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# Informant-based assessment instruments for dementia and their measurement properties in persons with intellectual disability: a systematic review protocol



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# Title: Informant-based assessment instruments for dementia and their measurement properties in persons with intellectual disability: a systematic review protocol

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## ABSTRACT

### Introduction

Persons with intellectual disability (ID) are at a higher risk of developing dementia than persons without ID, with an expected earlier onset. Assessment methods for the general population cannot be applied for persons with ID due to their pre-existing intellectual and functional impairments. As there is no agreed-upon measure to assess dementia in persons with ID, multiple instruments for this purpose have been developed and adapted in the last decades. This review aims to identify all available informant-based instruments for the assessment of dementia in persons with ID and to evaluate and compare them according to their measurement properties. Additionally, an overview of the amount and quality of research on these instruments will be provided.

### Methods and analysis

This review will be conducted and reported according to the PRISMA statement. We will adhere to the *COnsensus-based Standards for the selection of health Measurement INstruments* (COSMIN) guidelines, and use a set of characteristics developed for assessment instruments for persons with ID, the *Characteristics of Assessment Instruments for Psychiatric Disorders in Persons with Intellectual Developmental Disorders* (CAPs-IDD). Two comprehensive, systematic literature searches will be applied in ten international databases, including ASSIA, CINAHL, Cochrane Library, ERIC, MEDLINE, PsycINFO, Scopus, Web of Science, OpenGrey, and ProQuest Dissertations & Theses Global. Risk of bias and quality assessment will be done according to COSMIN guidelines. We will apply the modified *Grading of Recommendations, Assessment, Development and Evaluation* (GRADE) approach to rate the overall quality of the available evidence.

## Ethics and dissemination

No ethics statement is needed for this study. The results will be submitted to a peer-reviewed journal, and presented at international conferences.

## Registration details

This review has been submitted for registration to PROSPERO on May 15, 2020.

## Keywords

mental health, dementia, old age psychiatry, statistics and research methods

## ARTICLE SUMMARY

### Strengths and limitations of this study

- This review follows the most up-to-date standards for conducting systematic reviews on assessment instruments, the PRISMA and COSMIN guidelines, and additionally uses the CAPs-IDD, a system especially developed for evaluating assessment instruments for psychiatric disorders in persons with ID.
- Two very comprehensive consecutive search strategies will be applied in a total of ten international databases, including grey and unpublished literature.
- We use language restrictions only for abstracts of studies, not for full texts, trying to minimise language bias.
- We only include informant-based instruments assessing dementia in our evaluation, and exclude direct cognitive tests.
- Due to expected heterogeneity in studies, a quantitative pooling of psychometric data will probably not be possible.

## INTRODUCTION

Intellectual disability (ID) is characterized by limitations in intellectual functioning (IQ < 70) and in adaptive behaviour originating in the developmental phase of an individual.[1] It is also known as Intellectual Developmental Disorder in the *Diagnostic and Statistical Manual of Mental Disorders 5* (DSM-5)[2] and Disorders of Intellectual Development in the *11th Revision of the International Classification of Diseases* (ICD-11).[3] Prevalence of ID is hard to establish, since in many countries no official records of persons with ID exist.[4] In large meta-analysis and reviews, the worldwide prevalence of ID is estimated to range from 1% to 3,3%.[5–7]

Persons with ID are at the same or higher risk to develop dementia than persons without ID.[8–10] Yet, it is often hard to recognize dementia in persons with ID, especially at an early stage. Well-evaluated assessment and screening instruments for the general population, such as the frequently used Mini-Mental State Examination (MMSE)[11] are not suitable for persons with ID due to their pre-existing disabilities.[12,13] Diagnostic overshadowing [16,17] makes it difficult to distinguish symptoms linked to the pre-existing disability from symptoms caused by the onset of dementia. Additionally, the presentation of dementia in persons with ID can differ from the presentation in persons without ID, with behavioural symptoms and personality changes being more frequent and probably earlier in the course of the illness, especially in persons with Down Syndrome.[18,19] To reliably detect dementia in persons with ID, it is recommended to compare a baseline assessment with periodic re-assessments.[14–16] Most dementia assessment methods for persons with ID rely on informant-based measures. The respondent of these instruments should be a person who knows the respective person with ID very well, for instance, a family member or care staff. In contrast to direct tests of cognitive functioning, informant-based instruments can be applied for all persons with ID, irrespective of their intellectual and functional capacity.

Early recognition of dementia is particularly important to start early interventions, to plan for the future, and to get adequate support for family-carers or care staff.[13–15] Not being able to recognize early signs of dementia constitutes a disadvantage for persons with ID, and contradicts the *Convention on the Rights of Persons with Disabilities by the United Nations* (UN-CRPD).[17] Article 25 and 26 of the UN-CRPD require States Parties to ensure that persons with disabilities can get the "highest attainable standard of health without discrimination on the basis of disability."[17]

There are several tools and screening instruments in use for the early recognition of dementia in persons with ID.[12,20] One systematic review found 114 instruments and four test-batteries that have been used to assess dementia in persons with ID. However, some of these instruments have never been designed or adapted to be used in persons with ID, or even to assess dementia.[13] Although there are already some reviews summarizing tools and screening instruments in use for assessing dementia in persons with ID,[13,18–20] no systematic review on measurement properties using up-to-date guidelines for review conduction and psychometric evaluation has been conducted so far. Our review adds to the existing body of knowledge by using a very inclusive systematic search of the literature and, most importantly, by providing a systematic evaluation of informant-based dementia assessment instruments following up-to-date guidelines.

The systematic evaluation will build on the *COnsensus-based Standards for the selection of health Measurement INstruments* (COSMIN) guidelines [21] and the *Characteristics of Assessment Instruments for Psychiatric Disorders in Persons with Intellectual Developmental Disorders* (CAPs-IDD).[22] The CAPs-IDD is a system especially developed for the structured collection of information and evidence-based evaluation of assessment instruments for persons with ID. We will not only evaluate the instruments, but also the existing body of research. For each instrument, we will systematically summarize the amount and quality of

available evaluation studies, depicting which measurement properties have been evaluated to what extent, and which measurement properties have not or insufficiently been evaluated.

The objectives of this systematic review are to (1) identify informant-based instruments suitable for the assessment of dementia in persons ID, to (2) systematically collect and evaluate information on evaluation data of these instruments, to (3) evaluate and compare the instruments found according to their evaluation data, and to (4) provide a systematic overview of the amount and quality of available research for each instrument and each measurement property.

## METHODS AND ANALYSIS

This review will be conducted and reported according to the *Preferred Reporting Items for Systematic Reviews and Meta-Analyses* (PRISMA) statement.[23] The review protocol has been developed using the PRISMA guidelines for protocols (PRISMA-P).[24,25] We will adhere to the COSMIN guidelines,[21] and complement them with a set of characteristics especially developed for assessment instruments for persons with ID, the CAPs-IDD.[22] The systematic review has been submitted for registration with the *International Prospective Register of Systematic Reviews* (PROSPERO) on May 15, 2020 (identification number:181773). If amendments to the protocol are needed, we will register these in PROSPERO, including date and rationale. In the final publication of our results, any amendments to the protocol will be depicted and explained.

### Search strategy

Two systematic searches will be applied consecutively, and carried out between May 2020 and August 2020. The first search should provide an inventory of available informant-based assessment instruments for dementia in persons with ID. The goal of the second search is to locate evaluation studies for each instrument found in the first search. Figure 1 and Figure 2 depict our search strategies using PRISMA flow charts.

First search

To identify instruments we will search in ten international electronic databases, including ASSIA, CINAHL, Cochrane Library, ERIC, MEDLINE, PsycINFO, Scopus, Web of Science, OpenGrey, and ProQuest Dissertations & Theses Global. The search string is depicted in Table 1 and will include various terms for the (1) output of interest, (2) measure of interest, and (3) the specified population. As persons with Down Syndrome are very prone to develop dementia, this subgroup of persons with ID is included in our search strategy. We will use a limit on the timespan of publication in the first search, not including publications before the year 2012. Instruments published up to the year of 2012 are summarized in a previous systematic review.[13] This review used a very inclusive search strategy and listed all assessment instruments that have been used to assess dementia in persons with ID. We will examine the total of 114 dementia assessment instruments listed in the review of 2013, and include those instruments that are in line with our inclusion criteria.

Table 1: Search strategy for the first search

| | 1: Output | 2: Measure | 3: Population |
|---|---|---|---|
| Search terms | Assessment instruments | Dementia | Intellectual disability |
| Synonyms | assessment; diagnostic; diagnosis; screening; instrument; tool; measurement; questionnaire; psychometrics; scale; interview | dementia; Alzheimer's disease | intellectual disability; learning disability; intellectual developmental disorder; trisomy 21, Down syndrome |
| Combined and truncated | assess* OR diagnosti* OR screen OR screening* OR | dement* OR Alzheimer* | ((intellectual* OR learning) AND disab*) OR (intellectual* AND |

| | | |
|---|---|---|
| | instrument* OR tool* OR measure* OR questionnaire* OR psychometr* OR scale* OR interview* | developmental* AND disorder*) OR trisom* 21 OR (down* AND syndrom*) |
| Example search string for SCOPUS | TITLE-ABS-KEY ( ( assess* OR diagnosti* OR screen OR screening* OR instrument* OR tool* OR measure* OR questionnaire* OR psychometr* OR scale* OR interview* ) AND ( dement* OR alzheimer* ) AND ( ( ( intellectual* OR learning ) AND disab* ) OR ( intellectual* AND developmental* AND disorder* ) OR trisom* 21 OR ( down* AND syndrom* ) ) ) AND PUBYEAR > 2011 | |

Inclusion criteria for the first search will be: (1) studies need to focus on assessing dementia in persons with ID, (2) include at least one informant-based instrument (development or evaluation) for the assessment of dementia, (3) this instrument has to be especially developed or adapted for persons with ID, (4) and studies need to have an English language abstract. Exclusion criteria: (1) classification systems like ICD-11, DSM-5, (2) scales including dementia, but focusing on a broader spectrum of disorders for screening purposes or differential diagnosis.

Second search

Once we have identified the instruments, we will conduct a search by citation strategy using the initial publications of each instrument as a reference point. This search strategy was chosen on the assumption that a paper evaluating an instrument would surely cite the initial publication of the respective instrument. The papers used as reference points will also be included in the further appraisal of the literature. For published papers, we will use five international databases allowing a search by citation strategy, including ERIC, PsycInfo, MEDLINE, Scopus, and Web of Science. For published manuals, not listed in at least one of the five databases, we will use Google Scholar. Additionally, all records fulfilling the

inclusion and exclusion criteria of the first search will be transferred and examined in the second search.

The following inclusion criteria will be used in the second search: (1) studies need to describe an evaluation of the respective instrument in persons with ID, and (2) have an English-language abstract. Exclusion criteria: (1) the respective instrument was used primarily for other investigations, not related to an evaluation of the instrument, (2) or the study is a review on assessment instruments, not providing novel information.

To further include grey and unpublished literature in both searches, we will apply an invisible college approach, contacting authors in the field for information or manuscripts on this topic, and we will follow up on meeting abstracts. Full texts of reviews on assessment instruments identified in the course of the two searches will be screened for possible further studies to include. References of papers meeting the inclusion criteria will be hand-searched. We will re-run the search before the final analyses to include the most recent publications.

For study selection, one reviewer will exclude duplicates. All remaining records will be screened and reviewed for eligibility by two team members independently, i.e. blinded to each other's decisions. In the case of disagreement, dissonances will be discussed until agreement is reached. In the case of non-agreement, a third team member will be included in discussion.

**Data extraction**

The first search will result in a list of instruments. Data extracted will be the names of the instruments and information on their initial publication(s). In the second search, we will extract evaluation data of instruments, i.e. measurement properties and characteristics as listed in the COSMIN checklist and the CAPs-IDD. For each characteristic/property extracted, we will record the study design and sample characteristics, including sample size, gender distribution, age distribution, and aetiology of ID. We will include all studies, irrespective of their design, but apply the COSMIN quality rating.

