STUDY PROTOCOL

Sedentary behaviour levels in adults with an intellectual disability: a systematic review protocol [version 1; peer review: 2 approved with reservations]

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Abstract

Background: Sedentary behaviour contributes to non-communicable diseases, which account for almost 71% of world deaths. Of these, cardiovascular disease is one of the largest causes of preventable death. It is not yet fully understood what level of sedentary behaviour is safe. People with an intellectual disability have poorer health than the general population with higher rates of multi-morbidity, obesity and inactivity. There is a paucity of evidence on whether this poorer health is due to sedentary behaviour or physical inactivity. This systematic review will investigate the sedentary behaviour levels of adults with an intellectual disability.
Method: The PRISMA-P framework will be applied to achieve high-quality articles. An extensive search will be conducted in Medline, Embase, psycINFO and Cinahl and grey literature sources. All articles will be independently reviewed by two reviewers and a third to resolve disputes. Initially, the articles will be reviewed by title and abstract and then the full article will be reviewed using stringent inclusion criteria. All article data will be summarised in a standardised tabular format. The National Institute of Health's quality assessment tool will be used to assess article quality. GRADE will be used to assess the quality of the evidence. The primary outcome of interest is the prevalence of sedentary behaviour levels for people with an intellectual disability. The definition of sedentary behaviour to be used for the purposes of this study is: 'low physical activity as identified by metabolic equivalent (MET) or step levels or as measured by the Rapid Assessment of Physical activity questionnaire (RAPA) or the International Physical Activity questionnaire (IPAQ) or sitting for more than 3 hours per day'.
Conclusion: This systematic review will provide a critical insight into the prevalence of sedentary behaviour in adults with an intellectual disability.

Keywords

Intellectual disability, sedentary behaviour, adults

Open Peer Review

Reviewer Status ? ?

	Invited Reviewers	
	1	**2**
version 2 (revision) 29 Mar 2021		
version 1 26 Aug 2020	? report	? report

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Any reports and responses or comments on the article can be found at the end of the article.

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Introduction

Rationale

According to the World Health Organisation (WHO, 2013), non-communicable diseases account for almost 71% of world deaths. Non-communicable diseases are non-infectious and chronic but can be prevented. Of these, cardiovascular disease (CVD) is one the largest causes of preventable death worldwide with over 17.9 million dying annually. CVD can manifest as increased blood pressure or elevated blood lipid levels, leading to heart attack or stroke. One of the main contributors to CVD is lack of physical activity (Forouzanfar *et al.*, 2016). Physical activity is any bodily movement which uses skeletal muscles and results in energy expenditure (WHO, 2019) while a sedentary lifestyle is one which has low levels of physical activity and consequently low levels of energy expenditure. In general, people with intellectual disability (ID) have poorer health than their non-disabled contemporaries (Emerson *et al.*, 2016) and often experience health disparities (Krahn & Fox, 2014). However, the real state of the science regarding sedentary behaviour and people with ID is not known. Further investigation is essential to understand if sedentary behaviour contributes to these health differences.

It is necessary to understand some of the known contributors to CVD, obesity and physical inactivity, as well as sedentary behaviours because these are all modifiable and inter-related health risks factors.

Sedentary behaviour. Sedentary comes from the Latin word *sedere* which means to sit and can describe a wide range of distinct activities which require low levels of energy expenditure in any setting (Thorp *et al.*, 2011). The first real attempt to define the term 'sedentary' was made in 2012 (Tremblay *et al.*, 2017). This was in an effort to avoid confusion by standardising the terms to refer to sedentary or inactive behaviours used in journals. A metabolic equivalent (MET), known as the resting metabolic rate, is an objective measurement scale used to classify activity types and levels. A MET is the amount of oxygen (O_2) burned at rest and is the equivalent of 3.5ml O_2 per kg body weight per minute (Jette *et al.*, 1990) or 1kilocalorie per kg of body weight per hour (Newton *et al.*, 2013). Tremblay *et al.* (2017) proposed to define sedentary behaviour as 'any waking behaviour characterized by an energy expenditure of ≤1.5 METs while in a sitting or reclining posture' for example watching television or working on a computer. Hence sedentary behaviour constitutes too much sitting or stationary activity as opposed to physical inactivity which is too little exercise or physical movement. Tudor-Locke *et al.* (2013) found a link between reduced steps per day (less than 5,000) and being more sedentary. In addition, sitting for prolonged periods (more than 3 hours per day) has been found to have adverse health effects (Pinto Pereira *et al.*, 2012).

