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Outcome Measures Evaluating Physical Functioning and their Measurement Properties in Adolescent Idiopathic Scoliosis: Protocol for a Systematic Review

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ABSTRACT

Introduction Adolescent Idiopathic Scoliosis (AIS) can impact on quality of life. Physical functioning is one outcome that provides an evaluation of meaningful aspects of an individual's life and can be assessed through Patient–Reported Outcome Measures (PROMs), Performance-Based Outcome Measures (PBOMs) or Impairment Measure (IMs). Measures need to be valid, reliable and responsive to change to evaluate the effects of an intervention on physical functioning ability. In the absence of existing evidence this systematic review will appraise the evidence on the measurement properties of physical functioning in AIS.

Methods/analysis A protocol for a systematic review and meta-analysis informed by Cochrane guidelines and reported in line with the Preferred Reporting Items for Systematic Reviews and Meta-Analysis-P. Key databases will be searched including MEDLINE, PsycINFO, EMBASE, CINHAL, SPORT discus, Web of science and PubMed. The search strategy will be in two stages to (1) identify outcome measures used for assessment of physical functioning in AIS (2) evaluate the measurement properties (i.e. validity, reliability and responsiveness) of the identified measures. Two reviewers will independently perform study selection, data extraction, risk of bias, and overall quality assessment. The Consensus-based Standards for the selection of Health Measurement Instruments (COSMIN) risk of bias and a modified Grading of Recommendations, Assessment, Development and Evaluation (GRADE) guidelines will be used.

Ethics and dissemination: Since no patient data are being collected, ethical approval is not necessary. The results will be disseminated through a peer-reviewed publication and conference presentation.

PrOsPErO registration number: CRD42019142335

Key words: Adolescent Idiopathic Scoliosis, Outcome Measure, Measurement Properties, Physical Functioning, Functioning

ARTICLE SUMMARY

Strengths and Limitations of This Study

- This will be the first systematic review to evaluate the measurement properties of all outcome measures evaluating physical functioning in AIS.
- This study will employ rigorous methods and using COnsensus-based Standards for the selection of health Measurement Instruments risk of bias tool and modified GRADE.
- Although other measures of functioning such as role and social functioning are important to an AIS population, this study is focused on outcome measures of physical functioning.

INTRODUCTION

Scoliosis is a complex three-dimensional deformity of the spine, including lateral curvature and rotation of the vertebrae [1] and characterised by a curve angle ≥10°. [2] There are two main types of scoliosis, idiopathic and non-idiopathic with the latter arising from congenital, neuromuscular or mesenchymal causes.[3] While the aetiology of idiopathic scoliosis remains unknown; genetic, hormonal, and mechanical factors are involved. [4] Adolescent Idiopathic Scoliosis (AIS) often develops between 10 and 16 years of age and represents ~85% of cases .[5] AIS is the most common spinal deformity among the pediatric population, with a prevalence ranging from 2% to 3%. [6] Nearly 80% of those affected present with a curvature of the thoracic or thoracolumbar/lumbar region.[3] Whilst males and females are equally affected, females are reported to be at 10 times greater risk of curve progression. [1]

A number of health-related problems are reported among AIS individuals including; lower quality of life, [7] back pain, [8] pulmonary dysfunction, [9] stress, [10] mental health disorders. [11] A major component of health status and health-related quality of life is physical functioning, [12] which can be used to identify individuals at risk of disability and to predict health and social care use. [13, 14] Accordingly, physical functioning is included in the Core Outcome Set (COS) for use within clinical trials for many musculoskeletal conditions, [12, 15, 16] including adolescents with spinal deformity.[16] Limitations in physical functioning are reported by individuals with AIS, e.g. walking, moving around, maintaining body position and lifting objects. [17-19] Additionally, pain is often reported in individuals with AIS which may cause functional limitations. [8, 20-22]

Physical functioning can be assessed with IMs, PROMs and PBOMs, [23] with IMs such as Cobb degree, a commonly reported outcome measure in AIS. [24] Impairment measures give an indication about dysfunctions in structure or organs, but fail to fully capture functional limitations. [23] The most widely used PROM for assessment of the quality of life as well as physical functioning of individuals with spinal deformity is the Scoliosis Research Society (SRS) questionnaire, [24, 25] and variants of such. [26-28] The SRS is mostly used among surgically treated AIS individuals, [25, 28, 29] but may not be applicable to those treated conservatively. [30] Although relevant, PROMs are influenced by patients' perception of their abilities to perform activities and lack sensitivity to change. [23] Measures such as PBOMs have the potential to provide unbiased and reproducible assessments of physical functioning during the performance of activities of daily living, [31-33] such as walking speed, trunk muscle endurance testing and balance. [23, 33] Within the AIS population, little is known about the available PBOMs for evaluating physical functioning. A recent COS study for adolescents and young adults with spinal deformity, identified the SRS-22r as a measure of quality of

life which includes physical functioning. [16] However, the SRS-22r fails to fully capture important aspects of physical functioning for AIS population e.g. self-care and mobility [17]. Where this study included all forms of spinal deformities, the heterogeneity limits it is applicability to AIS as a discrete population.