The extraction of all relevant data will be done via standardised and piloted excel spreadsheets by two team members independently. In the case of disagreement, dissonances will be discussed until agreement is reached. In the case of non-agreement, a third team member will be included in discussion. If we find important data missing in a study, we will contact study authors for this information.

**Risk of bias and quality assessment**

Quality and risk of bias will be assessed on study level, on single outcome level, and on an aggregated outcome level, i.e. for each study, each measurement property, and each assessment instrument. We will combine the COSMIN checklists [26–28] with the CAPs-IDD [22], a comprehensive tool especially developed for the evaluation of assessment instruments for psychiatric disorders in persons with ID.

All ratings will be done by two reviewers independently. In the case of disagreement, dissonances will be discussed until agreement is reached. In the case of non-agreement, a third team member will be included in the discussion. Initial interrater agreement will be determined using Cohen's Kappa, calculated in R.[29] The quality rating of the studies will go into the final appraisal of the quality of available evaluation data for each instrument.

As to publication bias, we assume that evaluation results not in favour of the respective instruments are likely to be underreported. This may be partly due to evaluations being frequently done and published by the author(s) of the respective instrument. We will address this by including grey literature and by discussing this aspect in the interpretation of our results.

**Strategy for data synthesis**

A narrative synthesis will be conducted. Assessment instruments will be presented in a table along with their measurement properties and quality ratings according to CAPs-IDD and the COSMIN checklists. Data pooling will probably not be possible. This is due to an expected

limited number of studies evaluating the same property (e.g. internal consistency) for an instrument, and an expected heterogeneity in the population studied (e.g. severity of ID, persons with Down syndrome vs. persons with ID of other aetiology). However, if applicable, we will calculate pooled estimates and 95% confidence intervals using R.[29]

**Analysis of subgroups**

We define persons with Down syndrome/trisomy 21 (DS) as a special subgroup, as they are more often affected by Alzheimer's dementia, with a suspected earlier onset.[30] We will group instruments according to their intended use, and studies according to their participants in four clusters: (1) persons with ID, including persons with DS, (2) only persons with DS, (3) only persons with ID, not including DS, (4) aetiology of ID not specified. For the fourth cluster, we will contact study authors to determine aetiology of ID in the respective sample or for the respective instrument. We will then allocate each study or instrument to the first three clusters according to the information provided by the authors. If no information is provided, the respective study or instrument remains in cluster four.

**Confidence in cumulative evidence**

The modified *Grading of Recommendations, Assessment, Development and Evaluation* (GRADE) approach as suggested by the COSMIN guidelines[21] will be applied to grade the quality of the evidence.

**Data management**

We will use ZOTERO for saving records and managing and storing literature. For extracting data and recording decisions on quality ratings we will use standardised and piloted excel spreadsheets.

**Patient and public involvement**

This research was done without patient involvement due to limited resources.

## DISCUSSION

This review will summarize measurement properties of available informant-based assessment instruments for persons with ID and give an overview of the quality of each instrument and the quality of available evaluation studies. For each instrument we will depict which psychometric properties are evaluated to what extent, and which properties need further evaluation in future research. This will be the first systematic review of dementia assessment instruments for persons with ID using the PRISMA and COSMIN guidelines as well as applying ID-specific criteria of the CAPs-IDD.

Our work will highlight gaps in research on these instruments, thus setting the ground for more effective research in the future. The results of this review will inform researchers and clinicians of the quality of available instruments to assess dementia in persons with ID, and guide them in choosing an adequate instrument. This will hopefully contribute to an improvement of dementia assessment in persons with ID and a better, earlier, and more adequate provision of healthcare services, as demanded by the UN-CRPD.[17]

### Ethics and dissemination

No ethics statement is needed for this study. The results of this systematic review will be submitted for publication to a leading peer-reviewed journal, and presented at international conferences and congresses in the fields of ID, ageing, and dementia.

### Author contributions

ELZ conceived the study, drafted the protocol, and is the guarantor of the review. SK, IZ and FF contributed to study design and drafting the protocol. ELZ, SK, and KW designed and tested the search strategy. FF and IZ tested quality rating tools and software options. All authors read and approved the final protocol.

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**Patient and public involvement**

This research was done without patient involvement.

**Competing interests**

The authors have no competing nor potential conflict interests to declare.

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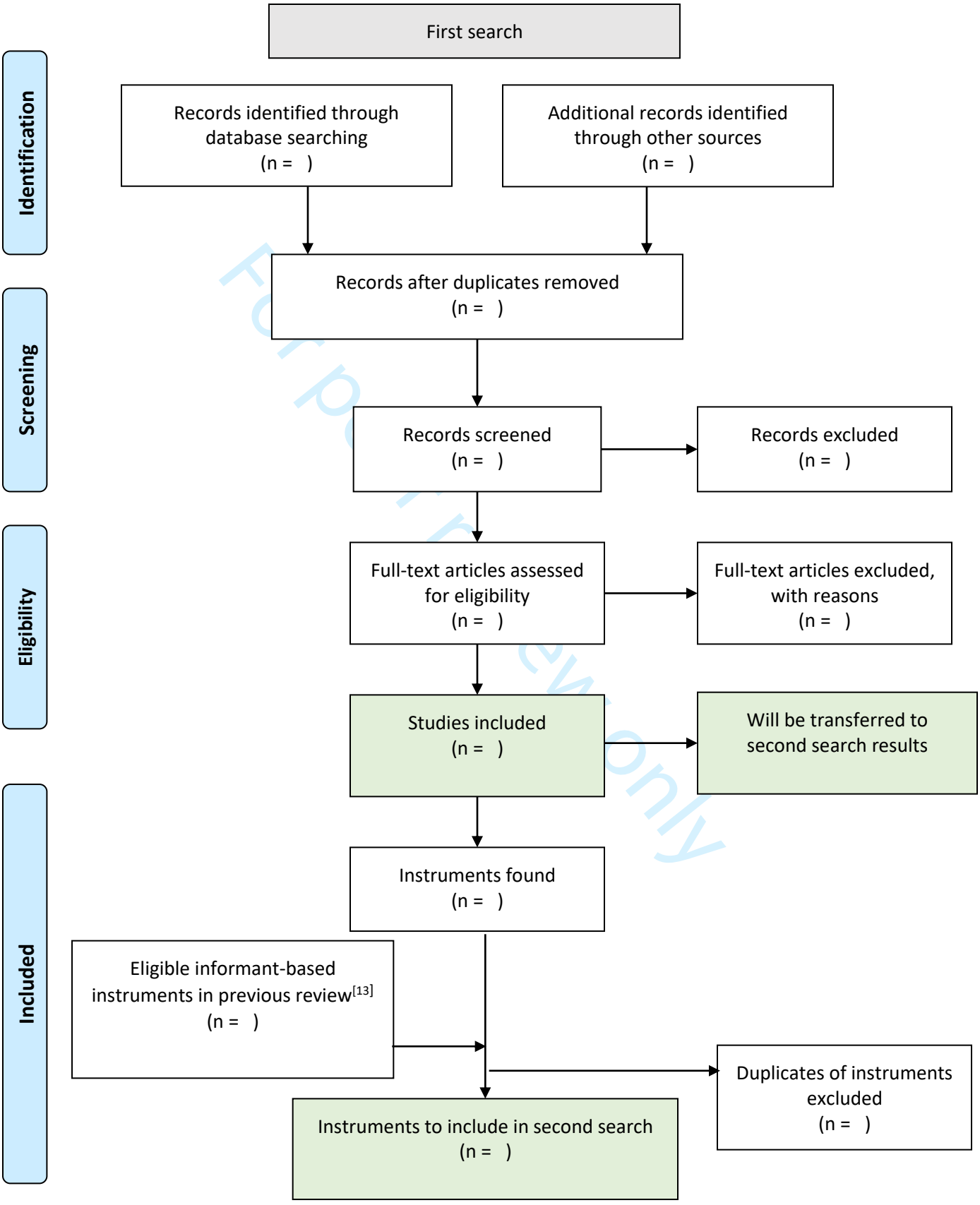
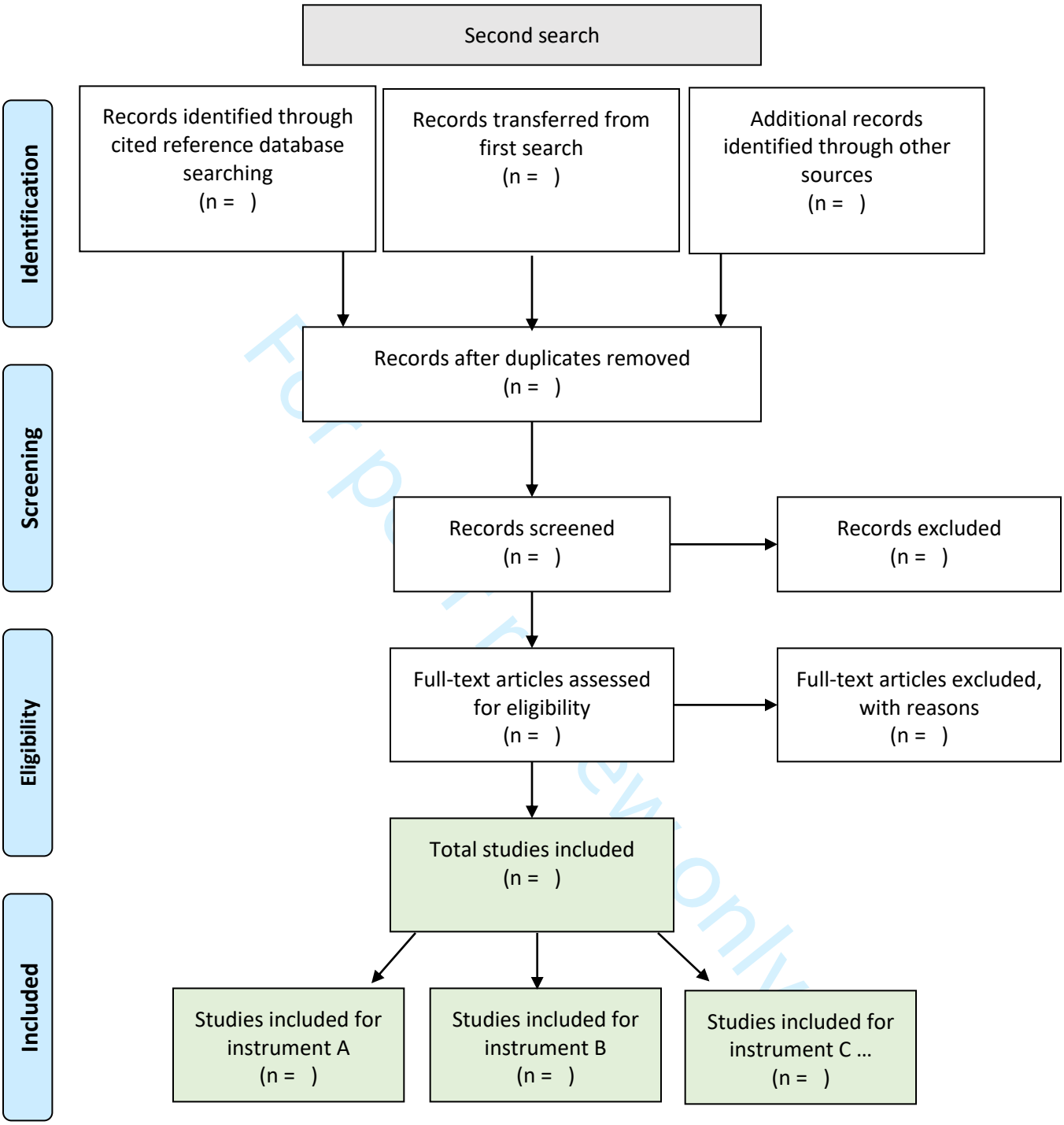
**Figure legends**

Figure 1: PRISMA flow chart of first search

Figure 2: PRISMA flow chart of second search





# Reporting checklist for protocol of a systematic review.

Based on the PRISMA-P guidelines.

