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Effects of exercise on fitness in adults with intellectual disability: a protocol of an overview of systematic reviews

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ABSTRACT

Introduction Adults with intellectual disability (ID) have lower physical fitness levels than their peers without disabilities, representing a risk to their health since physical activity and cardiorespiratory fitness are directly related to better health and quality of life. Therefore, it is essential to determine the effects that exercise can have on them, as adults with ID present high comorbidities and lower life expectancy, altogether with lower rates of physical activity. The current overview of systematic reviews aims to provide an outline of the exercise benefits in health-related and skill-related fitness in adults with ID.

Methods and analysis Research will be conducted in PubMed, CENTRAL, EMBASE, PEDro, SPORTDiscus and CINAHL. The search terms will be categorised through population (eg, adult, ID); intervention (eg, exercise, physical activity) and outcomes (eg, cardiorespiratory fitness, body composition, muscular strength, muscular endurance, flexibility, balance, power, speed, agility, coordination, mobility and reaction time). Each database will be searched from their earliest available record up to 30 September 2021.

Inclusion criteria will be: systematic reviews including at least one RCT that compare exercise interventions with a control group or another type of intervention; measure of fitness using objectives methods; inclusion of adults with ID (≥18 years old), and published in any language, with at least their abstract in English, Spanish, French and/or Portuguese.

Ethics and dissemination To our knowledge, our overview will be the first of its kind to address the topic in people with ID. The results could be used to determine which fitness components can be improved by exercise and to provide a valuable tool to develop comprehensive exercise programmes specific to people with ID. Ethical approval is not required. The knowledge generated will be disseminated electronically and in print and presented at conferences.

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Strengths and limitations of this study

► This is the first overview that will offer a comprehensive summary of the published systematic reviews analysing the effects of exercise in adults with intellectual disability (ID).
► The findings from this study will provide information to develop specific exercise programmes to improve physical fitness in adults with ID.
► All the systematic reviews will include at least one randomised controlled trial in order to establish a quality setting. Nevertheless, this could result in missing potentially relevant articles.
► A limitation of this overview is that the target population is adults with ID; therefore, the results should not be generalised to children and adolescents with ID.

INTRODUCTION

Intellectual disability (ID) is a disability characterised by significant intellectual functioning and adaptive behaviour limitations, which covers many everyday social and practical skills.[1] The criteria of onset of these limitations, which is the developmental period, is operationally defined as before the person attains the age of 22.[1] Adults with ID present different physical problems and comorbidities that situates this population at socioeconomic disadvantage.[2] It has been reported that diseases of the musculoskeletal system and connective tissue are present in 48.2% of people with ID.[1] That fact builds a big burden across lifespan development and ageing, which makes a deep impact on physical activity (PA) routines as well. In addition, one of the most important health conditions in older adults with ID is cardiovascular disease, which is present in 23.6% of older adults with ID.[2]

The functional and physical decline in people with ID occurs at earlier ages, around 40–50 years of age,[3 4] than in people who do not have a disability. This may cause a loss of physical fitness, an earlier onset of osteoporosis, diabetes, musculoskeletal disorders, dementia, hypertension and peripheral arterial disease.[5–7]

Although improved medical and healthcare have gradually increased the mean life expectancy of people with ID,[8] alternative and innovative solutions to maintain physical

function and improve health and quality of life should include PA and exercise training. Therefore, it is essential to determine the effects of exercise programmes on physical fitness in adults with ID.

Physical fitness is defined as the ability of someone to perform a determinate action/activity, a set of attributes that people have or achieve.[9] The lack of exercise and PA leads to low physical fitness levels in this population,[10] which declines even more with age. Thus, the deterioration of health associated with ageing is faster in individuals with ID than in persons without ID.[11]

In order to test the effects of exercise programmes and their impact on health and skills areas, these two domains are established: (1) health-related fitness (HRF), and (2) skill-related fitness (SRF).

