

A SYSTEMATIC REVIEW AND META ANALYSIS EXAMINING THE EFFECT OF
EXERCISE ON INDIVIDUALS WITH INTELLECTUAL DISABILITIES

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4 Abstract

5 This study examined the efficacy of exercise programs for individuals with ID based on
6 experimental designs. Multiple databases were searched from inception up until March 2019.
7 Randomised control trials were eligible for inclusion if they included a population with ID, used
8 an exercise intervention and included performance, cardiovascular health and/or psychological
9 measures. All data were pooled using random effects models of standardized mean differences.
10 The review included 18 studies which represented data from 799 individuals with ID. The largest
11 effect was seen in relation to anxiety and depression symptoms (SMD = -3.07). This study
12 represented the first comprehensive analysis on this topic area and illustrated that exercise can
13 play an important therapeutic role for this population.

14 **KEYWORDS:** Intellectual disability, exercise, intervention, systematic review

15 A systematic review and meta analyses on the effect of exercise for individuals with intellectual
16 disabilities.

17 The benefits of physical activity (PA) are numerous and have been so widely accepted
18 that even the World Health Organization presents guidelines for PA. While the evidence that PA
19 is good for both mental and physical health and that it may help improve overall quality of life is
20 strong, according to the WHO, few people actually engage in sufficient PA on a regular basis
21 (World Health Organization, 2002). Policy makers work to address this issue through mass
22 media and informational campaigns encouraging individuals to incorporate more PA into their
23 lives. Too often however, programs are designed, and resources designated in a way that best
24 serves the general population. Such an approach ignores the unique needs of individuals who
25 experience impairments due to cognitive deficits, such as those with intellectual disabilities. An
26 intellectual disability (ID) is a disability which occurs before the age of 18 and results in
27 adaptive, cognitive, and social impairments (Schalock, Luckasson, & Shrogen, 2007)

28 Adults with ID already comprise a disproportionately large number of annual hospital
29 visits and are dispensed more medications than adults without ID. One in three adults with an ID
30 in Canada are deemed high-cost patients, a designation used to refer to patients who rank within
31 the top 10% of annual health care spending (Lunsky et al., 2018). Given the fact that this cohort
32 tends to experience a disproportionate number of secondary health conditions compared to
33 typically developing adults, it would be reasonable to suggest that they, in turn, have lower
34 quality of life (May & Kennedy, 2010). Physical inactivity is one modifiable factor which is
35 linked to many conditions and diseases experienced by this population including motor
36 impairments, cardiovascular disease, obesity, diabetes and types of cancer (Durstine, Gordon,
37 Wang, & Luo, 2013). However, it is not enough to increase PA when attempting to create
38 meaningful changes to specific outcomes. Instead it is important to increase exercise behaviour

39 which is meaningful and structured and therefore, intentionally targets health and fitness related
40 outcomes (Caspersen & Christenson, 1985).

41 Exercise defined as a physical activity which is planned, structured, repetitive and
42 purposeful and focuses on improve or maintenance of one or more fitness related outcome
43 (Caspersen & Christenson, 1985). Exercise is one means by which individuals, including those
44 with ID, can meet recommended guidelines for PA (Department of health and human services,
45 2018). Regular engagement in exercise can also have positive benefits for an individual's mental
46 well-being and overall quality of life which can also significantly deteriorate as this population
47 continues to age (Ravindran et al., 2016). Approximately 4 in every 10 individuals with an ID
48 are diagnosed with a secondary mental illness. The most common conditions are anxiety,
49 depression, bipolar disorder and schizophrenia. Often adults with ID are often prescribed
50 multiple medications, as opposed to more holistic alternatives such as exercise (Finlayson,
51 Turner, & Granat, 2011), which has continually shown to regulate and improve symptoms
52 associated with mental illnesses (Ravindran et al., 2016). Therefore, a compelling argument can
53 be made that increasing exercise behaviour in this cohort may help to mitigate secondary
54 conditions associated with physical and mental health.

55 The Canadian physical activity guidelines suggest a minimum of 150 minutes of PA per
56 week to be considered sufficiently active (Dairo et al., 2016). Approximately 23% of typically
57 developing Canadians are sufficiently active, comparatively research has found that as few as 2%
58 of this individuals with intellectual disabilities are sufficiently active (Chow, Choi, & Huang,
59 2018), however this percentage does vary number ranges from 2% to 9% (Dairo, Collett, Dawes,
60 & Oskrochi, 2016). (Dairo et al., 2016).

61 While barriers to PA for this population have been acknowledged by researchers, the
62 most effective means to increase PA behaviour in this population remains unclear. As Temple
63 and colleagues (2017) stated in their recent review on physical activity promotion for adults with
64 intellectual disabilities:

65 It is clear from this review that experimental research focused on increasing
66 participation in physical activity and promoting physical activity to improve
67 the health of adults with intellectual disabilities is in its infancy...Despite
68 the potential benefits of physical activity and low levels among adults with
69 intellectual disabilities, this review demonstrates that research to document
70 the process and outcomes of physical activity interventions is sadly lacking.
71 (p.451-452)

72 Although this applies to PA behaviours in general, how best to increase participation
73 in exercise is also at this point unknown.

74 A meta-analysis by Shin & Park (2012) assessed the effect of exercise
75 programs on individuals with intellectual disabilities. The authors focused on
76 outcomes related to body composition, physiological outcomes (e.g., fitness) and
77 physical performance (i.e. balance) and found an overall positive effect of exercise
78 programs in this population. Additionally, it was found that programs which were
79 shorter in duration (e.g., 10 minutes) and ran 4 times per week were more effective
80 than those that ran for longer durations, but less frequently (e.g., 3 times per week; 13).
81 One of the major limitations of this review was related to the exclusion of individuals
82 with Down syndrome, despite the fact that this cohort makes up a large percentage of
83 individuals with ID. Additionally, the authors chose to focus on highlighting physical

84 health outcomes only, despite the fact that the psychological domain which can
85 include anxiety, depression and self-efficacy, can all be positively impacted by
86 exercise. Furthermore, the studies reviewed in the previous meta-analysis included a
87 broad range of research designs. Reducing the quality of evidence to only
88 experimental or randomized control trials can be used to examine the efficacy of
89 exercise-based intervention; Cross-sectional and longitudinal designs cannot.
90 Therefore, the objective of the current study is to assess the effectiveness of exercise
91 interventions based on experimental designs on individuals with intellectual
92 disabilities. The specific research questions are:

- 93 1. Do exercise interventions lead to positive physical and/or psychological outcomes in
94 individuals with intellectual disabilities when compared to a control?
- 95 2. What is the magnitude (i.e., effect size) of these changes?
- 96 3. Which of these outcomes, mental or physical, are most effected by exercise?