Sedentary behaviour has been linked to adverse health conditions in older adults, increased cardio metabolic risks, increased obesity and mortality in both men and women, as well as increased cancer risk (de Rezende *et al.*, 2014; Patel *et al.*, 2010; Same *et al.*, 2016; Thorp *et al.*, 2011;). Self-reported studies have shown that high levels of sedentary behaviour, even if minimum exercise guidelines are met, show increased metabolic risk (Patel *et al.*, 2010). This impact of sedentary behaviour can be mollified by interspersing periods of physical activity throughout the day (Healy *et al.*, 2008).

An ecological model of sedentary behaviour for older adults without an ID, proposed by Owen *et al.* (2011), could be used to assess the sedentary behaviours of people with ID. This model classed sedentary behaviour into four categories:

- Household (e.g. watching TV)
- Leisure time (increased screen-based and sitting activities)
- Transport (driving, sitting on public transport to/from work/activities)
- Occupation (e.g. screen-based computer work).

Sedentary behaviour and people with ID

In a systematic review by Melville *et al.* (2017), it was proposed that studies to determine sedentary behaviour in people with ID did not use enough randomly selected samples and sample sizes were too small, meaning that results could not be generalised for the ID population as a whole. Furthermore, insufficient studies have distinguished between sedentary behaviour and inadequate physical activity. Consequently, it is not clear what the actual sedentary behaviour of people with ID is.

Older people with an intellectual disability have been shown to have higher rates of multi-morbidity, obesity and inactivity than the general population (Gawlik *et al.*, 2018; McCarron *et al.*, 2013). In 2016 approximately 70,000 people, 1.4% of the overall Irish population (Census, 2016), were shown to have an ID. In an analysis of secondary data, Harris *et al.* (2017) deduced that people with ID were sedentary for over 70% a day. According to Graham & Reid (2000), adults with ID are more susceptible to age-related health risks, where sedentary behaviour could be a contributing factor.

While breaking up time spent doing sedentary activity has been shown to increase daily living activities and physical independence in older adults (Sardinha *et al.*, 2015), there is no similar information on adults with ID. Increased sedentary behaviour has been linked to obesity levels and increased likelihood of multi-morbidity (Melville *et al.*, 2017), but inconsistent evidence exists on links of sedentary behaviour to level of ID (Oppewal *et al.*, 2018). Often studies use proxy measures (e.g. watching TV) to determine sedentary behaviour which may be inaccurate, especially with regards to people with ID, as people with a more severe ID may be less likely to watch TV due to sensory or cognitive impairments (Owen *et al.*, 2011). Level of ID has been shown to be directly related to physical activity but not sedentary behaviour (Oppewal *et al.*, 2018).

Emerging evidence is highlighting the importance of reducing sedentary behaviour for improving cardio-metabolic health and adopting a holistic public health approach to improve activity levels as well as sedentary behaviour (van der Ploeg & Hillsdon, 2017).

For the purposes of this systematic review, sedentary behaviour will be defined as:

'low physical activity as identified by MET or step levels or as measured by the Rapid Assessment of Physical activity questionnaire (RAPA) or the International Physical Activity questionnaire (IPAQ) or sitting for more than 3 hours per day'

Obesity. Globally almost 38% of the world's population, a greater than 100% increase since 1980, and two-thirds of the American population, are either overweight or obese, with a BMI of greater than 25.0 kg/m^2 (Fryar *et al.*, 2012; Ng *et al.*, 2014; WHO, 2009). In Ireland, almost 23% of adults are obese with 50% of women and 66% of men being overweight (Ng *et al.*, 2014. This is a huge concern given the proven link between obesity and cancers, higher rates of type II diabetes, CVD and CVD mortality (Bhaskaran *et al.*, 2014; Hossain *et al.*, 2009; Ortega *et al.*, 2016).

Obesity and people with ID

A 2017 review found that the prevalence of overweight and obesity in people with ID varies from 28%–71% and 17%–43%, respectively (Ranjan *et al.*, 2018). The IDS-TILDA study found that overweight and obesity in people with ID increased from 66% in wave2 to 79.7% in wave3 and that 64% of participants considered themselves to be at the right weight (Burke *et al.*, 2017). According to a US based longitudinal study on people with ID women appear to be at a greater risk of developing morbid obesity while men were more likely to be overweight (Hsieh *et al.*, 2014)).