According to the American College of Sports Medicine,[12] the fitness domains and the medical-related domains have common benefits whenever one of the two is improved. HRF includes cardiorespiratory fitness, anthropometry, muscular strength, muscular endurance and flexibility, whereas SRF includes power, agility, speed, balance, coordination, reaction time and mobility.[9]

The combination of both fitness' components is included in many PA programmes for people with ID for several purposes. HRF components are key in the appropriate body functioning, particularly for this population, because of their accelerated ageing and low physical conditioning. It is clear that worse levels within anthropometric variables (eg, fat mass and muscle mass) are adding morbidities to this population that alters daily life activities and health status. Besides, a higher body mass index (BMI) will not allow these subjects to move easily, creating a vicious circle within BMI, movement and health. Then, although the BMI can be considered normal weight, cardiorespiratory fitness, muscular strength and endurance will determine the capacity of people with ID to perform activities of daily living, both qualitatively (economy and quality of the movements) and quantitatively (intensity and duration). In addition, balance and gait performance are linked to the incidence of falls and fall-related injuries among people with ID, and its training has already demonstrated a decrease of fall risks[13] and balance improvements.[14]

Literature reports benefits on different fitness components, but as far as we know, no previous studies compile the whole HRF and SRF benefits that exercise can trigger. This overview will bring a gathering onto which components of fitness are improved and this will help professionals to recommend or prescribe the type, volume and intensity of exercise that adults with ID might require according to their needs.

Therefore, the main aim of this study is to develop an overview of the evidence to report the effects of exercise (intervention) on HRF and SRF (outcome) in adults with ID (population). Usual care, waiting list control, placebo/sham treatment, other treatment or a combination of treatments, as long as the effect of exercise could
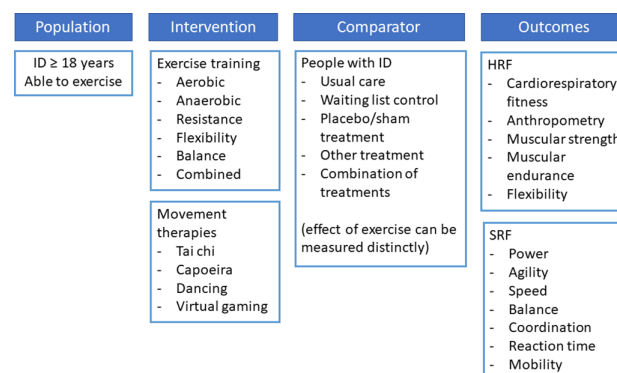

Figure 1 PICO components of the study. ID, intellectual disability; HRF, health-related fitness; PICO, population intervention comparator outcome; SRF, skill-related fitness.

be measured distinctly, will be the settings of comparison (control).

METHODS AND ANALYSIS
Design
This overview protocol has been reported following the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) Protocols guidelines[15] (online supplemental file 1). The ongoing overview will be reported following the updated PRISMA guidelines.[16]

The research will be conducted in the following electronic databases: PubMed, CENTRAL, EMBASE, PEDro, SPORTDiscus and CINAHL. In addition, the reference lists of included studies will be checked for study eligibility. PICO components can be seen in figure 1.

Inclusion and exclusion criteria
The inclusion criteria will be: (1) Systematic reviews will be selected as long as they include at least one RCT; (2) Study sample with participants with ID. If other populations are included, we will assess their eligibility as long as ID outcomes can be measured distinctly from non-ID individuals; (3) Systematic reviews that compare exercise interventions no matter types with usual care, waiting list control, placebo/sham treatment or other treatment as long as the effect of exercise can be measured and (4) Adult population. Study sample 18 years old or over. For studies that included adolescents and adults, at least 80% of the total sample must be 18 years or over; (5) For studies including samples with different types of disabilities, studies will be included if the proportion of participants with ID is reported; (6) Measures of fitness using objective methods and (7) Published in any language, with at least their abstract in English, Spanish, French and/or Portuguese.

Exclusion criteria will be: (1) The only available record is a conference abstract or poster; (2) The paper reported a lab-based study, for example, to calibrate accelerometer cut-offs; (3) The systematic review does not have any RCT; (4) The systematic review does not report the ID level of the participants and (5) Grey literature.

Population

We will include adults (≥18 years) with ID. There are no limitations on the participant's characteristics (such as sex, comorbidity and treatment course).