Establishment of measurement properties of outcome measures are important to avoid the risk of bias and ensure accuracy in the evaluation of test results.[34] The COnsensus-based Standards for the selection of health Measurement INstruments, (COSMIN) group developed a taxonomy of measurement properties to improve the selection of outcome measures. [35] Three main domains identified, reliability, validity and responsiveness with further subgrouping.[35] In the absence of existing evidence, a systematic review is needed to inform and summarise the evidence on physical functioning outcome measure in AIS population and to evaluate their measurement properties.

Objective

To synthesise evidence of physical functioning outcome measures in AIS population. A secondary aim is to evaluate measurement properties of outcome measures evaluating physical functioning in AIS.

METHODS

This protocol has been informed by expertise in the field including a surgeon, musculoskeletal rehabilitation experts including physiotherapists and individuals with methodological expertise. It has been designed in line with the Cochrane handbook [36] and is reported in line with the Preferred Reporting Items for Systematic Reviews and Meta-Analysis-P (PRISMA-P) (Supplementary File 1).[37]

Eligibility criteria

Inclusion criteria

Participants

Participants aged between the age of 10 years until the end of bone growth with a diagnosis of idiopathic scoliosis and ≥10° Cobb angle will be considered. The end of bone growth is estimated by the Risser classification, which grades the level of ossification and fusion of the iliac crest apophyses

into six stages [38] where stage 0 describes no ossification, and Stage 5 represents complete ossification and fusion of the iliac apophysis and end of bone growth. [38]

Outcome measures

Any study that includes assessments of the physical functioning of AIS using specific outcome measure will be included. Physical functioning defined according to the Core Outcome Measures in Effectiveness Trials (COMET) taxonomy [15] as any physical activities of daily living such as the ability to walk, independence, self-care, performance status, disability index. [15, 39] The outcome measures being defined as any one of the following:

1. Patient Reported Outcome Measures (PROMs) in the form of questionnaires or scales designed for AIS to evaluate physical functioning (e.g. SRS-22r) and/or
2. Performance-based outcome measures (PBOMs); a measure of physical functioning by clinician while the individual is performing a functional task (e.g. standing balance, walking speed and/or
3. Impairment measure (IMs) which means any dysfunction in a specific body part or system which limits function, such as muscle performance and range of motion. [23]

Measurement properties

Any study that has evaluated one or more measurement properties of the above-mentioned outcome measures in AIS. This will include all measurement properties in the three main domains of the COSMIN Taxonomy (i.e. reliability, validity, and responsiveness). [35] The definitions of measurement properties according to COSMIN taxonomy are summarised in the online supplementary file 2.

Study design

In the first stage, all study designs including; randomised clinical trials, cohort, observational studies and case studies will be included to identify all outcome measure of physical functioning being used within AIS population. The second stage will include the validation studies of the physical functioning measures identified in the first stage.

Information sources

A search strategy has been developed using medical subject headings, where available and relevant text words relating to AIS, physical functioning, outcome measures and measurement properties. An electronic search of databases will be conducted including MEDLINE (OVID interface), PsycINFO (OVID interface), EMBASE (OVID interface), SPORT discus (EBSCO interface), CINHAL (EBSCO interface), Web of Science and PubMed. A hand search in the key journals including Spine, The Spine Journal, Spine Deformity, Scoliosis and Spinal Disorder, Scoliosis and European Spine Journal as well as contacting relevant leading researchers in the field. Further, searching of the Grey literature, including conference proceedings, British National bibliography for report literature, open-Grey, dissertation abstracts and EThOS will be conducted.

Search strategy

Two reviewers (SA, EB) will independently complete searches and identify potential studies for eligibility. To ensure that all relevant data are included, no limitations will be applied. Initial search terms will be developed for MEDLINE and then adapted with relevant syntax and subject headings for other databases. Recommended search filters specifically designed for retrieving articles on measurement properties will be used where appropriate.[40] An example of the search strategy of both stages is available as an online supplementary file 3.

Data management

Search records will be imported into Endnote Version X9 (Clarivate Analytics). Using Endnote, the abstracts and full texts will be stored, and any duplicates will be identified and removed.

Selection Process

Two reviewers (SA, EB) will independently search information sources and assess study eligibility according to inclusion/exclusion criteria by grading each study as eligible/not eligible/might be eligible. [41] If a study cannot be clearly excluded based on the inclusion criteria, the full text will be retrieved. Study selection (included and excluded studies) with the reasons for exclusion, will be summarised in a PRISMA flow diagram (Supplementary file 1).[37] Articles will be included if both reviewers agreed that the eligibility criteria were met. Any disagreement will be first discussed and the third reviewer (NH) will mediate situations of disagreement. The percentage of agreement between reviewers on the extracted data will be reported.