97 **Methods**

98 **Search strategy and selection criteria**

99 In the Winter of 2019, a literature search of all relevant databases was conducted. Date
100 limits were applied up to March 2019. The following databases were searched: Medical
101 Literature Analysis and Retrieval System Online (MEDLINE), Psych Info, and SportDiscus.
102 Search terms included physical activity and/or exercise (title and abstract), intellectual
103 disabilities (title and abstract), adult (title and abstract), children (title and abstract). No specific
104 publication format restrictions were set. Only studies written in English were included in the
105 literature search.

106 In order to ensure the data in this analysis is of the highest methodological rigor and
107 contains studies with the highest quality evidence, only studies which randomly assigned
108 individuals or clusters to an experimental or control group were used.

109 Given the limited research on this topic, this analysis included studies on individuals
110 from all age groups (both children and adults). In order to be inclusive of the kinds of programs
111 created for individuals with ID, studies which focused on any intellectual disability population
112 were included. The intervention must have been one that was specific to exercise. Any and all
113 programs were included regardless of setting. Again, due to the relatively small body of studies
114 in this area, any and all exercise modalities were included. However, no cross-sectional studies
115 were included. Primary outcome variables included physical and psychological variables.
116 Physical outcome variables included were body composition measures (BMI, waist
117 circumference), blood pressure, oxygen consumption and aerobic capacity. Psychological
118 outcomes included anxiety, self-rated depression, and self-efficacy.

119

120 **Data collection and analysis**

121 **Selection of studies.** Two independent reviewers screened the titles and abstracts of all
122 obtained articles. Of those identified for potential inclusion, the full texts were obtained and
123 reviewed by two independent reviewers. The reviewers discussed and resolved any
124 discrepancies that were found. A total of 18 studies were included in the review (see Figure 1).

125 **Assessment of risk of bias in included studies.** In order to grade the strength of the
126 evidence, study quality was assessed using the Cochrane Risk of Bias 2.0 (RoB 2.0), specifically
127 designed for cluster randomized trials. The RoB 2.0 was used to assess bias related to threats to
128 internal validity such as flawed research design, poor study execution, and/or incomplete

129 reporting of results. Specifically, the RoB 2.0 assesses risks associated with randomization and
130 allocation sequence, blinding, incomplete or missing outcome data, and selective reporting
131 (Sterne, Egger, Moher, & Boutron, 2017). The RoB 2.0 was completed by two independent
132 reviewers. Any disagreements were discussed until a consensus was reached.

133 **Measurement of treatment effects.** To assess the effectiveness of the intervention on
134 the various outcome measures, standardized mean difference (SMD) and standard error (SE)
135 were calculated for all outcome variables. The use of SMD and SE allows for the summary
136 reporting of findings taking into account that different scales and measures were used across
137 different studies. The degree of the standardized mean difference was assessed using Cohen's
138 standardized conventions (Sterne et al., 2017) for effect size; small (0.2), medium (0.5) and large
139 (0.8). Studies typically reported pre-and post-intervention time points, however due to studies
140 not reporting variability in the change scores (i.e., standard deviation (SD) of the change score),
141 a comparison of the post intervention measurement score were used (Cohen, 1992). The post
142 intervention mean score and SD for measures of performance, body composition, cardiovascular
143 and psychological measures were all entered. A direct comparison of the exercise intervention
144 and control group was then completed.

145 **Assessment of heterogeneity.** For the purpose of this review, heterogeneity is defined as
146 follows: "Statistical heterogeneity manifests itself in the observed intervention effects being
147 more different from each other than one would expect due to random error (chance) alone."
148 (Higgins & Green, 2011). A visual and statistical examination of any study estimate
149 inconsistencies was completed by visually examining forest plots and consideration of the X^2
150 and I^2 values. The proposed thresholds from the Cochrane handbook chapter 9.5.2 (Higgins &
151 Green, 2011) were used to interpret I^2 values.

Data synthesis

Random effects models were generated for each outcome using RevMan (Version 5.3, 2014) software. Forest plots of the main analyses and tables containing the results of the sensitivity analyses were also generated.

Subgroup and sensitivity analyses. In cases where outcomes were assessed through unique measurements (e.g., musculoskeletal fitness was measured in both upper and lower body), subgroups would be used to analyze the data. This allowed for independent analyses of effect sizes for varying measures within the same outcome. Sensitivity analyses investigated the impact of varying intraclass correlation coefficient (ICC) values.

Results**Results of search**

A total of 715 records were obtained from all databases. After 129 duplicates were removed, 585 titles were screened. From this first level of screening (titles and abstracts), 489 studies were deemed irrelevant, leaving 96 full text articles to be reviewed for eligibility. Of those 96, 78 were excluded. Reason for exclusion was wrong study design (53 studies), wrong outcomes (11 studies), wrong intervention (7 studies), wrong patient population (4 studies), wrong route of administration (2 studies) and wrong setting (1 study). A total of 18 studies were included in this review (see Figure 1). The percent disagreement for the full text review was 7%, with discrepancy on seven articles. Of those seven articles, 0 were included in the final review.

Included studies

The 18 studies included 799 individuals with intellectual disabilities from studies conducted in Europe, the United States, Australia, and South Africa. Sixteen of the trials randomized individual participants into the intervention or control group, while two studies used cluster randomization (e.g., Day Activity centers). Intervention length ranged from 5 weeks to

176 52 weeks. Most interventions ran for 10-12 weeks and were typically performed 3 times per
177 week.