Intervention/control

We will include systematic reviews that examined the effects of any exercise and/or sport training (eg, aerobic; anaerobic; resistance; flexibility; balance) and/or movement therapies (eg, tai chi, capoeira, dancing or virtual gaming) for adults with ID. Usual care, waiting list control, placebo/sham treatment, other treatment, or a combination of treatments (as long as the effect of exercise could be measured distinctly) in people with ID will be used as comparators.

Context

No limits will be set for cultural or geographical settings. We will take into account that samples might be selected among different conditions (residential care, community care, occupational centres, home care…), but no restrictions will be applied to specific external services and/or facilities.

Outcomes

The main outcomes of this overview will be HRF components (eg, cardiorespiratory fitness, anthropometry, muscular strength, muscular endurance and flexibility) and SRF components (eg, power, agility, speed, balance, coordination, reaction time and mobility). The outcomes reported by the included articles must have been assessed quantitatively at least twice (eg, baseline and postintervention). There are different ways of measuring physical fitness components, as shown in figure 2, so we assume the results could be heterogeneous.

Search strategy

The search terms will be categorised as population (eg, adult, ID), intervention (eg, exercise, PA) and outcomes (eg, body composition, strength, cardiorespiratory fitness, flexibility, balance, endurance, speed…).

Figure 2 Examples of outcomes' variables and tests that could be obtained in the data extraction. BIA, bioimpedance analysis; BMI, body mass index; DEXA, dual X-ray absorptiometry; RER, respiratory exchange ratio; VO_2, volume of oxygen uptake.
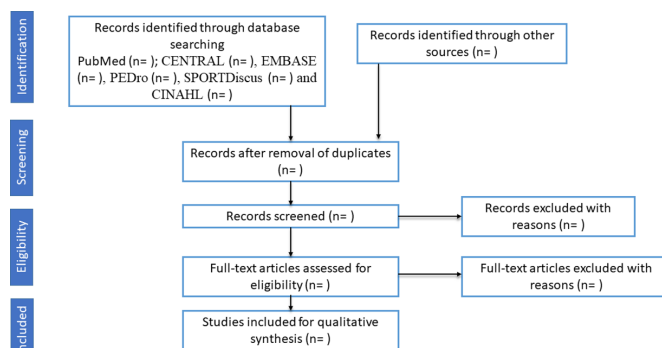

Figure 3 PRISMA flow chart. PRISMA, Preferred Reporting Items for Systematic Reviews and Meta-Analyses.

Syntaxis for PubMed search can be seen in online supplemental file 2. Each database will be searched from its earliest available record up to the 30 September 2021. No language restriction will be applied; nevertheless, the abstract should be published at least in one of the following languages: English, Spanish, French or Portuguese.

Data selection

After the search in databases, all references will be transferred to a citation manager software (Mendeley Desktop V.1.19.8) to identify and remove all duplicate articles. A manual check of all references will be performed to ensure that all duplicates have been removed.

The data processing will be carried out in two steps, by pairs: (1) reading of all titles and abstracts, (2) reading all articles in full. In the first step, we will discard or select the articles according to our inclusion and exclusion criteria only reading titles and abstracts. In the second step, we will decide the inclusion by reading the full text. This will allow for the appropriate screening, eligibility and inclusion. An exclusion and inclusion detailed list will be provided. All results will be checked in pairs (GRO, MF-F, MG-B and SSS), and disagreements will be solved by a third author (MBN-A). The process will be described in a PRISMA flow chart[16] as can be seen in figure 3.

Data extraction and synthesis

We will design a table that will include all data. Two independent authors (SSS and MF-F) will independently extract the characteristics and outcomes data from the studies that will be included. Two reviewers (GRO and MG-B) will check the data collected in the previous stage. Any disagreements will be solved by discussion within the whole team.

The following domains will be extracted: study characteristics (main author, publication year and journal, systematic review or meta-analysis), aims, funding, risk of bias assessment, databases, languages, intervention, participants (age range, mean age, sex, ID level, intervention group, control group, design setting, country, comorbidities and inclusion/exclusion criteria), outcomes (HRF and SRF objective measures), intervention (type of exercise, length, frequency, number of sessions and duration

of each session), number of RCT, results, conclusions and included studies. The overlap of the primary studies included in each review will be checked and analysed.