## Instructions to authors

Complete this checklist by entering the page numbers from your manuscript where readers will find each of the items listed below.

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In your methods section, say that you used the PRISMA-Preporting guidelines, and cite them as:

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| | | Reporting Item | Page Number | Line Number |
|---|---|---|---|---|
| **Title** | | | | |
| Identification | #1a | Identify the report as a protocol of a systematic review | 1 | 2-3 |

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|---|---|---|---|---|
| Update | #1b | If the protocol is for an update of a previous systematic review, identify as such | n.a. | |
| **Registration** | | | | |
| | #2 | If registered, provide the name of the registry (such as PROSPERO) and registration number | 6 | 137-138 |
| **Authors** | | | | |
| Contact | #3a | Provide name, institutional affiliation, e-mail address of all protocol authors; provide physical mailing address of corresponding author | 1 | 5-20 |
| Contribution | #3b | Describe contributions of protocol authors and identify the guarantor of the review | 12 | 271-275 |
| **Amendments** | | | | |
| | #4 | If the protocol represents an amendment of a previously completed or published protocol, identify as such and list changes; otherwise, state plan for documenting important protocol amendments | 6 | 138-140 |
| **Support** | | | | |
| Sources | #5a | Indicate sources of financial or other support for the review | 13 | 276-278 |
| Sponsor | #5b | Provide name for the review funder and / or sponsor | n.a. | |

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|---|---|---|---|---|
| Role of sponsor or funder | #5c | Describe roles of funder(s), sponsor(s), and / or institution(s), if any, in developing the protocol | n.a. | |
| **Introduction** | | | | |
| Rationale | #6 | Describe the rationale for the review in the context of what is already known | 5 | 104-114 |
| Objectives | #7 | Provide an explicit statement of the question(s) the review will address with reference to participants, interventions, comparators, and outcomes (PICO) | 6 | 124-129 |
| **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 | 7-8, 9 | 153-168, 180-184 |
| 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 | 7, 8, 9 | 148-150, 174-177, 185-190 |
| 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 | 8 | 160-161 |

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| Study records - data management | #11a | Describe the mechanism(s) that will be used to manage records and data throughout the review | 11 | 246-249 |
| Study records - selection process | #11b | State the process that will be used for selecting studies (such as two independent reviewers) through each phase of the review (that is, screening, eligibility and inclusion in meta-analysis) | 9 | 191-194 |
| Study records - 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 | 10 | 203-207 |
| Data items | #12 | List and define all variables for which data will be sought (such as PICO items, funding sources), any pre-planned data assumptions and simplifications | 9 | 195-202 |
| Outcomes and prioritization | #13 | List and define all outcomes for which data will be sought, including prioritization of main and additional outcomes, with rationale | n.a. | |
| Risk of bias in 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 | 10 | 208-223 |

| | | | | |
|---|---|---|---|---|
| Data synthesis | #15a | Describe criteria under which study data will be quantitatively synthesised | 10-11 | 227-231 |
| Data synthesis | #15b | If data are appropriate for quantitative synthesis, describe planned summary measures, methods of handling data and methods of combining data from studies, including any planned exploration of consistency (such as I2, Kendall's τ) | n.a. | |
| Data synthesis | #15c | Describe any proposed additional analyses (such as sensitivity or subgroup analyses, meta-regression) | n.a. | |
| Data synthesis | #15d | If quantitative synthesis is not appropriate, describe the type of summary planned | 10 | 224-227 |
| Meta-bias(es) | #16 | Specify any planned assessment of meta-bias(es) (such as publication bias across studies, selective reporting within studies) | 10 | 219-223 |
| Confidence in cumulative evidence | #17 | Describe how the strength of the body of evidence will be assessed (such as GRADE) | 11 | 242-245 |

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# Informant-based assessment instruments for dementia and their measurement properties in persons with intellectual disability: a systematic review protocol



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# Title: Informant-based assessment instruments for dementia and their measurement properties in persons with intellectual disability: a systematic review protocol

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## ABSTRACT

### Introduction

Persons with intellectual disability (ID) are at a higher risk of developing dementia than persons without ID, with an expected earlier onset. Assessment methods for the general population cannot be applied for persons with ID due to their pre-existing intellectual and functional impairments. As there is no agreed-upon measure to assess dementia in persons with ID, multiple instruments for this purpose have been developed and adapted in the last decades. This review aims to identify all available informant-based instruments for the assessment of dementia in persons with ID and to evaluate and compare them according to their measurement properties. Additionally, an overview of the amount and quality of research on these instruments will be provided.

### Methods and analysis

This review will be conducted and reported according to the PRISMA statement. We will adhere to the *COnsensus-based Standards for the selection of health Measurement INstruments* (COSMIN) guidelines, and use a set of characteristics developed for assessment instruments for persons with ID, the *Characteristics of Assessment Instruments for Psychiatric Disorders in Persons with Intellectual Developmental Disorders* (CAPs-IDD). Two comprehensive, systematic literature searches will be applied in ten international databases, including ASSIA, CINAHL, Cochrane Library, ERIC, MEDLINE, PsycINFO, Scopus, Web of Science, OpenGrey, and ProQuest Dissertations & Theses Global. Risk of bias and quality assessment will be done according to COSMIN guidelines. We will apply the modified *Grading of Recommendations, Assessment, Development and Evaluation* (GRADE) approach to rate the overall quality of the available evidence.

## Ethics and dissemination

No ethics statement is needed for this study. The results will be submitted to a peer-reviewed journal, and presented at international conferences.

## Registration details

PROSPERO registration number: CRD42020181773

## Keywords

mental health, dementia, old age psychiatry, statistics and research methods

## ARTICLE SUMMARY

### Strengths and limitations of this study

- This review follows the most up-to-date standards for conducting systematic reviews on assessment instruments, the PRISMA and COSMIN guidelines, and additionally uses the CAPs-IDD, a system especially developed for evaluating assessment instruments for psychiatric disorders in persons with ID.
- Two very comprehensive consecutive search strategies will be applied in a total of ten international databases, including grey and unpublished literature.
- We use no language restrictions to minimise language bias.
- We only include informant-based instruments assessing dementia in our evaluation, and exclude direct cognitive tests.
- Due to expected heterogeneity in studies, a quantitative pooling of psychometric data will probably not be possible.

## INTRODUCTION

Intellectual disability (ID) is characterized by limitations in intellectual functioning (IQ < 70) and in adaptive behaviour originating in the developmental phase of an individual.[1] It is also known as Intellectual Developmental Disorder in the *Diagnostic and Statistical Manual of Mental Disorders 5* (DSM-5)[2] and Disorders of Intellectual Development in the *11th Revision of the International Classification of Diseases* (ICD-11).[3] Prevalence of ID is hard to establish, since in many countries no official records of persons with ID exist.[4] In large meta-analysis and reviews, the worldwide prevalence of ID is estimated to range from 1% to 3,3%.[5–7]

Persons with ID are at the same or higher risk to develop dementia than persons without ID.[8–10] Yet, due to their limitations in intellectual functioning, it is often hard to recognize dementia in this population, especially at an early stage. Well-evaluated assessment and screening instruments for the general population, such as the frequently used Mini-Mental State Examination (MMSE)[11] are not suitable for persons with ID due to their pre-existing disabilities.[12,13] Diagnostic overshadowing[14,15] makes it difficult to distinguish symptoms linked to the pre-existing disability from symptoms caused by the onset of dementia. Additionally, the presentation of dementia in persons with ID can differ from the presentation in persons without ID, with behavioural symptoms and personality changes being more frequent and probably earlier in the course of the illness, especially in persons with Down Syndrome.[16,17] To reliably detect dementia in persons with ID, it is recommended to compare a baseline assessment with periodic re-assessments.[18–20] Most dementia assessment methods for persons with ID rely on informant-based measures. The respondent of these instruments should be a person who knows the respective person with ID very well, for instance, a family member or care staff. In contrast to direct tests of cognitive functioning,

informant-based instruments can be applied for all persons with ID, irrespective of their intellectual and functional capacity.

Early recognition of dementia is particularly important to start early interventions, to plan for the future, and to get adequate support for family-carers or care staff.[21–23] Not being able to recognize early signs of dementia constitutes a disadvantage for persons with ID, and contradicts the *Convention on the Rights of Persons with Disabilities by the United Nations* (UN-CRPD).[24] Article 25 and 26 of the UN-CRPD require States Parties to ensure that persons with disabilities can get the "highest attainable standard of health without discrimination on the basis of disability."[24]

There are several tools and screening instruments in use for the early recognition of dementia in persons with ID.[13,25] These instruments can be distinguished in three categories: medical test (e.g. fMRI, gene-markers), direct cognitive tests, and informant-based scales, which are also called observer-rated scales. In this review we focus solely on informant-based scales, which include observer-reported outcome measures (ObsROM), as well as clinician-reported outcome measures (ClinROM). [26]

One systematic review found 114 instruments and four test-batteries that have been used to assess dementia in persons with ID. However, some of these instruments have never been designed or adapted to be used in persons with ID, or even to assess dementia.[13] Although there are already some reviews summarising tools and screening instruments in use for assessing dementia in persons with ID,[13,25,27,28] no systematic review on measurement properties using up-to-date guidelines for review conduction and psychometric evaluation has been conducted so far. We want to provide an inventory of available informant-based instruments and their measurement properties. This should help clinicians and researches in choosing the adequate instrument for their respective purpose. Our review adds to the existing body of knowledge by using a very inclusive systematic search of the literature and, most

importantly, by providing a systematic evaluation of informant-based dementia assessment instruments following up-to-date guidelines.

For each instrument, we will systematically summarise the amount and quality of available evaluation studies, depicting which measurement properties have been evaluated to what extent, and which measurement properties have not or insufficiently been evaluated.

The objectives of this systematic review are to (1) identify informant-based instruments suitable for the assessment of dementia in persons with ID, to (2) provide a systematic overview of descriptive aspects for each instrument (e.g. respondent requirements, response format), to (3) provide a systematic overview of the amount and quality of available research for each instrument and each measurement property, and to (4) provide a recommendation for the most suitable instrument(s) based on all information collected.

## METHODS AND ANALYSIS

This review will be conducted and reported according to the *Preferred Reporting Items for Systematic Reviews and Meta-Analyses* (PRISMA) statement,[29] and the review protocol has been developed using the PRISMA guidelines for protocols (PRISMA-P).[30,31] We will adhere to the *COnsensus-based Standards for the selection of health Measurement INstruments* (COSMIN) guidelines,[32] and complement them with a set of characteristics especially developed for assessment instruments for persons with ID, the *Characteristics of Assessment Instruments for Psychiatric Disorders in Persons with Intellectual Developmental Disorders* (CAPs-IDD).[33] The systematic review has been registered with the *International Prospective Register of Systematic Reviews* (PROSPERO) with registration number CRD42020181773. If amendments to the protocol are needed, we will register these in PROSPERO, including date and rationale. In the final publication of our results, any amendments to the protocol will be depicted and explained.

## Search strategy

Two systematic searches will be applied consecutively, and carried out between May 2020 and August 2020. The first search should provide an inventory of available informant-based assessment instruments for dementia in persons with ID. The goal of the second search is to locate evaluation studies for each instrument found in the first search. Figure 1 and Figure 2 depict our search strategies using PRISMA flow charts.

### First search

To identify instruments we will search in ten international electronic databases, including ASSIA, CINAHL, Cochrane Library, ERIC, MEDLINE, PsycINFO, Scopus, Web of Science, OpenGrey, and ProQuest Dissertations & Theses Global. The search string is depicted in Table 1 and will include various terms for the (1) output of interest, (2) construct of interest, and (3) the specified population. As persons with Down Syndrome are very prone to develop dementia, this subgroup of persons with ID is included in our search strategy. We will use a limit on the timespan of publication in the first search, not including publications before the year 2012. Instruments published up to the year of 2012 are summarised in a previous systematic review.[13] This review used a very inclusive search strategy and listed all assessment instruments that have been used to assess dementia in persons with ID. We will examine the total of 114 dementia assessment instruments listed in the review of 2013, and include those instruments that are in line with our inclusion criteria.