Data collection process

Two reviewers (SA, EB) will independently extract the data of eligible studies. A bespoke data extraction form will be used and piloted on 3 studies. If information is not available in the studies, authors will be contacted. Any disagreement between reviewers will be mediated through discussion with a third reviewer (NH) if needed.

Data items

The data that will be extracted from each study at each stage is summarised in Table 1. In the case of missing data, the authors of the study will be contacted.

Table 1: Summary of items to be extracted from included studies

General information	Author(s), Year of publication, Country
Study and participants Characteristics	Design of study, Sample size, Age, Gender. Type of intervention (bracing, physiotherapy, exercise, or surgery)
Outcome measure	Type of measures (patient-reported measure, performance-based measure, impairment measure)
Outcome domain	Physical Functioning
Measurement properties	Measurement properties assessing statistical method used and results

Outcomes and prioritisation

Since there is no gold standard outcome measure for physical functioning, no primary outcome measure is identified for this review.

Risk of bias in individual studies

The COSMIN checklist for assessment of risk of bias and methodological quality in individual studies will be used.[42] It was revised and specifically designed for use in systematic reviews of PROMs to evaluate studies on measurement properties. [42] The checklist includes standards for each measurement property for both design and preferred statistical methods, and rates each study as either very good, adequate, doubtful or inadequate quality.[42, 43] The COSMIN group recommend researchers to adapt the checklist to other measures (i.e. PBOMs, IBOM) since it was originally developed for PROMs [43]. Two independent reviewers (SA, EB) will assess the risk of bias for all included studies. Any disagreement will be resolved through discussion, and if no agreement is

reached, a third reviewer (NH) will be consulted. The percentage of agreement between reviewers will be reported in the final results.

Data synthesis

Scoping searches of the currently available literature indicate heterogeneity of the outcome measures used for assessment of physical functioning in AIS. Thus, a meta-analysis may not be possible. If deemed not possible, a narrative synthesis will be conducted. The COSMIN guidelines for systematic reviews will be used for the synthesis of the results.[43] The methodological quality of each single study on a measurement property will be assessed using the COSMIN Risk of Bias checklist (Online supplementary file 2).[42] The result will be rated against the predefined criteria for good measurement properties as sufficient (+), insufficient (–), inconsistent (±), or indeterminate (?). [43] The evidence will be summarised for each measurement property for PROM, PBOM or IM and the overall result is rated against the criteria for good measurement properties. [43]

Confidence in cumulative evidence

The overall quality and strength of evidence will be assessed using a modified Grading of Recommendations Assessment, Development and Evaluation (GRADE) approach for systematic reviews of clinical trials. [43] The GRADE approach uses five factors to determine the quality of the evidence: risk of bias (quality of the studies), inconsistency (of the results of the studies), indirectness (evidence comes from different populations, interventions or outcomes than the ones of interest in the review), imprecision (wide confidence intervals), and publication bias (negative results are less often published).[44] For evaluating measurement properties in systematic reviews of PROMs, only four factors will be assessed as recommended by COSMIN, while the fifth factor (publication bias) was removed as there is no registry exists for measurement properties. [43]

Patient and public involvement

The study question and systematic review protocol were informed following discussion at a patient and public involvement meeting at the Centre of Precision Rehabilitation for Spinal Pain at the University of Birmingham. Since no patient data is needed, patients will not be involved in data collection or analysis. However, the results of the study will be shared at public engagement events.

Implications of this study

The AIS is a complex deformity of the spine and causes a significant impact on physical activities of individuals' daily living such as lifting objects and maintaining body position. [17, 19] In consequence, the quality of life is affected. Physical functioning gives an indication about the current health status and identifies people at risk of disability.[12, 13] Therefore, physical functioning is considered as one of the outcomes that should be assessed and reported in clinical trials of musculoskeletal conditions. [15] Within the AIS population, little attention has been paid to the physical functioning measures and their measurement properties. This systematic review will be the first assessing the measurement properties of physical functioning outcome measures among the AIS population. This will inform clinicians and researchers of the best available tools for assessment of physical functioning in AIS.

Declarations

Ethics and Dissemination

No ethics approval is required for this systematic review. The results will be disseminated through a peer-reviewed publication and conference presentation.

Author Contributions

All authors conceptualized and designed the protocol. SA is a PhD student and NH (lead supervisor), AR and DF are supervisors. SA drafted the initial manuscript with NH, AR and DF all providing guidance on design, topic, methodology and analyses. All authors reviewed and commented on each draft of the protocol. All authors have approved and contributed to the final manuscript.

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Competing interests

None declared.

Patient consent for publication

Not required.