178 **Risk of bias in included studies**

179 Table 1 documents data regarding the risk of bias assessments for all included studies.

180 Overall, two studies were low risk of bias, nine studies were deemed to have some concerns, and
181 the remaining seven were rated as having a high risk of bias. Out of the 18 studies, 9 studies had
182 at least some risk of bias regarding deviations due to intended intervention, usually due to lack of
183 proper participant/trial personnel blinding.

184 **Results of Pooled Sample**

185 Participants in this review included individuals with mild to moderate ID (n=279),
186 individuals with Down syndrome with ID (n=130), individuals with profound ID (n=37), and
187 individuals with ID of unspecified etiology (n= 353). Of those, 349 (44%) were male; however,
188 the gender of 30 participants was unspecified (Beasley,1982).

189 **Effects of intervention**

190 The following results come from 18 studies.

191 **Performance Measures**

192 All results for performance measures are outlined in Table 2.

193 **Submaximal exercise.** In this review, submaximal exercise is defined as a type of
194 exercise that is terminated before reaching ventilatory threshold or maximum heart rate (HR). It
195 is used to estimate VO_2 max or aerobic fitness (Heyward,2009). Estimates of aerobic fitness
196 were tested using the six-minute walk test (Boer et al., 2014; Boer, & Moss, 2016; Calder et al.,
197 2011; Marks, Sisirak, & Chang, 2013), and the shuttle run test (Ozmen, Ryildirim, Yuktasir, &
198 Beets, 2007; Schijndel-Speet, Evenhuis, Wijck, & Echteld, 2014). Pooled estimates of all

199 measures of aerobic fitness from eight studies (Beasley,1982, Boer et al., 2014; Boer, & Moss,
200 2016; Calders et al., 2011; Lee, Lee, & Song, 2016; Marks, Sisirak, & Chang, 2013; Ozmen et
201 al., 2007; Schijndel-Speet et al., 2014) with a combined sample size of 333 participants showed
202 almost no increase in aerobic fitness when comparing exercise interventions to sedentary
203 controls (SMD = 0.13, 95% confidence interval (CI) range from -0.11 to 0.37). The range of
204 effects shows moderate possible harm, no effect, and small benefit. The overall $I^2 = 17\%$
205 indicated low heterogeneity.

206 **Balance.** For the purpose of this review, balance is defined as static balance or the ability
207 to maintain the body in a fixed position (Rival, Ceyte, & Olivier, 2005). Three studies (Borji, et
208 al., 2018; Lee et al., 2016; Schijndel-Speet et al., 2014) (pooled $n=162$) assessed static balance.
209 Pooled estimates show that although the effect size is large (SMD = 1.25), CI ranged from -0.39
210 to 2.90, indicating no effect of the intervention. This range indicates that results show a small
211 degree of harm, no effect, and large benefit.

212 **Functional fitness.** Functional fitness reflects one's ability to perform physical activities
213 of daily life with relative ease (Heyward, 2009). Functional fitness included five studies (Boer et
214 al., 2014; Boer, & Moss, 2016; Calders et al., 2011; Lee et al., 2016; Marks et al., 2013) and a
215 total of 325 participants, with pooled estimates indicating a minimal effect (SMD = -0.07).
216 Overall, results were imprecise: showing high benefit, no effect, and a moderate degree of harm.
217 When subgroup analysis was examined, Sit to Stand was found to favor the controls (SMD =
218 0.37), while the Get up and Go test did have a large, benefit observed in the intervention group
219 (SMD = -0.77) (the decrease in the Get up and Go scores does indicate an improvement on the
220 test, as decreased time indicates better functional fitness). The X^2 test of subgroup differences

221 was statistically significant ($p=0.02$) indicating the effects differ across between the two
222 subgroups.

223 **Musculoskeletal strength.** For this review, muscular strength was defined as any
224 activity which elicited the maximum force that a muscle or muscle group can generate at a
225 specific velocity. Studies looked at pooled estimates from all measures from six studies (Boer et
226 al., 2014; Calders et al., 2011, Giagazoglou et al., 2013; Schijndel-Speet et al., 2014; Shields et
227 al., 2013; Suomi, 1998) of 351 participants showed an effect estimate indicating strong, positive
228 increase when comparing the intervention group to the control group (SMD = 0.70, 95% CI
229 range from 0.24 to 1.16), indicating benefit. Subgroup analyses show slightly less conclusive
230 results for upper body strength (95% CI range from -0.17 to 1.26), while the results for low body
231 musculoskeletal strength indicate a strong effect (SMD = 0.86, 95% CI range from 0.30 to 1.42).
232 The overall $I^2 = 74\%$, indicating a high degree of heterogeneity.

233 **Flexibility.** Two studies (Giagazoglou et al., 2013; Marks et al., 2013) were included in
234 the pooled analysis of flexibility. Results from 152 participants were inconclusive, showing
235 possible benefit, no effect, and possible harm (SMD = -0.19, 95% CI from -1.73 to 1.34).

236 **Body composition.** Body composition included body mass index (Boer et al., 2014,
237 Calders et al., 2011; Melville et al., 2015; Ozmen et al., 2007), weight in pounds (Marks et al.,
238 2013), weight in kilograms (Schijndel-Speet et al., 2014). Pooled estimates from all measures
239 from six studies of 343 participants showed a small effect size for weight (SMD = 0.13, 95% CI
240 from -0.12 to 0.37). The 95% CI indicates the possibility of benefit, no effect and possible harm.
241 Only two studies (Melville et al., 2015; Schijndel-Speet et al., 2014) measured waist
242 circumference, and results were inconsistent: possible benefit, no effect, and possible harm (95%
243 CI from -0.37 to 0.50). All results for body composition are outlined in Table 3.

244 Cardiovascular Health

245 All results for cardiovascular health are outlined in Table 4.

246 Four studies (Boer & Moss, 2016; Boer et al., 2014; Calders et al., 2011; Rosety-
247 Rodriguez et al., 2014) examined maximal oxygen uptake ($n = 109$); the pooled estimates
248 indicated a medium effect ($SMD = 0.55$) when comparing the exercise intervention to the
249 sedentary control. The 95% CI ranged from 0.17 to 0.94.