According to Fock & Khoo (2013), excessive calorific intake and increased sedentary behaviour are the main contributors to increased obesity levels but obesity levels may be ameliorated by a combination of healthy eating, a reduction in sedentary behaviour and an increase in physical activity

Physical inactivity. Physical inactivity is classified as not meeting the minimum activity requirements. According to the American College of Sports Medicine, moderate-intensity aerobic physical activity (PA) of between 150 and 250 minutes per week is the minimum necessary for health and weight management in adults (Donnelly *et al.*, 2009; Health Service Executive, 2009; US Department of Health, 2018). Insufficient PA or physical inactivity contributes to adverse health issues like obesity, CVD and cancer as well as increased mortality (Lee *et al.*, 2012). According to the World Health Organisation (WHO, 2009), physical inactivity is the fourth leading risk factor for all-cause mortality, with over three million deaths annually. Of concern is that Ireland is one of the least active countries in Europe (Loyen *et al.*, 2016).

Physical inactivity and people with ID

For People with an ID, the amount of moderate PA done, and the number of hours spent watching TV was found to be significantly associated with obesity level (Hsieh *et al.*, 2014). A 2016 Australian based study found that over 66% of participants did not meet minimum exercise guidelines (Koritsas & Iacono, 2016), while another study found that 77% of participants did not meet the minimum exercise recommendations (Barnes *et al.*, 2013). It must be noted that physical impairments leading to the use of walking aids or wheelchairs may inhibit physical activity for some people with an ID (Ranjan *et al.*, 2018).

Hence sedentary behaviour and physical inactivity are different and should be addressed separately with distinct guidelines for each. While specific recommendations for movement and physical activity levels in adults have been long established, corresponding recommendations for sedentary behaviour have not. The recommendations from emerging evidence are to minimise the amount of time being sedentary, but no specifics have yet been established for the general population or people with ID (WHO, 2020).

Developing the question

A focused and well-defined question avoids bias in literature searches, ensures clarity and therefore ensures the identification of the concepts for the focused search. PICo, which is used for qualitative studies is being used to define the question as follows (Schardt *et al.*, 2007):

- P [Population or problem]: Adults aged 18+ with an Intellectual Disability
- I [Interest]: Sedentary behaviour level
- C [Context]: Sedentary behaviour in line with the definition of sedentary behaviour as defined for this review.

The research question to be addressed by this systematic review protocol is

'What are the sedentary behaviour levels of older Adults with an Intellectual Disability?'.

Methods

PRISMA-P, for the reporting and development of systematic review protocols is used as the guide in the writing of this protocol (Shamseer *et al.*, 2015). The completed PRISMA-P checklist for this protocol is available as extended data (Lynch, 2020).

Eligibility criteria

The criteria for inclusion in the review are as follows:

- Population: adults aged 18+ with an Intellectual Disability
- Language: English
- Study type: All types of studies including primary studies, peer reviewed, grey literature
- Study design: Randomised controlled trials, cohort, cross-sectional
- Content: Must reference sedentary behaviours of adults with ID to be eligible for inclusion
- Timeframe: no restriction on timeframes up to March 2020.

The criteria for exclusion in the review are as follows:

- Population: Children with or without an ID and Adults without ID
- Language: Articles that are not available in English
- Study design: Any type of reviews
- Conference proceedings and published conference abstracts only

Information sources

Databases

The following four databases will be used to perform the search:

Medline

Embase

psycINFO

Cinahl

In addition, the following sources will be explored for grey literature sources:

The CORDIS library

Grey Literature Database from the Canadian Evaluation Society

The U.S. Department of Housing and Urban Development (HUD) User database

National Technical Information Service (NTIS)

Open Grey

Social Care Online

Social Science Research Network (SSRN) eLibrary

RIAN

Google Scholar

Proquest (Dissertations and Theses)

Search strategy

The search strategy was refined into two concepts following the application of PICo. Concept 1 is 'Sedentary behaviour or inactivity' and Concept 2 is 'Intellectual Disability'. Each of the two concepts will be searched using MESH terms and keywords and then combined using OR. Then the total results of each concept will be combined using AND (See Figure 1). This search will be repeated for each of the four databases. The resulting article list will be the complete combined database search results. This list will be screened for inclusion.