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PRISMA-P (Preferred Reporting Items for Systematic review and Meta-Analysis Protocols) 2015 checklist: recommended items to address in a systematic review protocol*

Section and topic	Item No	Checklist item
ADMINISTRATIVE INFORMATION		
Title:		
Identification	1a	Identify the report as a protocol of a systematic review
Update	1b	If the protocol is for an update of a previous systematic review, identify as such
Registration	2	If registered, provide the name of the registry (such as PROSPERO) and registration number
Authors:		
Contact	3a	Provide name, institutional affiliation, e-mail address of all protocol authors; provide physical mailing address of corresponding author
Contributions	3b	Describe contributions of protocol authors and identify the guarantor of the review
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
Support:		
Sources	5a	Indicate sources of financial or other support for the review
Sponsor	5b	Provide name for the review funder and/or sponsor
Role of sponsor or funder	5c	Describe roles of funder(s), sponsor(s), and/or institution(s), if any, in developing the protocol
INTRODUCTION		
Rationale	6	Describe the rationale for the review in the context of what is already known
Objectives	7	Provide an explicit statement of the question(s) the review will address with reference to participants, interventions, comparators, and outcomes (PICO)
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
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
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
Study records:		
Data management	11a	Describe the mechanism(s) that will be used to manage records and data throughout the review

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)
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
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
Outcomes and prioritization	13	List and define all outcomes for which data will be sought, including prioritization of main and additional outcomes, with rationale
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
Data synthesis	15a	Describe criteria under which study data will be quantitatively synthesised
	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 I^2, Kendall's τ)
	15c	Describe any proposed additional analyses (such as sensitivity or subgroup analyses, meta-regression)
	15d	If quantitative synthesis is not appropriate, describe the type of summary planned
Meta-bias(es)	16	Specify any planned assessment of meta-bias(es) (such as publication bias across studies, selective reporting within studies)
Confidence in cumulative evidence	17	Describe how the strength of the body of evidence will be assessed (such as GRADE)

*** It is strongly recommended that this checklist be read in conjunction with the PRISMA-P Explanation and Elaboration (cite when available) for important clarification on the items. Amendments to a review protocol should be tracked and dated. The copyright for PRISMA-P (including checklist) is held by the PRISMA-P Group and is distributed under a Creative Commons Attribution Licence 4.0.**

From: Shamseer L, Moher D, Clarke M, Ghersi D, Liberati A, Petticrew M, Shekelle P, Stewart L, PRISMA-P Group. Preferred reporting items for systematic review and meta-analysis protocols (PRISMA-P) 2015: elaboration and explanation. BMJ. 2015 Jan 2;349(jan02 1):g7647.

COSMIN Taxonomy Measurement property definitions [1]

Term			
Domain	**Measurement property**	**Aspect of a measurement property**	**Definitions**
Reliability			The degree to which the measurement is free from measurement error
Reliability(extended definition)			The extent to which scores for patients who have not changed are the same for repeated measurement under several conditions: for example, using different sets of items from the same HR-PROs(internal consistency), over time (testeretest) by different persons on the same occasion (interrater) or by the same persons (i.e., raters or responders) on different occasions (intrarater)
	Internal consistency		The degree of the interrelatedness among the items
	Reliability		The proportion of the total variance in the measurements which is because of ‘‘true’’[a] differences among patients
Validity			The degree to which an HR-PRO instrument measures the construct(s)it purports to measure
	Content validity		The degree to which the content of an HR-PRO instrument is an adequate reflection of the construct to be measured
		Face validity	The degree to which (the items of) an HR-PRO instrument indeed looks as though they are an adequate reflection of the construct to be measured
	Construct validity		The degree to which the scores of an HR-PRO instrument are consistent with hypotheses (for instance

			with regard to internal relationships, relationships to scores of other instruments, or differences between relevant groups) based on the assumption that the HR-PRO instrument validly measures the construct to be measured
		Structural validity	The degree to which the scores of an HR-PRO instrument are an adequate reflection of the dimensionality of the construct to be measured
		Hypotheses testing	Idem construct validity
		Cross-cultural validity	The degree to which the performance of the items on a translated or culturally adapted HR-PRO instrument are an adequate reflection of the performance of the items of the original version of the HR-PRO instrument
	Criterion validity		The degree to which the scores of an HR-PRO instrument are an adequate reflection of a ‘‘gold standard’’
Responsiveness			The ability of an HR-PRO instrument to detect change over time in the construct to be measured
	Responsiveness		Idem responsiveness
Interpretability [b]			The degree to which one can assign qualitative meaning d that is, clinical or commonly understood connotations to an instrument’s quantitative scores or change in scores.

a The word ‘‘true’’ must be seen in the context of the CTT, which states that any observation is composed of two components true score and error associated with the observation. ‘‘True’’ is the average score that would be obtained if the scale were given an infinite number of times. It refers only to the consistency of the score and not to its accuracy

b Interpretability is not considered a measurement property but an important characteristic of a measurement instrument.