Finally, we will create a table with a descriptive synthesis and a summary of the findings.

Quality assessment

The AMSTAR2 tool[17] will be used to assess the risk of bias of each systematic review included by duplicate (GRO and MF-F; MG-B and SSS). Discrepancies will be solved by a third author (MBN-A). A pre-established form will be used to fill in the results from the assessment. Authors will agree on critical domains in order to establish confidence criteria for the reviews: high, moderate, low and critically low. A coloured chart and a comprehensive discussion of the results will be given.

Strength of evidence assessment

In order to assess the strength of the body evidence across studies, we will use the Grade of Recommendations Assessment, Development and Evaluation (GRADE) tool[18] to assess and report the certainty of the evidence for each outcome of interest obtained. Depending on the results obtained from the data extraction, GRADE degrees will be obtained directly from each systematic review, or, in case that data is missing, we will assess GRADE strength of recommendation.[19 20] The level of evidence obtained will be high, moderate, low or very low.

Patient and public involvement statement

No patient involved.

DISCUSSION

We will not provide quantitative analysis between the results of the systematic reviews. In contrast, we will use different tools and agreements to be as accurate and objective as possible for developing this protocol.

First, our data will always be screened and studied for eligibility and inclusion by pairs. Percentages of agreement will be specified. To avoid the risk of bias in individuals' studies, we will apply the AMSTAR2 tool to assess the study quality. Nevertheless, we will not be able to analyse publication bias as no funnel plots will be performed, so we will not have tools to determine if negative results have not been published, ensuring that only the positive outcomes are shown in the selected articles. We do not expect to detect selection bias related to language as we assume that no language restriction is established, except for the abstract and title search. Finally, GRADE assessments will be performed to determine the strength of body evidence.

The low fitness levels of adults with ID may contribute to an increased risk of suffering physical dysfunction and mobility disability.[4 7] Our study will also be conditioned by the conceptualisation of ID due to differences between sociocultural issues that are found in different countries, as the population with ID is very heterogeneous.

As far as we know, this will be the first overview of systematic reviews assessing physical and exercise programmes to improve the HRF and SRF on adults with ID.

If exercise interventions could be established as a treatment based on existing scientific evidence to improve the fitness of adults with ID, healthcare systems and providers will hold a powerful and economic weapon to fight multimorbidities and enhance the health of people with ID.

It is essential to determine the current effectiveness of exercise in both HRF and SRF, to support the care of people with ID. Likewise, this overview will help to identify gaps in the knowledge regarding the effects of exercise on the components of fitness mentioned previously. Furthermore, we would like to make recommendations for future studies based on the results from our overview.

ETHICS AND DISSEMINATION

As this is an overview of systematic reviews, ethical approval is not required. The knowledge generated will be disseminated electronically, in print and presented at conferences relevant to ID.

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Supplemental file 2

1. "Intellectual disabilit*"

2. "intellectual development disorder"

3. "learning disabilit*"

4. "mental retardation"

5. "cerebral palsy"

6. "genetic syndrome"

7. "down syndrome"

8. "trisomy 21"

9. "autism spectrum disorder"

10. "autistic disorder"

11. "pervasive developmental disorder not otherwise specified"

12. "fetal alcohol syndrome"

13. "fragile X syndrome"

14. "Prader-Willi syndrome"

15. "Rett syndrome"

16. 1 OR 2 OR 3 OR 4 OR 5 OR 6 OR 7 OR 8 OR 9 OR 10 OR 11 OR 12 OR 13 OR 14 OR 15

17. Exercise

18. "exercise therapy"

19. "physical activity"

20. fitness

21. training

22. sports

23. 17 OR 18 OR 19 OR 20 OR 21 OR 22

24. "physical conditioning"

25. "physical fitness"

26. "health-related fitness"

27. "skill-related fitness"

28. "cardiorespiratory fitness"

29. "body composition"

30. "weight loss"