Table 1: Search strategy for the first search

| | 1: Output | 2: Construct | 3: Population |
|---|---|---|---|
| Search terms | Assessment instruments | Dementia | Intellectual disability |
| Synonyms | assessment; diagnostic; diagnosis; screening; | dementia; Alzheimer's disease | intellectual disability; learning disability; |

| | | | |
|---|---|---|---|
| | instrument; tool; measurement; questionnaire; psychometrics; scale; interview | | intellectual developmental disorder; trisomy 21, Down syndrome |
| Combined and truncated | assess* OR diagnosti* OR screen OR screening* OR instrument* OR tool* OR measure* OR questionnaire* OR psychometr* OR scale* OR interview* | dement* OR Alzheimer* | ((intellectual* OR learning) AND disab*) OR (intellectual* AND developmental* AND disorder*) OR trisom* 21 OR (down* AND syndrom*) |
| Example search string for SCOPUS | TITLE-ABS-KEY ( ( assess* OR diagnosti* OR screen OR screening* OR instrument* OR tool* OR measure* OR questionnaire* OR psychometr* OR scale* OR interview* ) AND ( dement* OR alzheimer* ) AND ( ( ( intellectual* OR learning ) AND disab* ) OR ( intellectual* AND developmental* AND disorder* ) OR trisom* 21 OR ( down* AND syndrom* ) ) ) AND PUBYEAR > 2011 | | |

Inclusion criteria for the first search will be: (1) studies need to focus on assessing dementia in persons with ID, (2) describe the development or evaluation of an informant-based instrument for the assessment of dementia, (3) and this instrument has to be especially developed or adapted for persons with ID. Exclusion criteria: (1) classification systems like ICD-11, DSM-5, (2) scales including dementia, but focusing on a broader spectrum of disorders for screening purposes or differential diagnosis, such as the *Psychiatric Assessment Schedule for Adult with Developmental Disability* (PAS-ADD).[34]

Second search

Once we have identified the instruments, we will conduct a search by citation strategy using the initial publications of each instrument as a reference point. This search strategy was

chosen on the assumption that a paper evaluating an instrument would surely cite the initial publication of the respective instrument. The papers used as reference points will also be included in the further appraisal of the literature. For published papers, we will use five international databases allowing a search by citation strategy, including ERIC, PsycInfo, MEDLINE, Scopus, and Web of Science. For published manuals, not listed in at least one of the five databases, we will use Google Scholar. Additionally, all records fulfilling the inclusion and exclusion criteria of the first search will be transferred and examined in the second search.

The following inclusion criterion will be used in the second search: (1) studies need to describe an evaluation of the respective instrument in persons with ID. Exclusion criteria comprise: (1) the respective instrument was used primarily for other investigations, not related to an evaluation of the instrument, (2) or the study is a review on assessment instruments, not providing novel information.

To further include grey and unpublished literature in both searches, we will apply an invisible college approach, contacting authors in the field for information or manuscripts on this topic, and we will follow up on meeting abstracts. Full texts of reviews on assessment instruments identified in the course of the two searches will be screened for possible further studies to include. References of papers meeting the inclusion criteria will be hand-searched. We will re-run both searches before the final analyses to include the most recent publications.

For study selection, one reviewer will exclude duplicates. All remaining records will be screened and reviewed for eligibility by two team members independently, i.e. blinded to each other's decisions. In the case of disagreement, dissonances will be discussed until agreement is reached. In the case of non-agreement, a third team member will be included in discussion.

**Data extraction**

The first search will result in a list of instruments. Data extracted will be the names of the

instruments and information on their initial publication(s). In the second search, we will extract evaluation data of instruments, i.e. measurement properties and characteristics as listed in the COSMIN checklists and the CAPs-IDD. For each characteristic/property extracted, we will record the study design and sample characteristics, including sample size, gender distribution, age distribution, aetiology of ID, and country (language) in which the instrument was evaluated. We will include all studies, irrespective of their design.

The extraction of all relevant data will be done via standardised and piloted excel spreadsheets by two team members independently. In the case of disagreement, dissonances will be discussed until agreement is reached. In the case of non-agreement, a third team member will be included in discussion. If data necessary for coding is missing in a study, we will contact the respective study authors for this information.

**Risk of bias and quality assessment**

Quality and risk of bias will be assessed on study level (for each measurement property), on outcome level (for each assessment instrument), and on an aggregated outcome level, applying the *Grading of Recommendations, Assessment, Development and Evaluation* (GRADE) approach. We will combine the COSMIN checklists [35–37] with the CAPs-IDD [33], a comprehensive tool specifically developed for the evaluation of assessment instruments for psychiatric disorders in persons with ID. The CAPs-IDD consists of two parts: (1) conceptual and measurement model (including descriptive aspects of instruments, e.g. respondent requirements, theoretical foundation), and (2) psychometric properties. We will only use the first part, as the second part is more comprehensively covered by the COSMIN checklists.

All ratings will be done by two reviewers independently. In the case of disagreement, dissonances will be discussed until agreement is reached. In the case of non-agreement, a

third team member will be included in the discussion. Initial interrater agreement will be determined using percentage agreement, calculated in R.[38]

As to publication bias, we assume that evaluation results not in favour of the respective instruments are likely to be underreported. This may be partly due to evaluations being frequently done and published by the developer(s) of the respective instrument. We will address this by including grey literature and by discussing this aspect in the interpretation of our results.

**Strategy for data synthesis**

A narrative synthesis will be conducted. Assessment instruments will be presented in a table along with descriptive aspects according to CAPs-IDD, and their measurement properties and quality ratings according to the COSMIN checklists. Quantitative data pooling will probably not be possible. This is due to an expected limited number of studies evaluating the same property (e.g. internal consistency) for an instrument, and an expected heterogeneity in the population studied (e.g. severity of ID, persons with Down Syndrome vs. persons with ID of other aetiology). However, if applicable, we will calculate pooled estimates and 95% confidence intervals using R.[38]

**Analysis of subgroups**

We define persons with Down Syndrome/trisomy 21 (DS) as a special subgroup, as they are more often affected by Alzheimer's dementia, with a suspected earlier onset.[16] We will group instruments according to their intended use, and studies according to their participants in four clusters: (1) persons with ID, including persons with DS, (2) only persons with DS, (3) only persons with ID, not including DS, (4) aetiology of ID not specified. For the fourth cluster, we will contact study authors to determine aetiology of ID in the respective sample or for the respective instrument. We will then allocate each study or instrument to the first three

clusters according to the information provided by the authors. If no information is provided, the respective study or instrument remains in cluster four.

**Confidence in cumulative evidence**

The modified GRADE approach as suggested by the COSMIN guidelines[32] will be applied to grade the quality of the evidence.

**Data management**

We will use ZOTERO for saving records and managing and storing literature, including managing duplicates. For extracting data and recording decisions on quality ratings we will use standardised and piloted excel spreadsheets.

**Patient and public involvement**

This research was done without patient involvement due to limited resources.

## DISCUSSION

This review will summarise measurement properties of available informant-based assessment instruments for persons with ID and give an overview of the quality of each instrument and the quality of available evaluation studies. For each instrument we will depict which psychometric properties are evaluated to what extent, and which properties need further evaluation in future research. This will be the first systematic review of dementia assessment instruments for persons with ID using PRISMA and COSMIN guidelines as well as applying the ID-specific criteria of the CAPs-IDD.

Our work will highlight gaps in research on these instruments, thus setting the ground for more effective research in the future. The results of this review will inform researchers and clinicians of the quality of available instruments to assess dementia in persons with ID, and guide them in choosing an adequate instrument. This will hopefully contribute to an

improvement of dementia assessment in persons with ID and a better, earlier, and more adequate provision of healthcare services, as demanded by the UN-CRPD.[24]

## Ethics and dissemination

No ethics statement is needed for this study. The results of this systematic review will be submitted for publication to a leading peer-reviewed journal, and presented at international conferences and congresses in the fields of ID, ageing, and dementia.

## Author's contributions

ELZ conceived the study, drafted the protocol, and is the guarantor of the review. SK, IZ and FF contributed to study design and drafting the protocol. ELZ, SK, and KW designed and tested the search strategy. FF and IZ tested quality rating tools and software options. All authors read and approved the final protocol.

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The authors have no competing nor potential conflict interests to declare.