Criteria for good measurement properties [2]

Measurement property	Rating	Criteria
Structural validity	+	**CTT:**CFA: CFI or TLI or comparable measure > 0.95 OR RMSEA < 0.06 OR SRMR < 0.08a **IRT/Rasch:** No violation of unidimensionality [b]: CFI or TLI or comparable measure > 0.95 OR RMSEA < 0.06 OR SRMR < 0.08 *AND* no violation of local independence: residual correlations among the items after controlling for the dominant factor < 0.20 OR Q3's < 0.37 *AND* no violation of monotonicity: adequate looking graphs OR item scalability > 0.30 *AND* adequate model fit IRT: $\chi 2 > 0.001$ Rasch: infit and outfit mean squares ≥ 0.5 and ≤ 1.5 OR Z-standardized values > −2 and < 2
	?	CTT: not all information for '+' reported IRT/Rasch: model fit not reported
	−	Criteria for '+' not met
Internal consistency	+	At least low evidence for sufficient structural validity AND Cronbach's alpha(s) ≥ 0.70 for each unidimensional scale or subscale
	?	Criteria for "At least low evidence [c] for sufficient structural validity [d]" not met
	−	At least low evidence[c] for sufficient structural validity[d] AND Cronbach's alpha(s) < 0.70 for each unidimensional scale or subscale[e]
Reliability	+	ICC or weighted Kappa ≥ 0.70
	?	ICC or weighted Kappa not reported
	−	ICC or weighted Kappa < 0.70
Measurement error	+	SDC or LoA < MIC[d]
	?	MIC not defined
	−	SDC or LoA > MIC[d]
Hypotheses testing for construct validity	+	The result is in accordance with the hypothesis [f]
	?	No hypothesis defined (by the review team)
	−	The result is not in accordance with the hypothesis f
Cross-cultural validity\measurement invariance	+	No important differences found between group factors (such as age, gender, language) in multiple group factor analysis OR no important DIF for group

		factors (McFadden's R2 < 0.02)
	?	No multiple group factor analysis OR DIF analysis performed
	–	Important differences between group factors OR DIF was found
Criterion validity	+	Correlation with gold standard ≥ 0.70 OR AUC ≥ 0.70
	?	Not all information for '+' reported
	–	Correlation with gold standard < 0.70 OR AUC < 0.70
Responsiveness	+	The result is in accordance with the hypothesis [f] OR AUC ≥ 0.70
	?	No hypothesis defined (by the review team)
	–	The result is not in accordance with the hypothesis [f] OR AUC < 0.70

AUC area under the curve, *CFA* confirmatory factor analysis, *CFI* comparative fit index, *CTT* classical test theory, *DIF* differential item functioning,
ICC intraclass correlation coefficient, *IRT* item response theory, *LoA* limits of agreement, *MIC* minimal important change, *RMSEA* root mean square error of approximation, *SEM* standard error of measurement, *SDC* smallest detectable change, *SRMR* standardized root mean
residuals, *TLI* Tucker–Lewis index
"+" = sufficient
"–" = insufficient
"?" = indeterminate
a To rate the quality of the summary score, the factor structures should be equal across studies
b Unidimensionality refers to a factor analysis per subscale, while structural validity refers to a factor analysis of a (multidimensional) patient reported
outcome measure
c As defined by grading the evidence according to the GRADE approach
d This evidence may come from different studies
e The criteria 'Cronbach alpha < 0.95' was deleted, as this is relevant in the development phase of a PROM and not when evaluating an existing
PROM
f The results of all studies should be taken together and it should then be decided if 75% of the results are in accordance with the hypotheses

1. Mokkink, L.B., et al., The COSMIN study reached international consensus on taxonomy, terminology, and definitions of measurement properties for health-related patient-reported outcomes. *J Clin Epidemiol*, 2010. **63**(7):737-45.
2. Prinsen, C.A.C., et al., COSMIN guideline for systematic reviews of patient-reported outcome measures. *Quality of Life Research*, 2018. **27**(5):1147-57.

Supplementary File 3: Example of search strategy

Stage 1

1. scoliosis.mp.
2. exp Scoliosis/
3. Idiopathic scoliosis.mp.
4. exp Spinal Curvatures/
5. Adolescen$.mp.
6. exp Adolescent/
7. Physical functioning.mp.
8. exp Physical Functional Performance/
9. Functional activity.mp.
10. independence.mp.
11. Functional independence.mp.
12. exp Health Status/
13. exp performance status/
14. exp Health Behavior/
15. exp Movement/
16. mobility.mp.
17. Functional limitation.mp.
18. Activity limitation.mp.
19. exp Motor Activity/
20. Recovery of function/
21. (Recover$ adj5 function$).tw.
22. exp Motor Skills/
23. exp Disability Evaluation/
24. exp Disabled Persons/
25. exp physical examination/
26. exp "Activities of Daily Living"/
27. ((daily or domestic or house or home) adj5 (activit$ or task$ or skill$ or chore$)).t
28. (Activities of daily living or adl$ or eadl$ or iadl$).tw.
29. exp Self Care/
30. ((self or personal) adj5 (Care or manage$)).tw.
31. (Dressing or feeding or eating or toilet$ or bathing or mobil$ or driving or public transport$).tw.
32. exp Lifting/
33. Bending.mp.
34. exp sitting/