250 Blood pressure was examined in three studies (Boer & Moss, 2016; Calders et al., 2011;
251 Schijndel-Speet et al., 2014), and the results of pooled estimates revealed a medium effect (SMD
252 $= -0.30$, 95% CI from -0.56 to -0.03). Subgroup analyses of systolic and diastolic blood pressure
253 found a medium effect of exercise on systolic blood pressure ($SMD = -0.47$, 95% CI from -0.085
254 to -0.10), while exercise had a small effect on diastolic ($SMD = -0.12$, 95% CI from -0.48 to
255 0.23 ; See Figure) with no significant differences between subgroups ($p = 0.19$).

256 While results from the pooled estimates of three studies (Boer & Moss, 2016; Boer et al.,
257 2014; Calders et al., 2011) ($n=89$) examining heart rate did show a small effect of exercise
258 benefiting the intervention group ($SMD = 0.11$) the 95% CI show indefinite results: possible
259 harm, no effect and possible benefit.

260 Psychological Outcomes

261 All results for psychological outcomes are outlined in Table 5.

262 Anxiety and depression was assessed by three studies (Carraro & Gobbi, 2012; Carraro &
263 Gobbi, 2014; Schijndel-Speet, 2014). The pooled estimates from 140 participants showed a
264 large effect of exercise on anxiety and depression related symptoms ($SMD = -3.07$). However,
265 the CI were extremely wide and provides unspecified results (95% CI from -6.81 , 0.66).

266 Self-efficacy was examined in two studies (Marks et al., 2013; Melville et al., 2015) (n =
267 152) and pooled estimates indicated a large effect of exercise when comparing the intervention to
268 the sedentary control (SMD = 0.74, 95% CI from -0.33 to 1.80).

269 **Discussion**

270 **Summary of main results and certainty of evidence**

271 Interest in exercise as an intervention for various outcome measures for individuals with
272 IDs has steadily increased, with a greater number of interventions appearing in the literature.
273 However, the inclusion of all available trials provided inconsistent results. Some evidence did
274 indicate improvements with regards to lower body muscular endurance, blood pressure, reaction
275 time, and self-efficacy. Results for other outcomes were inconsistent, even potentially harmful.
276 Furthermore, the best available evidence for other outcomes is uncertain due to the quality of
277 evidence. It is possible, however that those with ID need extra support to fully and consistently
278 maximize the benefits of exercise.

279 **Overall completeness and applicability of evidence**

280 The studies within this review include individuals with various types of intellectual
281 disabilities (Down syndrome, IDD, PDD-NOS), various exercise modalities, and a wide range of
282 age groups. Very few of these studies are adequately powered, and limited number had examined
283 multiple outcomes in the same study. The problem of low sample size resulted in confidence
284 intervals were wide and could not provide quality, determinant results. Additionally, few
285 studies reported ICC values, which also made it difficult to assess quality. It is important to
286 consider, however the difficulty associated with recruiting this population, which could largely
287 account for the small participant sample sizes. In general, studies had at least some concern or
288 high concern with regards to risk of bias. Typically, studies did not have blinded outcome

289 assessors, and as a result, the risk of bias assessment with regards to measurement outcome was
290 high.

291 Reporting errors were also a major cause of concern. One study, in particular, did not
292 include any information regarding the duration, frequency, intensity or modality of the exercise
293 intervention (Schijndel-Speet, 2014). Additionally, some studies included information for some
294 of these categories, but not all. As a result, it was not possible to conduct post hoc analyses
295 exploring the effect of intervention duration, frequency, and intensity on outcomes. Therefore,
296 the body of research must become more robust in order to identify detectable differences/effects
297 estimates resulting from various durations, frequencies, and intensities. Specifically, increases in
298 sample size and the quality surrounding the measurement of the interventions need to be
299 improved.

300 **Agreements and disagreements with other studies or reviews**

301 Overall, the results from this meta-analysis show that while definitive, and salient
302 changes were not observed in all domains/outcomes, some improvements were apparent.
303 Notably, there is at least some modest evidence that exercise may lead to positive changes in
304 musculoskeletal strength, maximal oxygen uptake, and blood pressure. Previous research
305 supports the findings from the current meta-analysis as previous work has also found that
306 exercise programs improved muscle force, VO_2 , and self-esteem in a similar population (Shin &
307 Park, 2012). Furthermore, in the general population, sustained and ongoing exercise behaviour
308 can help to improve total blood pressure score, and reduce symptoms associated with
309 hypertension (Carpio-Rivera, Moncada-Jiménez, Salazar-Rojas, & Solera-Herrera, 2016) and it
310 appears this extends to populations with intellectual disabilities as well.

311 The current review was the first to examine the impact that exercise can have on mental
312 health. The evidence showed that there were large (though, somewhat imprecise) gains in
313 mental health outcomes for adults with ID. Specifically, when investigating exercises impact on
314 anxiety and depressive symptoms. This finding not only points out the link between mental
315 health and exercise, but the complexity of this link in those with ID and the importance of
316 exercise as a means of providing holistic treatment for secondary mental health conditions.
317 However, the results from this study show strong but variable results due to few studies actually
318 investigating mental health in this population. This provides more reason for researchers and
319 practitioners to continue their consideration of exercise and mental health in this group. More
320 research needs to explore how these benefits are maintained beyond the duration of the
321 intervention.

322 While research has previously shown that exercise can indeed improve body composition
323 outcomes (Swift, Johannsen, Lavie, Earnest, & Churh, 2013), the current meta-analysis found
324 only a small effect on body composition despite the fact that each of the six studies used exercise
325 as a means to facilitate weight loss. Beyond the small effect size, CI indicated no effect and
326 possibility of harm. However, when it comes to weight loss, exercise is only one small aspect
327 related to weight loss. Individuals must also maintain a healthy diet and must reduce caloric
328 intake while increasing energy input. Furthermore, weight loss is highly unique and differs from
329 person to person (Swift et al., 2013). One factor which hinders weight loss and improvements in
330 body composition for individuals with ID is medication. A large percentage of this population is
331 prescribed antipsychotic and antidepressant medications (Doan, Lennox, Taylor-Gomez, &
332 Ware, 2013), both of which can cause weight gain (Wharton, Raiber, Serodio, Lee, &
333 Christensen, 2018). Medication-induced weight gain is significant and often difficult to reverse.