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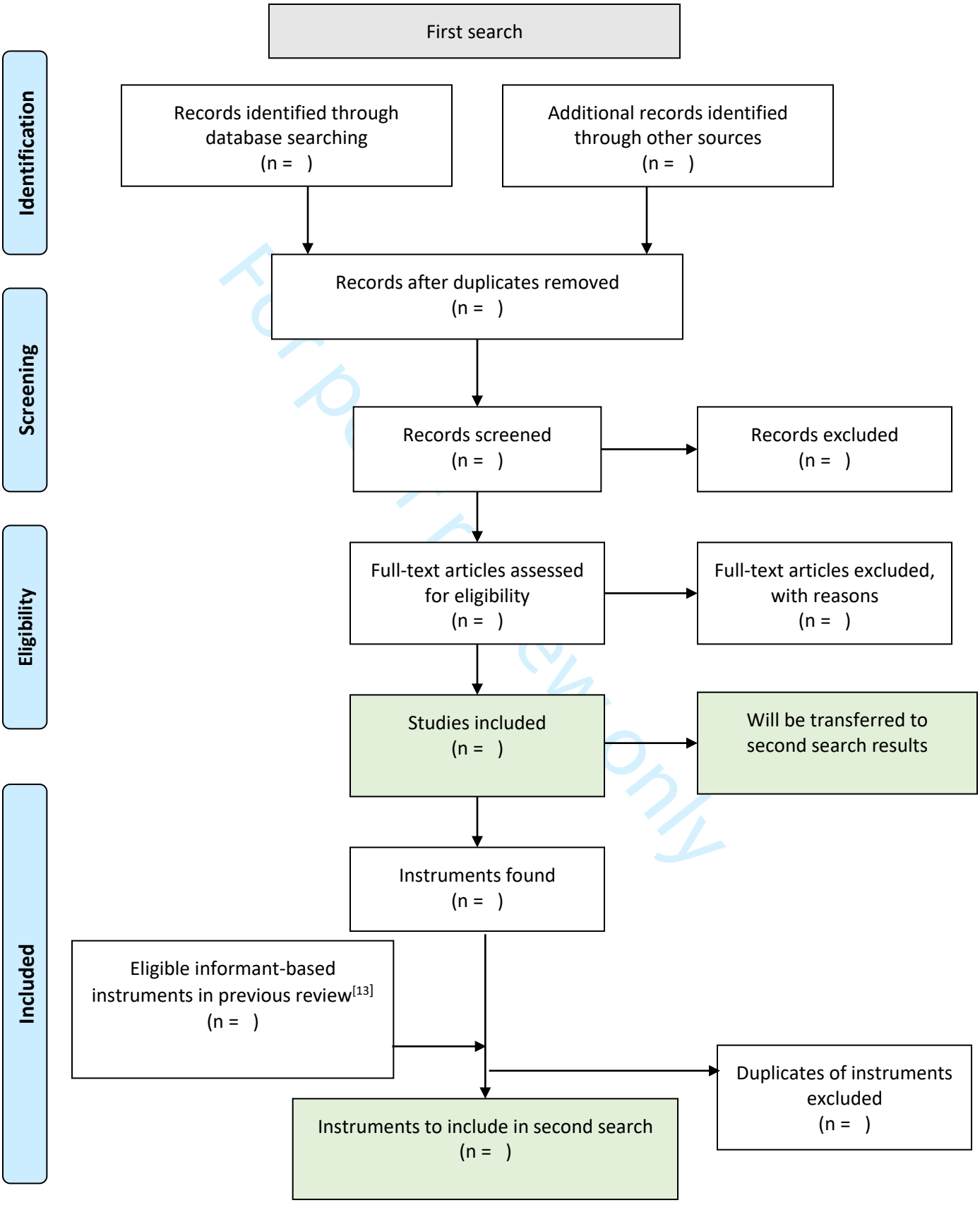
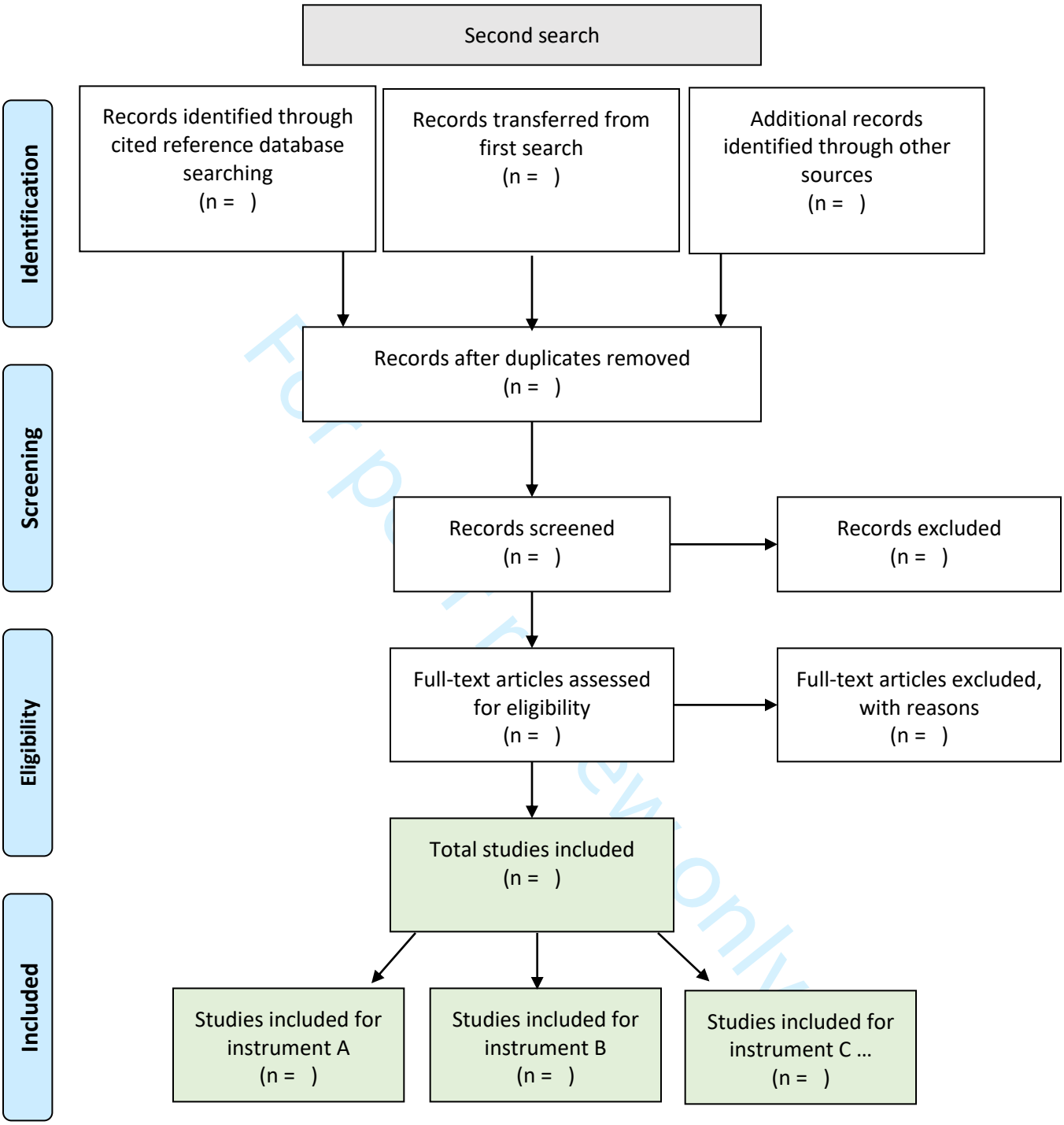
38 R Core Team. *R: A Language and Environment for Statistical Computing*. Vienna, Austria: : R Foundation for Statistical Computing 2020. https://www.R-project.org/

## Figure legends

Figure 1: PRISMA flow chart of first search

Figure 2: PRISMA flow chart of second search





**Supplementary information**

**Informant-based assessment instruments for dementia and their measurement properties in persons with intellectual disability: a systematic review protocol**

Detailed search strategy in electronic databases

**ASSIA**
(via ProQuest)

noft(Assess* OR diagnosti* OR screen OR screening* OR instrument* OR tool* OR measure* OR questionnaire* OR psychometr* OR scale* OR interview*) AND noft(Dement* OR Alzheimer*) AND noft(((intellectual* OR learning) AND disab*) OR (intellectual* AND developmental* AND disorder*) OR trisom* 21 OR (down* AND syndrom*))

Additional limits: Date: From 2012 to 2020

**CINAHL**
(via EBSCOhost) (MEDLINE records excluded, since they are already in the MEDLINE Search):

(AB ( Assess* OR diagnosti* OR screen OR screening* OR instrument* OR tool* OR measure* OR questionnaire* OR psychometr* OR scale* OR interview* ) AND AB ( Dement* OR Alzheimer* ) AND AB ( ((intellectual* OR learning) AND disab*) OR (intellectual* AND developmental* AND disorder*) OR trisom* 21 OR (down* AND syndrom*) ) ) OR (TI ( Assess* OR diagnosti* OR screen OR screening* OR instrument* OR tool* OR measure* OR questionnaire* OR psychometr* OR scale* OR interview* ) AND TI ( Dement* OR Alzheimer* ) AND TI ( ((intellectual* OR learning) AND disab*) OR (intellectual* AND developmental* AND disorder*) OR trisom* 21 OR (down* AND syndrom*) ) ) OR (SU ( Assess* OR diagnosti* OR screen OR screening* OR instrument* OR tool* OR measure* OR questionnaire* OR psychometr* OR scale* OR interview* ) AND SU ( Dement* OR Alzheimer* ) AND SU ( ((intellectual* OR learning) AND disab*) OR (intellectual* AND developmental* AND disorder*) OR trisom* 21 OR (down* AND syndrom*) ) )

**Limiters** - Published Date: 20120101-20201231; Exclude MEDLINE records

**Cochrane Library**

(Assess* OR diagnosti* OR screen OR screening* OR instrument* OR tool* OR measure * OR questionnaire* OR psychometr* OR scale* OR interview*):ti,ab,kw *AND* (Dement* OR Alzheimer*):ti,ab,kw *AND* (((intellectual* OR learning) AND disab*) OR (intellectual* AND developmental* AND d isorder*) OR trisom* 21 OR (down* AND syndrom*)):ti,ab,kw

with Cochrane Library publication date from Jan 2012 to Dec 2020

**ERIC, MEDLINE, and PsycInfo**
(via Ovid) [mp=ab, ti, hw, id, tc, ot, tm, mh]

((Assess* or diagnosti* or screen or screening* or instrument* or tool* or measure* or questionnaire* or psychometr* or scale* or interview*) and (Dement* or Alzheimer*) and (((intellectual* or learning) and disab*) or (intellectual* and developmental* and disorder*) or trisom* 21 or (down* and syndrom*))).mp

limit to yr="2012 - 2020"

**SCOPUS**

TITLE-ABS-KEY ( ( assess* OR diagnosti* OR screen OR screening* OR instrument* OR tool* OR measure* OR questionnaire* OR psychometr* OR scale* OR interview* ) AND ( dement* OR alzheimer* ) AND ( ( ( intellectual* OR learning ) AND disab* ) OR ( intellectual* AND developmental* AND disorder* ) OR trisom* 21 OR ( down* AND syndrom* ) ) ) AND PUBYEAR > 2011

**Web of Science Core Collection**:

**TOPIC:**(Assess* OR diagnosti* OR screen OR screening* OR instrument* OR tool* OR measure* OR questionnaire* OR psychometr* OR scale* OR interview*) *AND*
**TOPIC:**(Dement* OR Alzheimer*) *AND*
**TOPIC:**(((intellectual* OR learning) AND disab*) OR (intellectual* AND developmental* AND disorder*) OR trisom* 21 OR (down* AND syndrom*))

**Timespan:** 2012-2020. **Indexes:** SCI-EXPANDED, SSCI, A&HCI, CPCI-S, CPCI-SSH, BKCI-S, BKCI-SSH, ESCI, CCR-EXPANDED, IC.

**OpenGrey**
(Assess* OR diagnosti* OR screen OR screening* OR instrument* OR tool* OR measure* OR questionnaire* OR psychometr* OR scale* OR interview*) *AND*
(Dement* OR Alzheimer*) *AND*
(((intellectual* OR learning) AND disab*) OR (intellectual* AND developmental* AND disorder*) OR trisom* 21 OR (down* AND syndrom*))

**ProQuest Dissertations & Theses**
noft(Assess* OR diagnosti* OR screen OR screening* OR instrument* OR tool* OR measure* OR questionnaire* OR psychometr* OR scale* OR interview*) AND noft(Dement* OR Alzheimer*) AND noft(((intellectual* OR learning) AND disab*) OR (intellectual* AND developmental* AND disorder*) OR trisom* 21 OR (down* AND syndrom*))

Additional limits: Date: From 2012 to 2020

# Reporting checklist for protocol of a systematic review.

Based on the PRISMA-P guidelines.

## Instructions to authors

Complete this checklist by entering the page numbers from your manuscript where readers will find each of the items listed below.

Your article may not currently address all the items on the checklist. Please modify your text to include the missing information. If you are certain that an item does not apply, please write "n/a" and provide a short explanation.

Upload your completed checklist as an extra file when you submit to a journal.

In your methods section, say that you used the PRISMA-Preporting guidelines, and cite them as:

Moher D, Shamseer L, Clarke M, Ghersi D, Liberati A, Petticrew M, Shekelle P, Stewart LA. Preferred Reporting Items for Systematic Review and Meta-Analysis Protocols (PRISMA-P) 2015 statement. Syst Rev. 2015;4(1):1.