35. exp Walking/
36. exp Walking Speed/
37. exp Postural Balance/
38. Standing balance.mp.
39. exp Hand Strength/ or Grip strength.mp.
40. 1- 4/OR
41. 5 OR 6
42. 7-39/OR
43. 40 and 41 and 42
44. Limit 43 to humans

Stage 2

45. Name of the Identified outcome measure
46. validity.mp
47. exp validation studies/
48. reliability.mp
49. exp reproducibility of results/
50. interpretability.mp
51. internal consistency.mp
52. exp sensitivity and Specificity/
53. clinical sensitivity.mp
54. exp psychometrics/
55. responsiveness.mp
56. exp Evaluation studies/
57. measurement error.mp
58. measurement properties.mp
59. 46-58/OR
60. 1 AND 59
61. Limit 60 to humans

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Outcome Measures Evaluating Physical Functioning and their Measurement Properties in Adolescent Idiopathic Scoliosis: Protocol for a Systematic Review

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Outcome Measures Evaluating Physical Functioning and their Measurement Properties in Adolescent Idiopathic Scoliosis: Protocol for a Systematic Review

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ABSTRACT

Introduction Physical functioning (PF) is the ability to carry out physical activity of daily living. It is an important outcome that provide meaningful evaluation of individuals' life. PF can be assessed using Patient-Reported outcome measures, Performance-Based Outcome Measures or Body Structure and Function Measure. Measures need to be valid, reliable and responsive to change to evaluate effects of an intervention. Adolescent Idiopathic Scoliosis (AIS) is the most common deformity among the paediatric population. It causes significant impact on individuals' life. In the absence of existing evidence, this systematic review will appraise evidence on measurement properties of PF tools in individuals with AIS.

Methods/analysis A protocol for systematic review and meta-analysis informed by Cochrane guidelines and, reported in line with Preferred Reporting Items for Systematic Reviews and Meta-Analysis-P. MEDLINE, PsycINFO, EMBASE, CINHAL, SPORTdiscus, Web of science and PubMed will be searched in two stages (inception until December 2019). Search one will include all studies that assessed PF in participants with AIS without any limitations. Search terms will be Scoliosis, Adolescent and PF related terms. Search two will include studies which investigated instrument measurement properties in the same population for measures identified in search one. Two reviewers will independently perform study selection, data extraction, risk of bias, and overall quality assessment. The Consensus-based Standards for the selection of Health Measurement Instruments risk of bias and a modified Grading of Recommendations, Assessment, Development and Evaluation guidelines will be used. Meta-analysis will be conducted if possible, or the evidence will be synthesised and summarized for each outcome measure and according to specific measurement properties.

Ethics and dissemination: This review will provide recommendations for practice and future research, considering psychometric properties of outcome measures of PF in AIS. The results of this study will be disseminated through a peer-reviewed publication and conference presentation.

Keywords: Adolescent Idiopathic Scoliosis, measurement properties, physical function, systematic review

ARTICLE SUMMARY

Strengths and Limitations of this Study

1. This review synthesises evidence of patient-reported, performance-based or, body structure and function outcome measures of physical functioning, for use in practice or research involving individuals with AIS.
2. The search strategy of this review comprises two stages, to ensure that all measures of physical functioning of adolescent idiopathic scoliosis are included.
3. This study will employ rigorous methods and using COnsensus-based Standards for the selection of health Measurement Instruments risk of bias tool and modified GRADE.
4. This review is limited to studies of the English language that assess measurement properties among adolescents with idiopathic scoliosis.

INTRODUCTION

Scoliosis is a complex three-dimensional deformity of the spine, including lateral curvature and rotation of the vertebrae, [1] and characterised by a curve angle ≥10°. [2] There are two main types of scoliosis, idiopathic and non-idiopathic with the latter arising from congenital, neuromuscular or mesenchymal causes. [3] While the aetiology of idiopathic scoliosis remains unknown; genetic, hormonal, and mechanical factors are involved. [4] Adolescent Idiopathic Scoliosis (AIS) often develops between 10 and 16 years of age and represents ~85% of cases. [5] AIS is the most common spinal deformity among the pediatric population, with a prevalence ranging from 2% to 3%. [6] Nearly 80% of those affected presents with a curvature of the thoracic or thoracolumbar/lumbar region.[3] Whilst males and females are equally affected, females are reported to be at 10 times greater risk of curve progression. [1]