Search string. An example of the search string used for the Medline database is shown in Table 1.

Screening process. All identified articles from each database that is searched, as well as all grey literature sources, will be combined and duplicates removed. Endnote software will be used to store all the identified articles. The articles will be stored in folders which are named after the search process used. Using the inclusion criteria as detailed above, all articles will initially be screened by title and then by abstract. The remaining full text articles will be retrieved and read thoroughly. Those that do not meet the inclusion criteria will be omitted. The remaining articles will then be quality assessed using two separate assessors with a third person as an adjudicator should any discrepancies arise.

Quality assessment and risk of bias. The remaining articles will be assessed using a quality assessment tool for observational cohort and cross-sectional studies from the National Institute of Health. The tool used is available as extended data (Lynch, 2020).

These tools are used to critically assess the internal validity of each article and identify any issues or sources of potential bias. According to Cochrane, effectively evaluating the quality of a study is done by looking at its design, methodology, results, analysis and reporting, and how they relate to the original research question (Higgins *et al.*, 2011).

There are different types of study quality assessment tools for the different study types. For Controlled Intervention Studies and Observational Cohort and Cross-sectional studies, 14 criteria are used to evaluate the study quality, while for Case-Control studies 12 criteria are used. This means that a

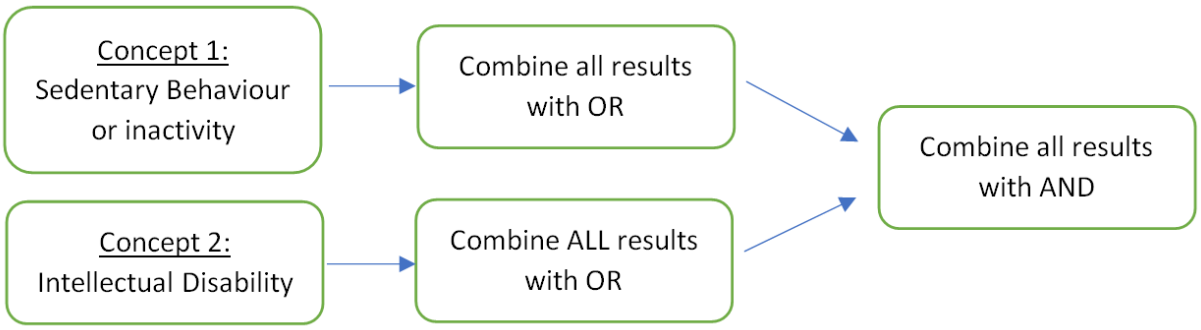

Figure 1. Search strategy.

Table 1. Medline search string.

Concept	Index	Keywords
Concept 1: Sedentary behaviour & physical inactivity	(MH "Sedentary Behavior")	sedentary lifestyle* OR sedentary behavior* OR sedentary behaviour* OR physical* inactiv* OR inactive lifestyle
Concept 2: Intellectual disability or learning disability	(MH "Intellectual Disability+") OR (MH "Learning Disabilities+")	((intellectual AND disabilit* OR 'mental retardation'/exp OR 'mental retardation' OR (mental AND ('retardation'/exp OR retardation)) OR 'learning'/exp OR learning) AND disabilit* OR developmental) AND disabilit* OR 'learning disabilities'/exp OR 'learning disabilities' OR (('learning'/exp OR learning) AND disabilities)

maximum quality score of 12-14 can be achieved. This quality score will be used to determine if the study should be included in the review. Quality scores are divided into 3 main categories: Good, Fair or Poor. See Table 2 for details.

Any studies that are excluded will be tracked with reasons for rejection.

Quality scoring

Scores are attributed to distinct parts of the study design for example type of study, design and blinding, where a 'yes' answer gives a score of '1', a 'no' answer a score of '0' and could potentially highlight an issue with the article. See Table 3.

Table 2. Quality assessment Scoring System.

Quality Rating	Observational Cohort & Cross-Sectional Studies	Case-Control Studies	Action
Good	9 - 12	10 - 14	Data extraction
Fair	6 - 8	7 - 9	2 reviewers to discuss. Adjudicate with 3rd reviewer if required.
Poor	<=5	<= 6	2 reviewers to discuss. Reject
Other	CD, NR, NA *		

* CD = Cannot determine, NR = Not reported, NA = Not applicable

Table 3. Study assessment scoring.