| | | Reporting Item | Page Number | Line Number |
|---|---|---|---|---|
| **Title** | | | | |
| Identification | #1a | Identify the report as a protocol of a systematic review | 1 | 2-3 |

| | | | | |
|---|---|---|---|---|
| Update | #1b | If the protocol is for an update of a previous systematic review, identify as such | n.a. | |
| **Registration** | | | | |
| | #2 | If registered, provide the name of the registry (such as PROSPERO) and registration number | 6 | 141-142 |
| **Authors** | | | | |
| Contact | #3a | Provide name, institutional affiliation, e-mail address of all protocol authors; provide physical mailing address of corresponding author | 1 | 5-20 |
| Contribution | #3b | Describe contributions of protocol authors and identify the guarantor of the review | 13 | 279-283 |
| **Amendments** | | | | |
| | #4 | If the protocol represents an amendment of a previously completed or published protocol, identify as such and list changes; otherwise, state plan for documenting important protocol amendments | 6 | 142-144 |
| **Support** | | | | |
| Sources | #5a | Indicate sources of financial or other support for the review | 13 | 284-286 |

| | | | | |
|---|---|---|---|---|
| Sponsor | #5b | Provide name for the review funder and / or sponsor | n.a. | |
| Role of sponsor or funder | #5c | Describe roles of funder(s), sponsor(s), and / or institution(s), if any, in developing the protocol | n.a. | |
| **Introduction** | | | | |
| Rationale | #6 | Describe the rationale for the review in the context of what is already known | 5-6 | 105-122 |
| Objectives | #7 | Provide an explicit statement of the question(s) the review will address with reference to participants, interventions, comparators, and outcomes (PICO) | 6 | 126-131 |
| **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 | 7-8, 9 | 157-172, 180-188 |
| 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 | 7, 9 | 152-154, 178-181, 189-194 |

| | | | | |
|---|---|---|---|---|
| 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 | 8; supplement | 164-165 |
| Study records - data management | #11a | Describe the mechanism(s) that will be used to manage records and data throughout the review | 12 | 254-257 |
| Study records - selection process | #11b | State the process that will be used for selecting studies (such as two independent reviewers) through each phase of the review (that is, screening, eligibility and inclusion in meta-analysis) | 9 | 195-198 |
| Study records - 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 | 10 | 207-211 |
| Data items | #12 | List and define all variables for which data will be sought (such as PICO items, funding sources), any pre-planned data assumptions and simplifications | 9-10 | 199-206 |
| Outcomes and prioritization | #13 | List and define all outcomes for which data will be sought, including prioritization of main and additional outcomes, with rationale | n.a. | |

| | | | | |
|---|---|---|---|---|
| Risk of bias in 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 | 10-11 | 212-231 |
| Data synthesis | #15a | Describe criteria under which study data will be quantitatively synthesised | 11 | 235-240 |
| Data synthesis | #15b | If data are appropriate for quantitative synthesis, describe planned summary measures, methods of handling data and methods of combining data from studies, including any planned exploration of consistency (such as I2, Kendall's τ) | n.a. | |
| Data synthesis | #15c | Describe any proposed additional analyses (such as sensitivity or subgroup analyses, meta-regression) | n.a. | |
| Data synthesis | #15d | If quantitative synthesis is not appropriate, describe the type of summary planned | 11 | 232-235 |
| Meta-bias(es) | #16 | Specify any planned assessment of meta-bias(es) (such as publication bias across studies, selective reporting within studies) | 11 | 227-231 |

| | | | | |
|---|---|---|---|---|
| Confidence in cumulative evidence | #17 | Describe how the strength of the body of evidence will be assessed (such as GRADE) | 12 | 251-253 |

BMJ Open

# Informant-based assessment instruments for dementia and their measurement properties in persons with intellectual disability: a systematic review protocol



| | |
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# Title: Informant-based assessment instruments for dementia and their measurement properties in persons with intellectual disability: a systematic review protocol

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## ABSTRACT

### Introduction

Persons with intellectual disability (ID) are at a higher risk of developing dementia than persons without ID, with an expected earlier onset. Assessment methods for the general population cannot be applied for persons with ID due to their pre-existing intellectual and functional impairments. As there is no agreed-upon measure to assess dementia in persons with ID, multiple instruments for this purpose have been developed and adapted in the last decades. This review aims to identify all available informant-based instruments for the assessment of dementia in persons with ID, to evaluate and compare them according to their measurement properties, and to provide a recommendation for the most suitable instrument(s). Additionally, an overview of the amount and quality of research on these instruments will be provided.

### Methods and analysis

This review will be conducted and reported according to the PRISMA statement. We will adhere to the *COnsensus-based Standards for the selection of health Measurement INstruments* (COSMIN) guidelines, and use a set of characteristics developed for assessment instruments for persons with ID, the *Characteristics of Assessment Instruments for Psychiatric Disorders in Persons with Intellectual Developmental Disorders* (CAPs-IDD). Two comprehensive, systematic literature searches will be applied in ten international databases, including ASSIA, CINAHL, Cochrane Library, ERIC, MEDLINE, PsycINFO, Scopus, Web of Science, OpenGrey, and ProQuest Dissertations & Theses Global. Risk of bias and quality assessment will be done according to COSMIN guidelines. We will apply the modified *Grading of Recommendations, Assessment, Development and Evaluation* (GRADE) approach to rate the overall quality of the available evidence.

## Ethics and dissemination

No ethics statement is needed for this study. The results will be submitted to a peer-reviewed journal, and presented at international conferences.

## Registration details

PROSPERO registration number: CRD42020181773

## Keywords

mental health, dementia, old age psychiatry, statistics and research methods

## ARTICLE SUMMARY

### Strengths and limitations of this study

- This review follows the most up-to-date standards for conducting systematic reviews on assessment instruments, the PRISMA and COSMIN guidelines, and additionally uses the CAPs-IDD, a system especially developed for evaluating assessment instruments for psychiatric disorders in persons with ID.
- Two very comprehensive consecutive search strategies will be applied in a total of ten international databases, including grey and unpublished literature.
- We use no language restrictions to minimise language bias.
- We only include informant-based instruments assessing dementia in our evaluation, and exclude direct cognitive tests.
- Due to expected heterogeneity in studies, a quantitative pooling of psychometric data will probably not be possible.

## INTRODUCTION

Intellectual disability (ID) is characterized by limitations in intellectual functioning (IQ < 70) and in adaptive behaviour originating in the developmental phase of an individual.[1] It is also known as Intellectual Developmental Disorder in the *Diagnostic and Statistical Manual of Mental Disorders 5* (DSM-5)[2] and Disorders of Intellectual Development in the *11th Revision of the International Classification of Diseases* (ICD-11).[3] Prevalence of ID is hard to establish, since in many countries no official records of persons with ID exist.[4] In large meta-analysis and reviews, the worldwide prevalence of ID is estimated to range from 1% to 3,3%.[5–7]

Persons with ID are at the same or higher risk to develop dementia than persons without ID.[8–10] Yet, due to their limitations in intellectual functioning, it is often hard to recognize dementia in this population, especially at an early stage. Well-evaluated assessment and screening instruments for the general population, such as the frequently used Mini-Mental State Examination (MMSE)[11] are not suitable for persons with ID due to their pre-existing disabilities.[12,13] Diagnostic overshadowing[14,15] makes it difficult to distinguish symptoms linked to the pre-existing disability from symptoms caused by the onset of dementia. Additionally, the presentation of dementia in persons with ID can differ from the presentation in persons without ID, with behavioural symptoms and personality changes being more frequent and probably earlier in the course of the illness, especially in persons with Down Syndrome.[16,17] To reliably detect dementia in persons with ID, it is recommended to compare a baseline assessment with periodic re-assessments.[18–20] Most dementia assessment methods for persons with ID rely on informant-based measures. The respondent of these instruments should be a person who knows the respective person with ID very well, for instance, a family member or care staff. In contrast to direct tests of cognitive functioning,

informant-based instruments can be applied for all persons with ID, irrespective of their intellectual and functional capacity.

Early recognition of dementia is particularly important to start early interventions, to plan for the future, and to get adequate support for family-carers or care staff.[21–23] Not being able to recognize early signs of dementia constitutes a disadvantage for persons with ID, and contradicts the *Convention on the Rights of Persons with Disabilities by the United Nations* (UN-CRPD).[24] Article 25 and 26 of the UN-CRPD require States Parties to ensure that persons with disabilities can get the "highest attainable standard of health without discrimination on the basis of disability."[24]

There are several tools and screening instruments in use for the early recognition of dementia in persons with ID.[13,25] These instruments can be placed into one of three categories: medical test (e.g. fMRI, gene-markers), direct cognitive tests, and informant-based scales, which are also called observer-rated scales. In this review we focus solely on informant-based scales, which include observer-reported outcome measures (ObsROM), as well as clinician-reported outcome measures (ClinROM). [26]

One systematic review found 114 instruments and four test-batteries that have been used to assess dementia in persons with ID. However, some of these instruments have never been designed or adapted to be used in persons with ID, or even to assess dementia.[13] Although there are already some reviews summarising tools and screening instruments in use for assessing dementia in persons with ID,[13,25,27,28] no systematic review on measurement properties using up-to-date guidelines for review conduction and psychometric evaluation has been conducted so far. We want to provide an inventory of available informant-based instruments and their measurement properties. This should help clinicians and researches in choosing the adequate instrument for their respective purpose. Our review adds to the existing body of knowledge by using a very inclusive systematic search of the literature and, most

importantly, by providing a systematic evaluation of informant-based dementia assessment instruments following up-to-date guidelines.

For each instrument, we will systematically summarise the amount and quality of available evaluation studies, depicting which measurement properties have been evaluated to what extent, and which measurement properties have not or insufficiently been evaluated.

The objectives of this systematic review are to (1) identify informant-based instruments suitable for the assessment of dementia in persons with ID, to (2) provide a systematic overview of descriptive aspects for each instrument (e.g. respondent requirements, response format), to (3) provide a systematic overview of the amount and quality of available research for each instrument and each measurement property, and to (4) provide a recommendation for the most suitable instrument(s) based on all information collected.

## METHODS AND ANALYSIS

This review will be conducted and reported according to the *Preferred Reporting Items for Systematic Reviews and Meta-Analyses* (PRISMA) statement.[29] The review protocol has been developed using the PRISMA guidelines for protocols (PRISMA-P).[30,31] We will adhere to the *COnsensus-based Standards for the selection of health Measurement INstruments* (COSMIN) guidelines,[32] and complement them with a set of characteristics especially developed for assessment instruments for persons with ID, the *Characteristics of Assessment Instruments for Psychiatric Disorders in Persons with Intellectual Developmental Disorders* (CAPs-IDD).[33] The systematic review has been registered with the *International Prospective Register of Systematic Reviews* (PROSPERO) with registration number CRD42020181773. If amendments to the protocol are needed, we will register these in PROSPERO, including date and rationale. In the final publication of our results, any amendments to the protocol will be depicted and explained.

## Search strategy

Two systematic searches will be applied consecutively, and carried out between May 2020 and August 2020. The first search should provide an inventory of available informant-based assessment instruments for dementia in persons with ID. The goal of the second search is to locate evaluation studies for each instrument found in the first search. Figure 1 and Figure 2 depict our search strategies using PRISMA flow charts.

### First search

To identify instruments we will search in ten international electronic databases, including ASSIA, CINAHL, Cochrane Library, ERIC, MEDLINE, PsycINFO, Scopus, Web of Science, OpenGrey, and ProQuest Dissertations & Theses Global. The search strategy is described in Table 1 and depicted in detail in the supplementary file. It will include various terms for the (1) output of interest, (2) construct of interest, and (3) the specified population. As persons with Down Syndrome are very prone to develop dementia, this subgroup of persons with ID is included in our search strategy. We will use a limit on the timespan of publication in the first search, not including publications before the year 2012. Instruments published up to the year of 2012 are summarised in a previous systematic review.[13] This review used a very inclusive search strategy and listed all assessment instruments that have been used to assess dementia in persons with ID. We will examine the total of 114 dementia assessment instruments listed in the review of 2013, and include those instruments that are in line with our inclusion criteria.

Table 1: Search strategy for the first search

| | 1: Output | 2: Construct | 3: Population |
|---|---|---|---|
| Search terms | Assessment instruments | Dementia | Intellectual disability |

| | | | |
|---|---|---|---|
| Synonyms | assessment; diagnostic; diagnosis; screening; instrument; tool; measurement; questionnaire; psychometrics; scale; interview | dementia; Alzheimer's disease | intellectual disability; learning disability; intellectual developmental disorder; trisomy 21, Down syndrome |
| Combined and truncated | assess* OR diagnosti* OR screen OR screening* OR instrument* OR tool* OR measure* OR questionnaire* OR psychometr* OR scale* OR interview* | dement* OR Alzheimer* | ((intellectual* OR learning) AND disab*) OR (intellectual* AND developmental* AND disorder*) OR trisom* 21 OR (down* AND syndrom*) |
| Example search string for SCOPUS | TITLE-ABS-KEY ( ( assess* OR diagnosti* OR screen OR screening* OR instrument* OR tool* OR measure* OR questionnaire* OR psychometr* OR scale* OR interview* ) AND ( dement* OR alzheimer* ) AND ( ( ( intellectual* OR learning ) AND disab* ) OR ( intellectual* AND developmental* AND disorder* ) OR trisom* 21 OR ( down* AND syndrom* ) ) ) AND PUBYEAR > 2011 | | |

Inclusion criteria for the first search will be: (1) studies need to focus on assessing dementia in persons with ID, (2) describe the development or evaluation of an informant-based instrument for the assessment of dementia, (3) and this instrument has to be especially developed or adapted for persons with ID. Exclusion criteria: (1) classification systems like ICD-11, DSM-5, (2) scales including dementia, but focusing on a broader spectrum of disorders for screening purposes or differential diagnosis, such as the *Psychiatric Assessment Schedule for Adult with Developmental Disability* (PAS-ADD).[34]

Second search

Once we have identified the instruments, we will conduct a search by citation strategy using

the initial publications of each instrument as a reference point. This search strategy was chosen on the assumption that a paper evaluating an instrument would surely cite the initial publication of the respective instrument. The papers used as reference points will also be included in the further appraisal of the literature. For published papers, we will use five international databases allowing a search by citation strategy, including ERIC, PsycInfo, MEDLINE, Scopus, and Web of Science. For published manuals, not listed in at least one of the five databases, we will use Google Scholar. Additionally, all records fulfilling the inclusion and exclusion criteria of the first search will be transferred and examined in the second search.