A number of health-related problems are reported among individuals with AIS including; lower quality of life, [7] back pain, [8] pulmonary dysfunction, [9] stress, [10] mental health disorders. [11] A major component of health status and health-related quality of life is physical functioning, [12] which can be used to identify individuals at risk of disability and to predict health and social care use. [13, 14] Accordingly, physical functioning is included in the Core Outcome Set (COS) for use within clinical trials for many musculoskeletal conditions, [12, 15, 16] including adolescents with spinal deformity.[16] Where the COS study includes all types of spinal deformity, there is a now need for a more specific systematic review of physical functioning outcome measures for this unique population subset. Limitations in physical functioning are reported by individuals with AIS, e.g. walking, moving around, and maintaining body position. [7, 17] Additionally, pain is often reported in individuals with AIS which may cause functional limitations. [8, 18, 19]

Physical functioning can be assessed with Patient-Reported Outcome Measures (PROMs), Performance-Based Outcome Measures (PBOMs) [20] or a measure of body structure and function e.g. Radiographs using Cobb method . [21] It can give an indication about dysfunctions in structure or organs but fail to fully capture functional limitations. [21] The most widely used PROM for assessment of the quality of life as well as physical functioning of individuals with spinal deformity is the Scoliosis Research Society (SRS) questionnaire, [22] and variants of such. [23-26] The SRS is mostly used among surgically treated individuals with AIS, [22, 25, 26] but may not be applicable to those treated conservatively. [26] Although relevant, PROMs are influenced by patients' perception of their abilities to perform activities and lack sensitivity to change. [21] Measures such as PBOMs have the potential to provide unbiased and reproducible assessments of physical functioning during the performance of activities of daily living, [21, 27] such as walking speed, trunk endurance testing. [21] Within individuals

with AIS, little is known about the available PBOMs for evaluating physical functioning. The SRS-22r questionnaire is the gold standard outcome measure of quality of life, which include physical functioning items as recommended by the recent COS study for adolescents and young adults with spinal deformity. [16] However, the SRS-22r fails to fully capture important aspects of physical functioning for individuals with AIS e.g. self-care and mobility. [7] This study included all forms of spinal deformities, the heterogeneity limits applicability to individuals with AIS as a discrete population.

Adequate measurement properties of outcome measures are important to avoid the risk of bias and ensure accuracy in the evaluation of test results.[28] The COnsensus-based Standards for the selection of health Measurement INstruments, (COSMIN) group developed a taxonomy of measurement properties to improve the selection of outcome measures. [29] Three main domains identified reliability, validity and responsiveness with further subgrouping.[28] A systematic review is needed to evaluate measures of assessment and, their measurement properties, of physical functioning for individuals with AIS. Review findings will inform clinicians and researchers on the best available tools for assessment of physical functioning in AIS. Furthermore, findings will inform future research drawing on a range of measures of physical functioning to investigate health status in AIS.

Objective

To identify outcome measures used to assess physical functioning in individuals with AIS. A secondary aim is to evaluate the measurement properties of physical functioning outcome measures in AIS.

METHODS

This protocol has been informed by experts in musculoskeletal orthopaedics including a consultant spine surgeon, musculoskeletal rehabilitation experts including physiotherapists and individuals with review, measurement properties and research experience. It has been designed in line with the COSMIN methodology for systematic reviews of Patient-Reported Outcome Measures [30] and is reported in line with the Preferred Reporting Items for Systematic Reviews and Meta-Analysis-P (PRISMA-P) [31] The search for this systematic review will be conducted in two parts. Stage one to identify studies used an outcome measure to evaluate physical functioning in individuals with AIS. This search will allow a list of outcome measures to be generated. Stage 2 will identify studies, which evaluated measurement properties of physical functioning outcome measure identified in the search one.

Stage one: Outcome measure

Eligibility criteria

Study design

All study designs including; randomised clinical trials, cohort, observational studies and case studies will be included to identify all outcome measure of physical functioning being used with individuals with AIS. No limitation on language or location will be applied at this stage.

Participants

Participants aged between the age of 10 years to 18 years of age, with a diagnosis of idiopathic scoliosis and ≥10° Cobb angle will be considered. No restrictions will be applied to the curve severity, evaluation settings, and type of treatment as well as to the language or country.

Outcome

Any study that includes assessments of the physical functioning of AIS using specific outcome measure will be included. Physical functioning defined according to the Core Outcome Measures in Effectiveness Trials (COMET) taxonomy, [15] as any physical activities of daily living such as the ability to walk, independence, self-care, performance status, disability index. [15, 32] The outcome measures being defined as any one of the following:

1. Patient-Reported Outcome Measures (PROMs) in the form of questionnaires or scales designed for AIS to evaluate physical functioning or if it is included as a sub-scale within a questionnaire.
2. Performance-based outcome measures (PBOMs); a measure of physical functioning by clinician while the individual is performing a functional task e.g walking, and/or
3. Body structure and function measure which means any dysfunction in a specific body part or system which may limits function, such as range of motion. [21]

Stage two: measurement properties

Eligibility criteria

Study design

Any study that has evaluated one or more measurement properties of the identified outcome measures in the first search will be eligible. Only full-text studies available in English will be included. Conference abstracts will be excluded due to the inability to effectively evaluate the quality of the study.