Answer	Score
Yes	1
No	0
Cannot determine/not reported/not applicable	0

Ethics

This research project is part of the IDS-TILDA project. Full ethical approval for IDS-TILDA has been granted by the Trinity College Dublin Faculty of Health Sciences Research Ethics Committee.

Study records

Data management

All search records will be kept in an excel spreadsheet detailing the database, type of search (keyword or MESH terms) and the resulting search numbers. The articles will be stored in Endnote. Each stage of the search and review will be recorded in excel. For each stage of the search process, articles will be stored in an appropriately named folder in EndNote X9 for windows.

Selection process

The selection process of studies for inclusion, which are identified by the search strategy, will be done by two independent review authors [LL and EB]. The initial screening will be done by title and abstract. If eligibility is inconclusive from the title and abstract, the full text of the article will be assessed. Any articles that do not match the inclusion criteria will be excluded. Any differences on article inclusion between the two authors will be resolved by discussions with the third review author [MMc]. Finally, the full-text article of all potential articles that could be included in the review will be independently assessed by the authors for inclusion as above.

Data collection process

An excel spreadsheet will act as the data extraction tool. This will be used to summarise all the shortlisted studies. The categories to be captured are as in Table 4.

Data items

The PICo framework will be used to define what data will be sought from variables as follows:

- P: Adults with an Intellectual Disability
    - Age, gender, living circumstance, country, number in study, level of ID

- I: Sedentary behaviour
  - Level, types of behaviour, quantify change
- C: Level of sedentary behaviour or physical inactivity
  - Level, intensity, types of activity/sedentary behaviour, type of employment

Outcomes and prioritisation

The outcomes of this investigation into sedentary behaviour will determine the sedentary behaviour levels of older adults with an intellectual disability.

Primary outcome

- Sedentary behaviour levels

Data synthesis

All article data will be summarised in a spreadsheet format as seen in Table 4 (McKenzie *et al.*, 2019). If studies are homogenous in nature a meta-analysis may be performed and a forest plot produced to summarise results. A narrative synthesis will be used to summarise all the study article data and relevant information. A thematic analysis of the semantic and latent topics of the articles using a 6-step process (see Table 5), will guide the derivation of a framework for the analysis of the outcome data (Braun & Clarke, 2006).

Statistical comparisons of article data will be reviewed on a case-by-case basis.

Confidence in cumulative evidence

The GRADE (Grading of Recommendations Assessment, Development and Evaluation) approach will be used to assess the strength of the body of evidence of the review. In line with the Cochrane methodology, each outcome will be ranked according to whether the quality is high, moderate, low or very low. The GRADE framework will be used to assess each outcome in the following areas: risk of bias, consistency of effect, imprecision, indirectness and publication bias (Schünemann *et al.*, 2019).

Table 4. Article Data Collection Categories.

Author, title, year
- study focus
- study type
- Intervention type
- country
- duration
- dates
- numbers
Participants
- number
- mean age
- gender (%)
- level of ID
- living circumstances
- employment type
Assessment
- type
- intervention
- Assessment type
- measurement device
- outcome/data
- Statistical results
Findings
Summary
Comment

Table 5. 6-step thematic analysis process.

Step number	Process	Explanation
1	Data familiarisation	Complete data immersion
2	Generate initial codes	Topics, patterns of data
3	Search for themes	Broader theme identification
4	Review of themes	Theme refinement
5	Define and name themes	Categorise. Include sub-themes if required
6	Produce report	Complete write-up

Dissemination of information
The dissemination plan will be to present at conferences for example the THEconf March 2021, Irish Gerontology Society PhD event and other ID or physical activity events or conferences as well as publishing in journals.

Study status
Searches are currently in progress.

Conclusion
This systematic review of the sedentary behaviour levels of older adults with an intellectual disability will provide a critical insight into the sedentary behaviours of this population group.