The following inclusion criterion will be used in the second search: (1) studies need to describe an evaluation of the respective instrument in persons with ID. Exclusion criteria comprise: (1) the respective instrument was used primarily for other investigations, not related to an evaluation of the instrument, (2) or the study is a review on assessment instruments, not providing novel information.

To further include grey and unpublished literature in both searches, we will apply an invisible college approach, contacting authors in the field for information or manuscripts on this topic, and we will follow up on meeting abstracts. Full texts of reviews on assessment instruments identified in the course of the two searches will be screened for possible further studies to include. References of papers meeting the inclusion criteria will be hand-searched. We will re-run both searches before the final analyses to include the most recent publications.

For study selection, one reviewer will exclude duplicates. All remaining records will be screened and reviewed for eligibility by two team members independently, i.e. blinded to each other's decisions. In the case of disagreement, dissonances will be discussed until agreement is reached. In the case of non-agreement, a third team member will be included in discussion.

## Data extraction

The first search will result in a list of instruments. Data extracted will be the names of the instruments and information on their initial publication(s). In the second search, we will extract evaluation data of instruments, i.e. measurement properties and characteristics as listed in the COSMIN checklists and the CAPs-IDD. For each characteristic/property extracted, we will record the study design and sample characteristics, including sample size, gender distribution, age distribution, aetiology of ID, and country (language) in which the instrument was evaluated. We will include all studies, irrespective of their design.

The extraction of all relevant data will be done via standardised and piloted excel spreadsheets by two team members independently. In the case of disagreement, dissonances will be discussed until agreement is reached. In the case of non-agreement, a third team member will be included in discussion. If data necessary for coding is missing in a study, we will contact the respective study authors for this information.

## Risk of bias and quality assessment

Quality and risk of bias will be assessed on study level (for each measurement property), on outcome level (for each assessment instrument), and on an aggregated outcome level, applying the *Grading of Recommendations, Assessment, Development and Evaluation* (GRADE) approach. We will combine the COSMIN checklists [35–37] with the CAPs-IDD [33], a comprehensive tool specifically developed for the evaluation of assessment instruments for psychiatric disorders in persons with ID. The CAPs-IDD consists of two parts: (1) conceptual and measurement model (including descriptive aspects of instruments, e.g. respondent requirements, theoretical foundation), and (2) psychometric properties. We will only use the first part, as the second part is more comprehensively covered by the COSMIN checklists.

All ratings will be done by two reviewers independently. In the case of disagreement, dissonances will be discussed until agreement is reached. In the case of non-agreement, a third team member will be included in the discussion. Initial interrater agreement will be determined using percentage agreement, calculated in R.[38]

As to publication bias, we assume that evaluation results not in favour of the respective instruments are likely to be underreported. This may be partly due to evaluations being frequently done and published by the developer(s) of the respective instrument. We will address this by including grey literature and by discussing this aspect in the interpretation of our results.

**Strategy for data synthesis**

A narrative synthesis will be conducted. Assessment instruments will be presented in a table along with descriptive aspects according to CAPs-IDD, and their measurement properties and quality ratings according to the COSMIN checklists. Quantitative data pooling will probably not be possible. This is due to an expected limited number of studies evaluating the same property (e.g. internal consistency) for an instrument, and an expected heterogeneity in the population studied (e.g. severity of ID, persons with Down Syndrome vs. persons with ID of other aetiology). However, if applicable, we will calculate pooled estimates and 95% confidence intervals using R.[38]

**Analysis of subgroups**

We define persons with Down Syndrome/trisomy 21 (DS) as a special subgroup, as they are more often affected by Alzheimer's dementia, with a suspected earlier onset.[16] We will group instruments according to their intended use, and studies according to their participants in four clusters: (1) persons with ID, including persons with DS, (2) only persons with DS, (3) only persons with ID, not including DS, (4) aetiology of ID not specified. For the fourth cluster, we will contact study authors to determine aetiology of ID in the respective sample or

for the respective instrument. We will then allocate each study or instrument to the first three clusters according to the information provided by the authors. If no information is provided, the respective study or instrument remains in cluster four.

**Confidence in cumulative evidence**

The modified GRADE approach as suggested by the COSMIN guidelines[32] will be applied to grade the quality of the evidence.

**Data management**

We will use ZOTERO for saving records and managing and storing literature, including managing duplicates. For extracting data and recording decisions on quality ratings we will use standardised and piloted excel spreadsheets.

**Patient and public involvement**

This research was done without patient involvement due to limited resources.

## DISCUSSION

This review will summarise measurement properties of available informant-based assessment instruments for persons with ID and give an overview of the quality of each instrument and the quality of available evaluation studies. For each instrument we will depict which psychometric properties are evaluated to what extent, and which properties need further evaluation in future research. This will be the first systematic review of dementia assessment instruments for persons with ID using PRISMA and COSMIN guidelines as well as applying the ID-specific criteria of the CAPs-IDD.

Our work will highlight gaps in research on these instruments, thus setting the ground for more effective research in the future. The results of this review will inform researchers and clinicians of the quality of available instruments to assess dementia in persons with ID, and guide them in choosing an adequate instrument. This will hopefully contribute to an

improvement of dementia assessment in persons with ID and a better, earlier, and more adequate provision of healthcare services, as demanded by the UN-CRPD.[24]

## Ethics and dissemination

No ethics statement is needed for this study. The results of this systematic review will be submitted for publication to a leading peer-reviewed journal, and presented at international conferences and congresses in the fields of ID, ageing, and dementia.

## Author's contributions

ELZ conceived the study, drafted the protocol, and is the guarantor of the review. SK, IZ and FF contributed to study design and drafting the protocol. ELZ, SK, and KW designed and tested the search strategy. FF and IZ tested quality rating tools and software options. All authors read and approved the final protocol.

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## Patient and public involvement

This research was done without patient involvement.

## Competing interests

The authors have no competing nor potential conflict interests to declare.

**Word Count:** 2,566 words.

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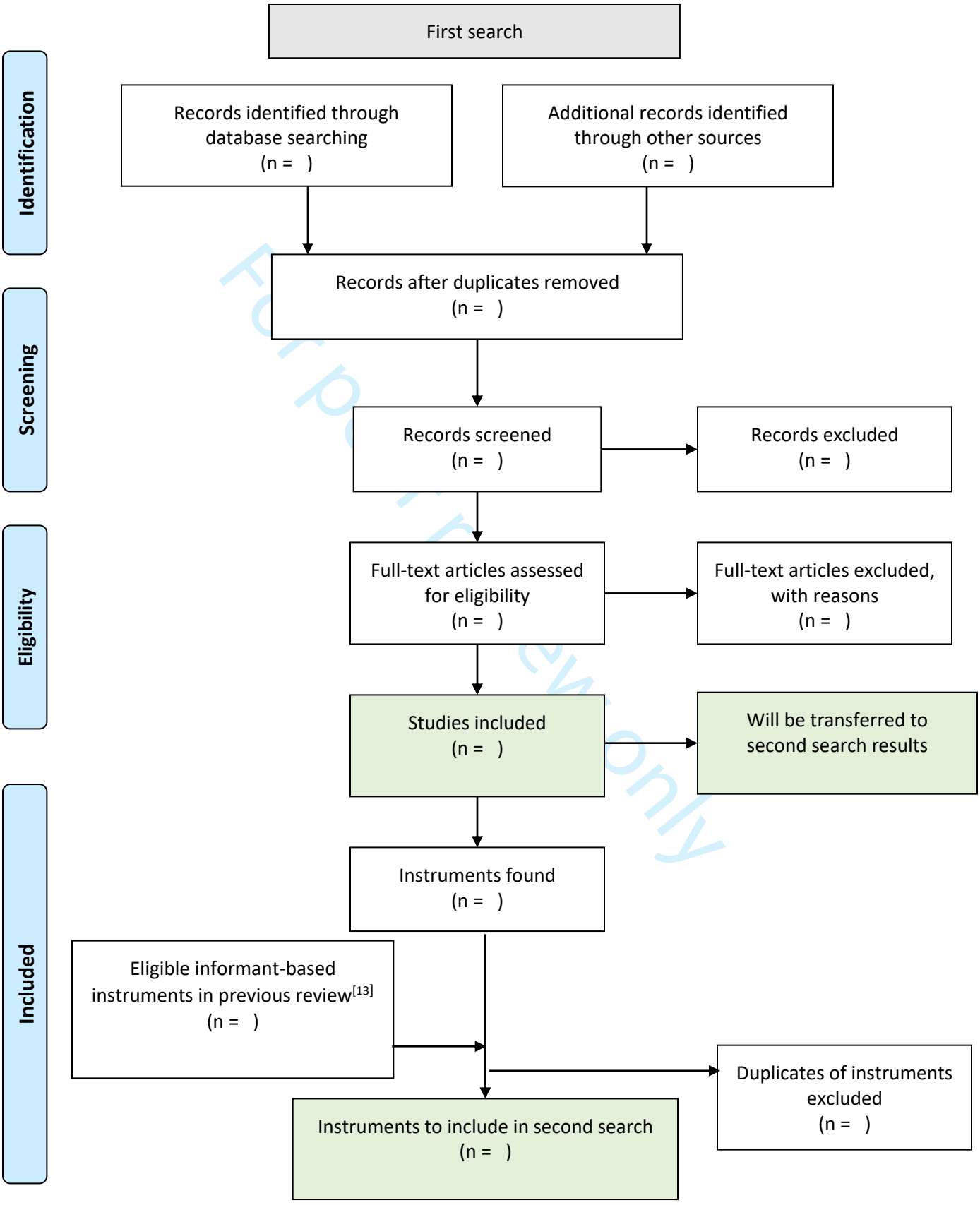
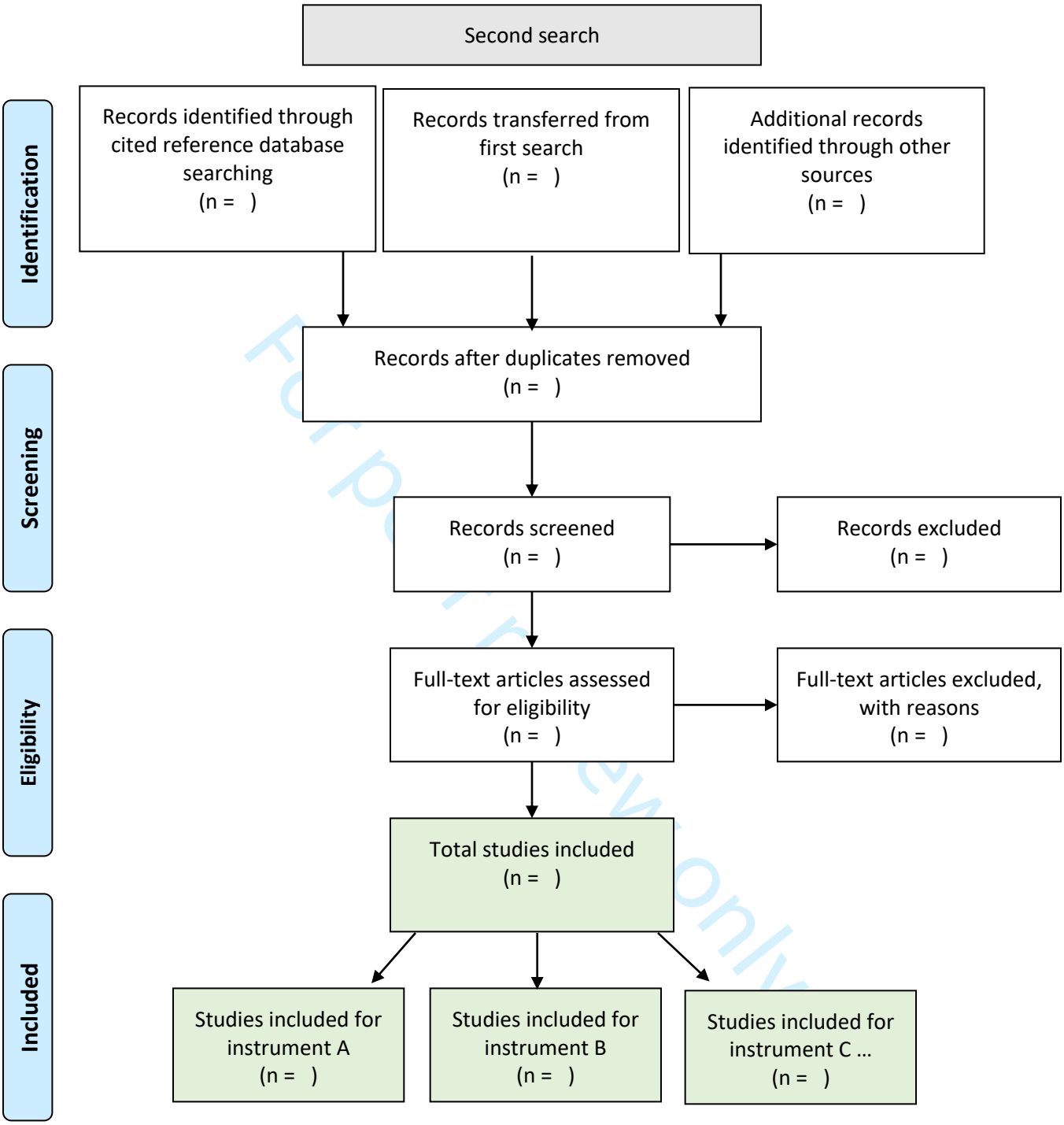
## Figure legends