334 While none of the studies in the current review indicated whether or not participants were taking
335 medication, it is possible that this, in combination with other individual factors (i.e., nutrition),
336 resulted in vague findings.

337 Flexibility, step count, and heart rate also had unclear results, which aligns with previous
338 work (Shin & Park, 2012). It is likely this is a result of small sample sizes as each of these
339 outcomes had less than 100 participants included in the pooled estimates. However, outcomes
340 with larger sample sizes (submaximal exercise, balance, functional fitness, anxiety/depression)
341 also had imprecise results. Many of the studies included in these outcomes were 12-week
342 exercise interventions, and it is possible that this length was not enough to produce any salient
343 changes.

344 **Conclusion**

345 The results of this review, while inconsistent, do provide some evidence indicating the
346 need for more research to determine the efficacy of fitness/exercise programs for this population.
347 It is clear that within this population, exercise and exercise related behaviours can lead to some
348 positive changes in specific outcomes. Current research is largely focused on physical outcomes
349 while there is an extreme lack of quality evidence supporting exercise as an alternative therapy
350 for mental health in this population. It is important to recognize that exercise may act as a
351 primary treatment for many co morbid conditions that are prominent within this population such
352 as anxiety and depression (Lunsky et al., 2018). While this review was not able to identify best
353 exercise practices for this population, it does provide evidence that exercise (through any
354 method) may be of benefit for individuals with ID.

355 This review is largely limited by the quality of evidence, which justifies the need for
356 future studies to employ methodologically sound, adequately powered interventions.

357 Additionally, very little information could be drawn regarding optimal program frequency,
358 timing, and length (which are fundamental to any exercise program) due to lack of evidence.
359 This also extends to adherence, as no studies completed follow-ups to confirm if the changes
360 they saw extended beyond the duration of the intervention. This is an important note for future
361 researchers as evidence which supports whether or not these changes are resistant to time is
362 lacking. Furthermore, no studies measured fidelity related to the implementation of the
363 intervention and if there were deviations from the intended program this may have effected the
364 final results. Therefore, it is important that future research ensure that interventions are being
365 carried out according to the initial design.

366 Insufficient evidence does not allow us to draw conclusions regarding several outcomes
367 including functional fitness, submaximal exercise performance, and heart rate. Overall this
368 review serves a pertinent reminder that while individual studies have identified exercise as a
369 prominent way to improve many lifestyle and health factors in those with ID this data should be
370 subject to reproduction before it can be taken as fact.

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References

373

Beasley, C. R. (1982). Effects of a jogging program on cardiovascular fitness and work

374

performance of mentally retarded adults. *American Journal of Mental Deficiency*, 86(6),

375

609–613.

376

Boer, P. H., Meeus, M., Terblanche, E., Rombaut, L., De Wandele, I., Hermans, L., ... Calders,

377

P. (2014). The influence of sprint interval training on body composition, physical and

378

metabolic fitness in adolescents and young adults with intellectual disability: A randomized

379

controlled trial. *Clinical Rehabilitation*, 28(3), 221–231.

380

<https://doi.org/10.1177/0269215513498609>

381

Boer, P. H., & Moss, S. J. (2016). Effect of continuous aerobic vs. interval training on selected

382

anthropometrical, physiological and functional parameters of adults with Down syndrome.

383

Journal of Intellectual Disability Research, 60(4), 322–334.

384

<https://doi.org/10.1111/jir.12251>

385

Borji, R., Sahli, S., Baccouch, R., Laatar, R., Kachouri, H., & Rebai, H. (2018). An open-label

386

randomized control trial of hopping and jumping training versus sensorimotor rehabilitation

387

programme on postural capacities in individuals with intellectual disabilities. *Journal of*

388

Applied Research in Intellectual Disabilities, 31(2), 318–323.

389

<https://doi.org/10.1111/jar.12324>

390

Bossink, L. W. M., van der Putten, A. A. J., Waninge, A., & Vlaskamp, C. (2017). A power-

391

assisted exercise intervention in people with profound intellectual and multiple disabilities

- 392 living in a residential facility: a pilot randomised controlled trial. *Clinical Rehabilitation*,
393 31(9), 1168–1178. <https://doi.org/10.1177/0269215516687347>
- 394 Rival, C., Ceyte, H., & Olivier, I. (2005). Developmental changes of static standing balance in
395 children. *Neuroscience Letters*, 376(2), 133–136.
396 <https://doi.org/10.1016/j.neulet.2004.11.042>
- 397 Calders, P., Elmahgoub, S., de Mettelinge, T. R., Vandenbroeck, C., Dewandele, I., Rombaut, L.,
398 ... Cambier, D. (2011). Effect of combined exercise training on physical and metabolic
399 fitness in adults with intellectual disability: a controlled trial. *Clinical Rehabilitation*,
400 25(12), 1097–1108. <https://doi.org/10.1177/0269215511407221>
- 401 Carpio-Rivera, E., Moncada-Jiménez, J., Salazar-Rojas, W., & Solera-Herrera, A. (2016). Acute
402 effects of exercise on blood pressure: A meta-analytic investigation. *Arquivos Brasileiros de*
403 *Cardiologia*. <https://doi.org/10.5935/abc.20160064>
- 404 Carraro, A., & Gobbi, E. (2012). Effects of an exercise programme on anxiety in adults with
405 intellectual disabilities. *Research in Developmental Disabilities*, 33(4), 1221–1226.
406 <https://doi.org/https://doi.org/10.1016/j.ridd.2012.02.014>
- 407 Carraro, A., & Gobbi, E. (2014). Exercise Intervention to Reduce Depressive Symptoms in
408 Adults with Intellectual Disabilities. *Perceptual and Motor Skills*, 119(1), 1–5.
409 <https://doi.org/10.2466/06.15.PMS.119c17z4>
- 410 Caspersen, C. J., & Christenson, G. M. (1985). Physical Activity , Exercise , and Physical
411 Fitness : Definitions and Distinctions for Health-Related Research, (April).

