary research studies	112
4.7.1	Participants with comparable physical deficits.....	112
4.7.2	Modifying physiotherapy interventions to meet participants' learning needs	113
4.7.3	Consideration of intervention approaches used in related clinical fields	113
4.7.4	Investigation of other GMSs.....	114
4.7.5	Reporting of results: improving clinical relevance	114
4.7.6	Use of outcome assessments validated for persons with intellectual disability.....	115
4.7.7	Assessment approaches	116
4.7.8	All study participants to receive an intervention.....	116
4.7.9	Incorporate the ICF structure	118
4.7.10	Considerations for service delivery and evidence-based practice	118
4.8	Future qualitative research.....	118

	10
4.8.1	Workplace factors affecting Best Practice 118
4.9	Future systematic reviews 119
4.9.1	Quantitative reviews to manage broad outcomes and sparse data 119
4.9.2	Search strategies 119
4.9.3	Qualitative systematic reviews 120
4.10	Considerations for physiotherapy clinical practice 120
4.10.1	Clinical assessment 120
4.10.2	Considerations for meeting learning needs of clients 121
4.10.3	Task specific practice 121
4.10.4	Safety considerations in choice of clinical approaches 122
4.10.4.1	Approaches for improving safety 122
4.10.4.2	Manual handling 123
4.11	Conclusion 123
5	Appendices 126
5.1	Appendix I: MASTARI Appraisal instruments 126
5.2	Appendix II: Data extraction instruments 129
5.3	Appendix III: Studies excluded after full-text review, with reasons 131
5.3.1	Summary of reasons for exclusion (number of studies) 131
5.3.2	List of excluded studies (citations) with reasons 131
6	References 139

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.

I give consent to this copy of my thesis, when deposited in the University Library, being made available for loan and photocopying, subject to the provisions of the Copyright Act 1968.

I also give permission for the digital version of my thesis to be made available on the web, via the University's digital research repository, the Library Search, and also through web search engines, unless permission has been granted by the University to restrict access for a period of time.

Signed

Judith Hocking,

on this date: .../.../..... .

Acknowledgements

I would like to formally acknowledge the thorough and expert supervisory support of Dr Jared Campbell and A/Prof Julian McNeil throughout my candidature. I also extend sincere thanks to Prof Alan Pearson for his valuable counsel in the development of the *a priori* research protocol. As well, I am grateful to Peter Hallett for co-reviewing the retrieved papers, Aderbal Aguiar Jr for the provision of raw data, and to Veronica Cimolin for providing summary statistical results.

During my candidature I have received ongoing and consistent encouragement from my family, most specially from my husband, daughter and mother. Dr Rebekah Das gave moral support at very timely intervals, and also helped me to refine my ideas during the early stages of writing this thesis.

I am also grateful for the support of The Joanna Briggs Institute, where I completed the studies reported in this thesis, and in particular to A/Prof Craig Lockwood, HDR Coordinator, for overseeing my Master of Clinical Science candidature within this School.

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