Figure 1: PRISMA flow chart of first search

Figure 2: PRISMA flow chart of second search





**Supplementary information**

**Informant-based assessment instruments for dementia and their measurement properties in persons with intellectual disability: a systematic review protocol**

Detailed search strategy in electronic databases

**ASSIA**
(via ProQuest)

noft(Assess* OR diagnosti* OR screen OR screening* OR instrument* OR tool* OR measure* OR questionnaire* OR psychometr* OR scale* OR interview*) AND noft(Dement* OR Alzheimer*) AND noft(((intellectual* OR learning) AND disab*) OR (intellectual* AND developmental* AND disorder*) OR trisom* 21 OR (down* AND syndrom*))

Additional limits: Date: From 2012 to 2020

**CINAHL**
(via EBSCOhost) (MEDLINE records excluded, since they are already in the MEDLINE Search):

(AB ( Assess* OR diagnosti* OR screen OR screening* OR instrument* OR tool* OR measure* OR questionnaire* OR psychometr* OR scale* OR interview* ) AND AB ( Dement* OR Alzheimer* ) AND AB ( ((intellectual* OR learning) AND disab*) OR (intellectual* AND developmental* AND disorder*) OR trisom* 21 OR (down* AND syndrom*) ) ) OR (TI ( Assess* OR diagnosti* OR screen OR screening* OR instrument* OR tool* OR measure* OR questionnaire* OR psychometr* OR scale* OR interview* ) AND TI ( Dement* OR Alzheimer* ) AND TI ( ((intellectual* OR learning) AND disab*) OR (intellectual* AND developmental* AND disorder*) OR trisom* 21 OR (down* AND syndrom*) ) ) OR (SU ( Assess* OR diagnosti* OR screen OR screening* OR instrument* OR tool* OR measure* OR questionnaire* OR psychometr* OR scale* OR interview* ) AND SU ( Dement* OR Alzheimer* ) AND SU ( ((intellectual* OR learning) AND disab*) OR (intellectual* AND developmental* AND disorder*) OR trisom* 21 OR (down* AND syndrom*) ) )

**Limiters** - Published Date: 20120101-20201231; Exclude MEDLINE records

**Cochrane Library**

(Assess* OR diagnosti* OR screen OR screening* OR instrument* OR tool* OR measure * OR questionnaire* OR psychometr* OR scale* OR interview*):ti,ab,kw *AND* (Dement* OR Alzheimer*):ti,ab,kw *AND* (((intellectual* OR learning) AND disab*) OR (intellectual* AND developmental* AND d isorder*) OR trisom* 21 OR (down* AND syndrom*)):ti,ab,kw

with Cochrane Library publication date from Jan 2012 to Dec 2020

**ERIC, MEDLINE, and PsycInfo**
(via Ovid) [mp=ab, ti, hw, id, tc, ot, tm, mh]

((Assess* or diagnosti* or screen or screening* or instrument* or tool* or measure* or questionnaire* or psychometr* or scale* or interview*) and (Dement* or Alzheimer*) and (((intellectual* or learning) and disab*) or (intellectual* and developmental* and disorder*) or trisom* 21 or (down* and syndrom*))).mp

limit to yr="2012 - 2020"

**SCOPUS**

TITLE-ABS-KEY ( ( assess* OR diagnosti* OR screen OR screening* OR instrument* OR tool* OR measure* OR questionnaire* OR psychometr* OR scale* OR interview* ) AND ( dement* OR alzheimer* ) AND ( ( ( intellectual* OR learning ) AND disab* ) OR ( intellectual* AND developmental* AND disorder* ) OR trisom* 21 OR ( down* AND syndrom* ) ) ) AND PUBYEAR > 2011

**Web of Science Core Collection**:

**TOPIC:**(Assess* OR diagnosti* OR screen OR screening* OR instrument* OR tool* OR measure* OR questionnaire* OR psychometr* OR scale* OR interview*) *AND*
**TOPIC:**(Dement* OR Alzheimer*) *AND*
**TOPIC:**(((intellectual* OR learning) AND disab*) OR (intellectual* AND developmental* AND disorder*) OR trisom* 21 OR (down* AND syndrom*))

**Timespan:** 2012-2020. **Indexes:** SCI-EXPANDED, SSCI, A&HCI, CPCI-S, CPCI-SSH, BKCI-S, BKCI-SSH, ESCI, CCR-EXPANDED, IC.

**OpenGrey**
(Assess* OR diagnosti* OR screen OR screening* OR instrument* OR tool* OR measure* OR questionnaire* OR psychometr* OR scale* OR interview*) *AND*
(Dement* OR Alzheimer*) *AND*
(((intellectual* OR learning) AND disab*) OR (intellectual* AND developmental* AND disorder*) OR trisom* 21 OR (down* AND syndrom*))

**ProQuest Dissertations & Theses**
noft(Assess* OR diagnosti* OR screen OR screening* OR instrument* OR tool* OR measure* OR questionnaire* OR psychometr* OR scale* OR interview*) AND noft(Dement* OR Alzheimer*) AND noft(((intellectual* OR learning) AND disab*) OR (intellectual* AND developmental* AND disorder*) OR trisom* 21 OR (down* AND syndrom*))

Additional limits: Date: From 2012 to 2020

# Reporting checklist for protocol of a systematic review.

Based on the PRISMA-P guidelines.

## Instructions to authors

Complete this checklist by entering the page numbers from your manuscript where readers will find each of the items listed below.

Your article may not currently address all the items on the checklist. Please modify your text to include the missing information. If you are certain that an item does not apply, please write "n/a" and provide a short explanation.

Upload your completed checklist as an extra file when you submit to a journal.

In your methods section, say that you used the PRISMA-Preporting guidelines, and cite them as:

Moher D, Shamseer L, Clarke M, Ghersi D, Liberati A, Petticrew M, Shekelle P, Stewart LA. Preferred Reporting Items for Systematic Review and Meta-Analysis Protocols (PRISMA-P) 2015 statement. Syst Rev. 2015;4(1):1.



| | | Reporting Item | Page Number | Line Number |
|---|---|---|---|---|
| **Title** | | | | |
| Identification | #1a | Identify the report as a protocol of a systematic review | 1 | 2-3 |

| | | | | |
|---|---|---|---|---|
| Update | #1b | If the protocol is for an update of a previous systematic review, identify as such | n.a. | |
| **Registration** | | | | |
| | #2 | If registered, provide the name of the registry (such as PROSPERO) and registration number | 6 | 141-142 |
| **Authors** | | | | |
| Contact | #3a | Provide name, institutional affiliation, e-mail address of all protocol authors; provide physical mailing address of corresponding author | 1 | 5-20 |
| Contribution | #3b | Describe contributions of protocol authors and identify the guarantor of the review | 13 | 279-283 |
| **Amendments** | | | | |
| | #4 | If the protocol represents an amendment of a previously completed or published protocol, identify as such and list changes; otherwise, state plan for documenting important protocol amendments | 6 | 142-144 |
| **Support** | | | | |
| Sources | #5a | Indicate sources of financial or other support for the review | 13 | 284-286 |

| | | | | |
|---|---|---|---|---|
| Sponsor | #5b | Provide name for the review funder and / or sponsor | n.a. | |
| Role of sponsor or funder | #5c | Describe roles of funder(s), sponsor(s), and / or institution(s), if any, in developing the protocol | n.a. | |
| **Introduction** | | | | |
| Rationale | #6 | Describe the rationale for the review in the context of what is already known | 5-6 | 105-122 |
| Objectives | #7 | Provide an explicit statement of the question(s) the review will address with reference to participants, interventions, comparators, and outcomes (PICO) | 6 | 126-131 |
| **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 | 7-8, 9 | 157-172, 180-188 |
| 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 | 7, 9 | 152-154, 178-181, 189-194 |

| | | | | |
|---|---|---|---|---|
| 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 | 8; supplement | 164-165 |
| Study records - data management | #11a | Describe the mechanism(s) that will be used to manage records and data throughout the review | 12 | 254-257 |
| Study records - selection process | #11b | State the process that will be used for selecting studies (such as two independent reviewers) through each phase of the review (that is, screening, eligibility and inclusion in meta-analysis) | 9 | 195-198 |
| Study records - 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 | 10 | 207-211 |
| Data items | #12 | List and define all variables for which data will be sought (such as PICO items, funding sources), any pre-planned data assumptions and simplifications | 9-10 | 199-206 |
| Outcomes and prioritization | #13 | List and define all outcomes for which data will be sought, including prioritization of main and additional outcomes, with rationale | n.a. | |

| | | | | |
|---|---|---|---|---|
| Risk of bias in 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 | 10-11 | 212-231 |
| Data synthesis | #15a | Describe criteria under which study data will be quantitatively synthesised | 11 | 235-240 |
| Data synthesis | #15b | If data are appropriate for quantitative synthesis, describe planned summary measures, methods of handling data and methods of combining data from studies, including any planned exploration of consistency (such as I2, Kendall's τ) | n.a. | |
| Data synthesis | #15c | Describe any proposed additional analyses (such as sensitivity or subgroup analyses, meta-regression) | n.a. | |
| Data synthesis | #15d | If quantitative synthesis is not appropriate, describe the type of summary planned | 11 | 232-235 |
| Meta-bias(es) | #16 | Specify any planned assessment of meta-bias(es) (such as publication bias across studies, selective reporting within studies) | 11 | 227-231 |

| | | | | |
|---|---|---|---|---|
| Confidence in cumulative evidence | #17 | Describe how the strength of the body of evidence will be assessed (such as GRADE) | 12 | 251-253 |