- 412 Chow, B. Choi, Peggy. Huang, W. (2018). Physical Activity and Physical Fitness of Adults with
413 Intellectual Disabilities in Group Homes in. *International Journal of Environmental*
414 *Research and Public Health*, 15. <https://doi.org/10.3390/ijerph15071370>
- 415 Cohen, J. (1992). A power primer. *Psychological Bulletin*. US: American Psychological
416 Association. <https://doi.org/10.1037/0033-2909.112.1.155>
- 417 Dairo, Y. M., Collett, J., Dawes, H., & Oskrochi, G. R. (2016). Physical activity levels in adults
418 with intellectual disabilities: A systematic review. *Preventive Medicine Reports*, 4, 209–
419 219. <https://doi.org/10.1016/j.pmedr.2016.06.008>
- 420 Department of Health & Human Services. (2018). 2018 Physical Activity Guidelines Advisory
421 Committee. *Physical Activity Guidelines Advisory Committee Scientific Report*, 779.
422 <https://doi.org/10.1111/j.1753-4887.2008.00136.x>
- 423 Doan, T. N., Lennox, N. G., Taylor-Gomez, M., & Ware, R. S. (2013). Medication use among
424 Australian adults with intellectual disability in primary healthcare settings: A cross-
425 sectional study. *Journal of Intellectual & Developmental Disability*, 38(June), 177–181.
426 <https://doi.org/10.3109/13668250.2013.778968>
- 427 Durstine, J. L., Gordon, B., Wang, Z., & Luo, X. (2013). Chronic disease and the link to physical
428 activity. *Journal of Sport and Health Science*, 2(1), 3–11.
429 <https://doi.org/10.1016/j.jshs.2012.07.009>
- 430 Finlayson, J., Turner, A., & Granat, M. H. (2011). Measuring the Actual Levels and Patterns of
431 Physical Activity / Inactivity of Adults with Intellectual Disabilities, 508–517.

- 432 Giagazoglou, P., Kokaridas, D., Sidiropoulou, M., Patsiaouras, A., Karra, C., & Neofotistou, K.
433 (2013). Effects of a trampoline exercise intervention on motor performance and balance
434 ability of children with intellectual disabilities. *Research in Developmental Disabilities*,
435 34(9), 2701–2707. <https://doi.org/10.1016/j.ridd.2013.05.034>
- 436 Heyward VH. *Advanced fitness assessment and exercise prescription*. 8th ed. Champaign, IL:
437 Human Kinetics; 2009. 369 p.
- 438 Higgins, J., & Green, S. (2011). Cochrane Handbook: Cochrane Reviews of Interventions. In
439 *Cochrane Handbook for: Systematic Reviews of Interventions* (Vol. 6, pp. 3–10).
- 440 Higgins J, T. S. (2008). Chapter 10-Addressing reporting biases. In *Cochrane Handbook for*
441 *Systematic Reviews of Interventions* (p. 10.1-10.33). Retrieved from
442 http://handbook.cochrane.org/chapter_10/10_addressing_reporting_biases.htm
- 443 Lee, K., Lee, M., & Song, C. (2016). Balance training improves postural balance, gait, and
444 functional strength in adolescents with intellectual disabilities: Single-blinded, randomized
445 clinical trial. *Disability and Health Journal*, 9(3), 416–422.
446 <https://doi.org/https://doi.org/10.1016/j.dhjo.2016.01.010>
- 447 Lunskey, Y., Balogh, R., Durbin, A., Selick, A., Volpe, T., & Lin, E. (2018). The Mental Health
448 of Adults with Developmental Disabilities in Ontario : Lessons from Administrative
449 Health Data, 21(1), 51–54.

- 450 Marks, B., Sisirak, J., & Chang, Y. C. (2013). Efficacy of the HealthMatters Program Train-the-
451 Trainer Model. *Journal of Applied Research in Intellectual Disabilities*, 26(4), 319–334.
452 <https://doi.org/10.1111/jar.12045>
- 453 May, M. E., & Kennedy, C. H. (1998). Health and Problem Behavior Among People With
454 Intellectual Disabilities, 3(2), 4–12.
- 455 Melville, C. A., Mitchell, F., Stalker, K., Matthews, L., McConnachie, A., Murray, H. M., ...
456 Mutrie, N. (2015). Effectiveness of a walking programme to support adults with intellectual
457 disabilities to increase physical activity: walk well cluster-randomised controlled trial. *The*
458 *International Journal of Behavioral Nutrition and Physical Activity*, 12, 125.
459 <https://doi.org/10.1186/s12966-015-0290-5>
- 460 Ozmen, T., Ryildirim, N. U., Yuktasir, B., & Beets, M. W. (2007). Effects of school-based
461 cardiovascular-fitness training in children with mental retardation. *Pediatric Exercise*
462 *Science*, 19(2), 171–178.
- 463 Ravindran, A. V, Balneaves, L. G., Faulkner, G., Ortiz, A., Mcintosh, D., Morehouse, R. L., ...
464 Parikh, S. V. (2016). Canadian Network for Mood and Anxiety Treatments (CANMAT)
465 2016 Clinical Guidelines for the Management of Adults with Major Depressive Disorder :
466 Section 5 . Complementary and Alternative Medicine Treatments.
467 <https://doi.org/10.1177/0706743716660290>
- 468 Rosety-Rodriguez, M., Diaz, A. J., Rosety, I., Rosety, M. A., Camacho, A., Fornieles, G., ...
469 Ordonez, F. J. (2014). Exercise reduced inflammation: But for how long after training?