31. Strength

32. Flexibility

33. Endurance

34. Speed

35. Agility

36. Balance

37. Power

38. "reaction time"

39. Coordination

40. Mobility

41. 24 OR 25 OR 26 OR 27 OR 28 OR 29 OR 30 OR 31 OR 32 OR 33 OR 34 OR

    35 OR 36 OR 37 OR 38 OR 39 OR 40

42. 16 AND 23 AND 41

43. Limit to systematic reviews AND metaanalysis

Supplemental file 1

Preferred reporting items for systematic review and meta-analysis protocols (PRISMA-P)			
Section and topic	**Item No**	**Checklist item**	**Page/line numbers**
Section 1: Administrative information			
Title:			
Identification	1a	Identify the report as a protocol of a systematic review	Page 1
Update	1b	If the protocol is for an update of a previous systematic review, identify as such	None
Registration	2	If registered, provide the name of the registry (such as PROSPERO) and registration number	Page 3
Authors:			
Contact information	3a	Provide name, institutional affiliation, and email address of all protocol authors; provide physical mailing address of corresponding author	Page 1
Contributions	3b	Describe contributions of protocol authors and identify the guarantor of the review	Page 11
Amendments	4	If the report represents an amendment of a previously completed or published protocol, identify as such and indicate what changes were made; otherwise, state plan for documenting important protocol amendments	None
Support:			
Sources	5a	Indicate sources of financial or other support for the review	Page 11-12
Sponsor	5b	Provide name of the review funder and/or sponsor	Page 11-12

Role of sponsor and/or funder	5c	Describe roles of funder(s), sponsor(s), and/or institution(s), if any, in developing the protocol	Page 11-12
Section 2: Introduction			
Rationale	6	Describe the rationale for the review in the context of what is already known	Pages 3-5
Objectives	7	Provide an explicit statement of the question(s) the review will address with reference to participants, interventions, comparators, and outcomes (PICO)	Page 5-6
Section 3: Methods			
Eligibility criteria	8	Specify the study characteristics (such as PICO, study design, setting, time frame) and report characteristics (such as years considered, language, publication status) to be used as criteria for eligibility for the review	Pages 6-8
Information sources	9	Describe all intended information sources (such as electronic databases, contact with study authors, trial registers or other grey literature sources) with planned dates of coverage	Page 6
Search strategy	10	Present draft of search strategy to be used for at least one electronic database, including planned limits, such that it could be repeated	Page 8 Suppl file 2
Study records			
Data management	11a	Describe the mechanism(s) that will be used to manage records and data throughout the review	Page 8-9
Selection process	11b	State the process that will be used for selecting studies (such as two independent reviewers) through each phase of the review (screening, eligibility, and inclusion in meta-analysis)	Page 8

Supplemental material

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Data collection process	11c	Describe planned method of extracting data from reports (such as piloting forms, done independently, in duplicate), any processes for obtaining and confirming data from investigators	Page 9
Data items	12	List and define all variables for which data will be sought (such as PICO items, funding sources) and any pre-planned data assumptions and simplifications	Page 7-8 Figure 2
Outcomes and prioritisation	13	List and define all outcomes for which data will be sought, including prioritisation of main and additional outcomes, with rationale	None
Risk of bias individual studies	14	Describe anticipated methods for assessing risk of bias of individual studies, including whether this will be done at the outcome or study level, or both; state how this information will be used in data synthesis	Page 9
Data synthesis	15a	Describe criteria under which study data will be quantitatively synthesised	None
	15b	If data are appropriate for synthesis, describe planned summary measures, methods of handling data, and methods of combining data from studies, including any planned exploration of consistency (such as I^2, Kendall's τ)	None
	15c	Describe any proposed additional analyses (e.g., sensitivity or subgroup analyses, meta-regression)	None
	15d	If quantitative synthesis is not appropriate, describe the type of summary planned	Page 9

| Meta-bias(es) | 16 | Specify any planned assessment of meta-bias(es) (such as publication bias across studies, selective reporting within studies) | None |
| Confidence in cumulative estimate | 17 | Describe how the strength of the body of evidence will be assessed (such as GRADE) | Page 9-10 |