Data availability
Underlying data
No data are associated with this article

Extended data
Harvard Dataverse: Replication Data for: Sedentary behaviour levels in adults with an intellectual disability: a systematic review protocol. https://doi.org/10.7910/DVN/TPS2HU (Lynch, 2020)

This project contains the following extended data:

- Study Quality Assessment Tools_sysrevprotocol.odt (Assessment tool to be used)

Reporting guidelines
Harvard dataverse: PRISMA-P checklist for 'Sedentary behaviour levels in adults with an intellectual disability: a systematic review protocol' https://doi.org/10.7910/DVN/TPS2HU (Lynch, 2020)

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Open Peer Review

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Thessa Hilgenkamp
Department of Physical Therapy, University of Nevada, Las Vegas, NV, USA

This protocol for a systematic review addresses a relevant and important topic for individuals with intellectual disabilities. Due to the current interest in this topic, this update of the previous 2017 systematic review on this topic is likely to include some newer studies published in the last 3 years. The search strategy is very inclusive, and the inclusion and exclusion criteria are clearly described. However, the quality assessment and risk of bias is not clearly described, which is a key element to a systematic review protocol. The tool included as extended data is a tool to evaluate the quality of a systematic review as a whole, not to assess the papers included in a systematic review. The authors then mention there are different types of study quality assessment tools for the different study types, and a number of criteria used to evaluate those study types, but it is not clear whether this is based on an existing and validated tool, or a self-designed list of criteria, nor is it clear which criteria these lists consist of. Referencing this tool or making this tool/list available as extended data would greatly improve the replicability of the methods.

Is the rationale for, and objectives of, the study clearly described?
Yes

Is the study design appropriate for the research question?
Yes

Are sufficient details of the methods provided to allow replication by others?
No

Are the datasets clearly presented in a useable and accessible format?
Not applicable

Competing Interests: No competing interests were disclosed.

Reviewer Expertise: Physical activity, fitness, intellectual disabilities, Down syndrome, exercise

physiology.

I confirm that I have read this submission and believe that I have an appropriate level of expertise to confirm that it is of an acceptable scientific standard, however I have significant reservations, as outlined above.

Author Response 13 Mar 2021

Louise Lynch, Trinity College, Dublin, Ireland

Thank you for taking the time to review my protocol for a Systematic review of sedentary behaviour in adults with an intellectual disability.

The correct National Institute Health (NIH) tools for observational, cohort, cross-sectional and randomised controlled trials have now been included in the extended data and the tool for assessing systematic reviews removed. In addition a reference which shows all quality assessment tools by the NIH is included. Furthermore the criteria for RCTS has been added to the table. Wording has been changed to highlight that the tools being used for quality assessment are validated tools.

Competing Interests: None

Reviewer Report 07 January 2021

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Catherine Ruth Hankey
College of Medical Veterinary and Life Sciences, Institute of Mental Health and Wellbeing, University of Glasgow, Glasgow, UK

This is an interesting topic - and the methods that have been proposed are robust. However, my concerns are that the topic of physical activity in those adults with ID has already been recently examined by Westrop SC in 2019[1]. This work, a systematic review, I would expect to have examined much the same area that is proposed in this proposed new systematic review. The Westrop publication has not been included in this protocol and should be.

Having looked at the published research - it seems that there are no systematic reviews in those children and young adults with ID with regard to their physical activity.

I think that the application is well written and clear despite being very similar to the Westrop work which was published last year. The field of ID is a small one and I imagine that there are not sufficient new works to lead to any differing conclusions than those of Westrop. However, I

understand that the HRB are keen to support reanalysis of literature and therefore indexing of this protocol may be justified.

Thank you for inviting me to review this work.

References

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Is the rationale for, and objectives of, the study clearly described?

Yes

Is the study design appropriate for the research question?

Yes

Are sufficient details of the methods provided to allow replication by others?

Yes

Are the datasets clearly presented in a useable and accessible format?

Not applicable

Competing Interests: No competing interests were disclosed.

Reviewer Expertise: Research experience in the field of lifestyle in those adults with ID.

I confirm that I have read this submission and believe that I have an appropriate level of expertise to confirm that it is of an acceptable scientific standard, however I have significant reservations, as outlined above.

Author Response 13 Mar 2021

Louise Lynch, Trinity College, Dublin, Ireland

Thank you for taking the time to review my protocol for a systematic review on the sedentary behaviour levels in adults with an intellectual disability.
The Westrop Systematic review has now been included in the introduction to this protocol. The Westrop review focussed on the gender differences of physical activity and sedentary behaviour while this systematic review will look at the overall prevalence of sedentary behaviour. In addition this systematic review will endeavour to do comparisons by age, level of ID and residence if studies are available. This review will include a comprehensive grey literature search as well as any updated articles from the standard database search. Hence it will add to the body of evidence on sedentary behaviour which is sparse for people with ID.

Competing Interests: none