- 470 *Journal of Intellectual Disability Research*, 58(9), 874–879.
471 <https://doi.org/10.1111/jir.12096>
- 472 Schalock, R. L., Luckasson, R. A., & Shogren, K. A. (2007). The Renaming of Mental
473 Retardation. *Intellectual and Developmental Disabilities*, 45(2), 116–124.
474 [https://doi.org/10.1352/1934-9556\(2007\)45\[116:tromru\]2.0.co;2](https://doi.org/10.1352/1934-9556(2007)45[116:tromru]2.0.co;2)
- 475 van Schijndel-Speet, M., Evenhuis, H. M., van Wijck, R., & Echteld, M. A. (2014).
476 Implementation of a group-based physical activity programme for ageing adults with ID: a
477 process evaluation. *Journal of Evaluation in Clinical Practice*, 20(4), 401–407.
478 <https://doi.org/10.1111/jep.12145>
- 479 Shields, N., Taylor, N. F., Wee, E., Wollersheim, D., O’Shea, S. D., & Fernhall, B. (2013). A
480 community-based strength training programme increases muscle strength and physical
481 activity in young people with Down syndrome: A randomised controlled trial. *Research in*
482 *Developmental Disabilities*, 34(12), 4385–4394.
483 <https://doi.org/https://doi.org/10.1016/j.ridd.2013.09.022>
- 484 Shin, I. S., & Park, E. Y. (2012). Meta-analysis of the effect of exercise programs for individuals
485 with intellectual disabilities. *Research in Developmental Disabilities*.
486 <https://doi.org/10.1016/j.ridd.2012.05.019>
- 487 Suomi, R. (1998). Self-directed strength training: its effect on leg strength in men with mental
488 retardation. *Archives of Physical Medicine and Rehabilitation*, 79(3), 323–328.

- 489 Swift, D. L., Johannsen, N. M., Lavie, C. J., Earnest, C. P., & Church, T. S. (2014). The role of
490 exercise and physical activity in weight loss and maintenance. *Progress in Cardiovascular*
491 *Diseases*, 56(4), 441–447. <https://doi.org/10.1016/j.pcad.2013.09.012>
- 492 Temple, V. A., Frey, G. C., Stanish, H. I., Va, T., Gc, F., & Hi, S. (2017). Interventions to
493 promote physical activity for adults with intellectual disabilities, 59(4).
- 494 Temple, V. A. (2007). Barriers, enjoyment, and preference for physical activity among adults
495 with intellectual disability. *International Journal of Rehabilitation Research*, 30(4), 281–
496 287. <https://doi.org/10.1097/MRR.0b013e3282f144fb>
- 497 Wharton, S., Raiber, L., Serodio, K. J., Lee, J., & Christensen, R. A. G. (2018). Medications that
498 cause weight gain and alternatives in Canada : a narrative review, 427–438.
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500 **FOOTNOTES**

501 **Contributors:** LSJ: Study conception/design, data acquisitions, data analysis and interpretation,
502 and drafting and revision of manuscript. GB: Data acquisition, data analysis, and revision of
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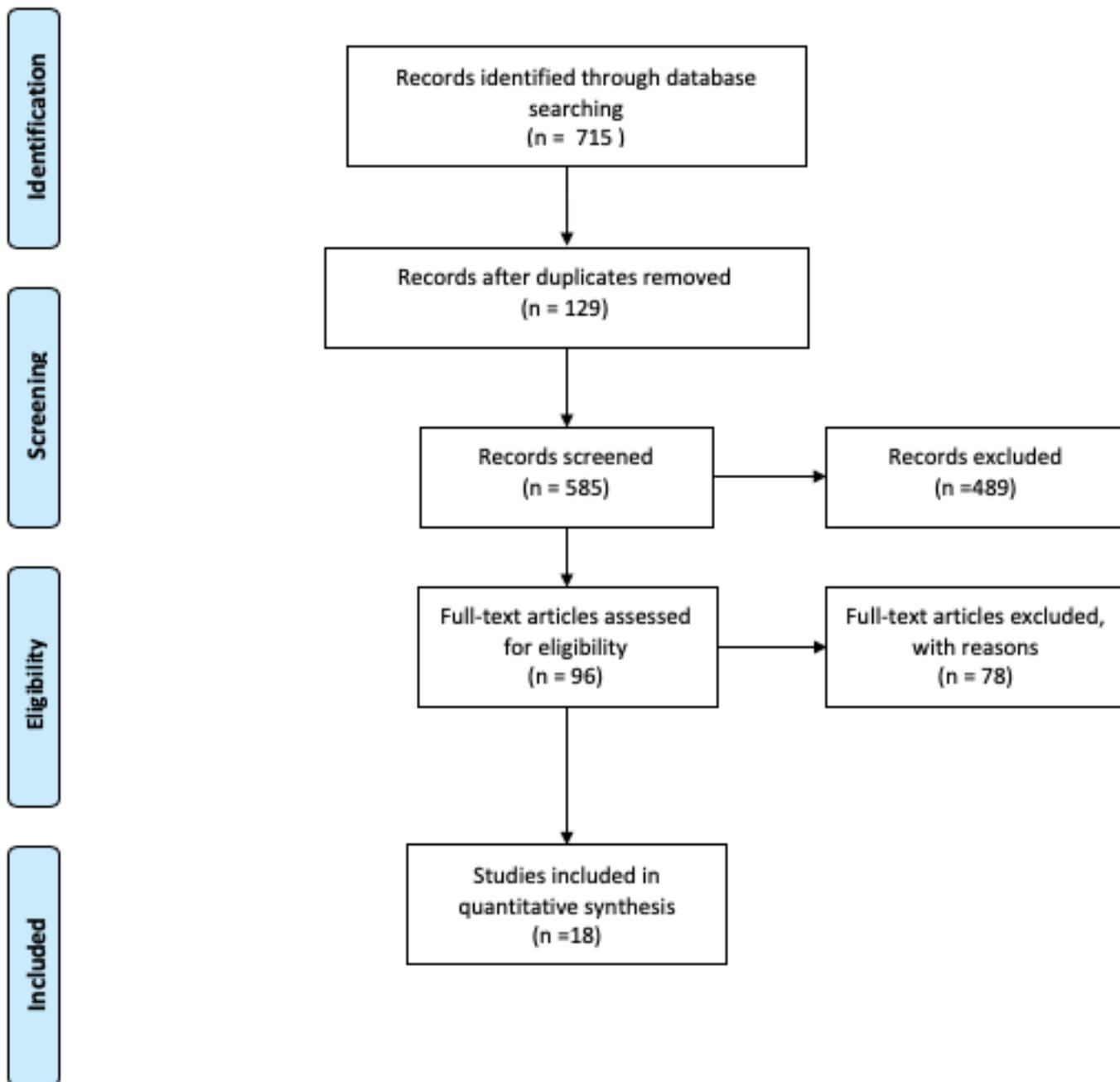


Figure 1. PRISMA flow chart.

Table 1
Risk of bias assessment for included studies

Study (Publication Date)	Randomization process bias over all judgement	Bias due to deviations from intended interventions	Bias due to missing outcome data	Bias in measurement of the outcome	Bias in selection of the reported result	Overall Judgment
Beasley (1982)	Some concerns	Some concerns	Low	Some concerns	Low	Some concerns
Boer (2014)	Low	Low	Some concerns	Low	Low	Some concerns
Boer (2016)	Low	Some concerns	Some concerns	Low	Low	Some concerns
Borji (2018)	High risk	Low	Low	Some concerns	Low	High
Bossink (2017)	Low	Low	Low	Low	High	High
Calders (2011)	High risk	High	Low	Low	Low	High
Carraro (2012)	Low	Some concerns	Low	High	High	High
Carraro (2014)	Low	Some concerns	Low	High	Low	High
Giagazoglou (2013)	Low	Low	Low	Some concerns	Low	Some concerns
Khalili (2009)	Low	Low	Low	Low	Low	Low
Lee (2016)	Low	Low	Low	Low	Low	Low
Marks (2013)	Some concern	Low	Some concerns	Low	Low	High
Melville (2015)	Low	Some concerns	Some Concerns	Low	Low	Some concerns
Ozmen (2007)	Some concern	Some concerns	Low	Low	Low	Some concerns
Rosety- Rodriguez (2014)	Low	Some concerns	Low	Some concerns	Low	Some concerns
Shield (2013)	Some concern	Low	Low	Low	Low	Some concerns
Suomi (1998)	Low	Some concerns	Low	Some concerns	Low	Some concerns
Van Schijndel Speet (2017)	Low	Some concerns	Some concerns	High	High	High

Table 1

Risk of bias assessment for included studies

Table 2

Performance measures comparison

Outcome or Subgroup Title	No. of studies	No. of participants	Statistical Method	Effect Size (95% CI)
1.1 Aerobic Fitness	8	333	Std. Mean Difference (IV, Random, 95% CI)	0.13 [-0.11, 0.37]
1.2 Balance	3	162	Std. Mean Difference (IV, Random, 95% CI)	1.25 [-0.39, 2.90]
1.3 Functional Fitness	5	325	Std. Mean Difference (IV, Random, 95% CI)	-0.07 [-0.60, 0.46]
1.3.1 Sit-to Stand	5	198	Std. Mean Difference (IV, Random, 95% CI)	0.37 [-0.20, 0.94]
1.3.2 Get up and Go	3	127	Std. Mean Difference (IV, Random, 95% CI)	-0.77 [-1.34, -0.19]
1.4 Musculoskeletal Strength	6	351	Std. Mean Difference (IV, Random, 95% CI)	0.70 [0.24, 1.16]
1.4.1 Upper body	4	223	Std. Mean Difference (IV, Random, 95% CI)	0.55 [-0.17, 1.26]

Table 2

1.4.2 Lower body	4	128	Std. Mean Difference (IV, Random, 95% CI)	0.86 [0.30, 1.42]
1.5 Flexibility	2	152	Std. Mean Difference (IV, Random, 95% CI)	-0.19 [-1.73, 1.34]
1.5.1 Upper Body	1	67	Std. Mean Difference (IV, Random, 95% CI)	0.08 [-0.40, 0.56]
1.5.2 Lower Body	2	85	Std. Mean Difference (IV, Random, 95% CI)	-0.30 [-3.16, 2.56]
1.7 Step Count	3	188	Std. Mean Difference (IV, Random, 95% CI)	0.30 [-0.15, 0.75]

Table 3

Body Composition Comparison

Outcome or Subgroup Title	No. of studies	No. of participants	Statistical Method	Effect Size (95% CI)
2.1 Weight	6	343	Std. Mean Difference (IV, Random, 95% CI)	0.13 [-0.12, 0.37]
2.2 Waist circumference	2	189	Std. Mean Difference (IV, Random, 95% CI)	0.06 [-0.37, 0.50]

Table 4

Cardiovascular Fitness Comparisons

Outcome or Subgroup Title	No. of studies	No. of participants	Statistical Method	Effect Size (95% CI)
3.1 Maximal Oxygen Uptake	4	109	Std. Mean Difference (IV, Random, 95% CI)	0.55 [0.17, 0.94]
3.2 Blood Pressure	3	242	Std. Mean Difference (IV, Random, 95% CI)	-0.30 [-0.56, -0.03]
3.2.1 Systolic	3	121	Std. Mean Difference (IV, Random, 95% CI)	-0.47 [-0.85, -0.10]
3.2.2 Diastolic	3	121	Std. Mean Difference (IV, Random, 95% CI)	-0.12 [-0.48, 0.23]
3.3 Heart Rate	3	89	Std. Mean Difference (IV, Random, 95% CI)	0.11 [-0.44, 0.65]

Table 5

Psychological Comparisons

Outcome or Subgroup Title	No. of studies	No. of participants	Statistical Method	Effect Size (95% CI)
4.1 Anxiety and Depression	3	140	Std. Mean Difference (IV, Random, 95% CI)	-3.07 [-6.81, 0.66]
4.2.1 Self Efficacy	2	152	Std. Mean Difference (IV, Random, 95% CI)	0.74 [-0.33, 1.80]