Participants

Participants aged between the age of 10 to 18 years of age, with a diagnosis of idiopathic scoliosis and ≥10° Cobb angle will be eligible. In studies with mixed cohorts, >50% of participants should be individuals with AIS for the study to be included. Authors of studies will be contacted in case of missing information about number of participants with AIS. Studies without original participant data (e.g. systematic review) will be excluded.

Outcome

Outcome of interest is the measurement properties: Reliability including (Internal consistency, test–retest, inter-rater and intra-rater), measurement error, validity (Content validity, Structural validity or Criterion validity), hypothesis testing, responsiveness [29] of the outcome measures identified in the search one will be eligible. Studies that provide indirect evidence on the measurement properties (by testing an alternative test against an outcome measure of interest, studies in which the outcome measure is used to measure an outcome) will be excluded. Also, studies that provide normative data will be excluded.

Information sources

The search strategy has been developed using medical subject headings, and relevant text words relating to AIS, physical functioning, outcome measures and measurement properties using the adapted search filter. [33] The electronic search of databases will be conducted including MEDLINE (1946- November 2019), PsycINFO (1967- December 2019), EMBASE (1974- December 2019), CINHAL (1937- December 2019), SPORTdiscus (1800- December 2019), Web of Science (1900- December 2019) and PubMed (1997- December 2019). No language limitations will be applied in the first search; however, the second search will be limited to the full-text article in English. A hand search in the key

journals including Spine, The Spine Journal, Spine Deformity, Scoliosis and Spinal Disorders and European Spine Journal as well as contacting relevant leading researchers in the field. Further, searching for the Grey literature, including British National bibliography for report literature, open-Grey, dissertation abstracts and Electronic Thesis Online Service (EThOS) will be conducted. Web of Science database will be searched for conference proceedings for the last 5 years.

Search strategy

Following discussions with the team and specialist librarian, the search be completed by one researcher (SA). Then, the potential studies will be independently assessed for eligibility by two researchers (SA, EB). To ensure that all relevant data are included, no limitations will be applied. Initial search terms will be developed for MEDLINE and then adapted with relevant syntax and subject headings for other databases. Recommended search filters specifically designed for retrieving articles on measurement properties will be used where appropriate.[33] An example of the search strategy of both stages is available as an online supplementary file 1.

Data management

Search records will be imported into Endnote Version X9 (Clarivate Analytics). Using Endnote, the abstracts and full texts will be stored. The duplicates will be identified through the Endnote software and exact duplicates will be removed.

Selection Process

A standardised eligibility assessment will be performed by two independent reviewers (SA, EB). All studies identified by the search strategy will assessed based on title/abstract for eligibility. If there is insufficient information to include/exclude study, full text will be retrieved and then, screened for eligibility. The Study selection (included and excluded studies) with the reasons for exclusion, will be summarised in a PRISMA flow diagram. [31] Articles will be included if both reviewers agreed that the eligibility criteria were met. Any disagreement will be first discussed and the third reviewer (NH) will mediate situations of disagreement. At each assessment stage, agreement between reviewers will be estimated with percentage of agreement and the Kappa statistic using SPSS for Windows statistical software package (IBM SPSS Statistics Version 25).

Data collection process

Two reviewers (SA, EB) will independently extract the data of eligible studies. A bespoke data extraction form will be used and piloted on 3 studies. Any disagreement between reviewers will be

mediated through discussion with a third reviewer (NH) if needed. If information is not clear or unavailable in the studies, corresponding authors will be contacted to request further details. A second and final reminder will then each be sent 2 weeks apart.

Data items

The data that will be extracted from each study at each stage is summarised in Table 1. In the case of missing data, the authors of the study will be contacted.

Table 1: Summary of items to be extracted from included studies

Study & Participants Characteristics	Reference, Year, Country, Design of Study, Age, Gender, Sample Size (used in the analysis), Curve Size, Curve Type, Type of Intervention (Bracing, Physiotherapy, Exercise, Or Surgery)
Outcome measure	**PROM:** Name of outcome measure, means of scores (standard deviation), mode of administration, recall period, sub-scale, number of items, response option, response rate, missing items, Setting, target population, scoring, original language, available translation
	PBOM: Name of outcome measure, equipment needed, number of assessments, outcome (e.g. time needed, ability/disability), setting, scoring
	Body structure and function measure: Name of outcome measure, equipment needed, mode of administration, setting, scoring, outcome (e.g. time needed, ability/disability)
Measurement properties	**Validity:** Name of outcome measure, type of validity, descriptive statistics, missing value, comparator outcome or predictor outcome, statistics method, confidence interval, validation results
	Reliability: Name of outcome measure, type of reliability, descriptive statistic, time interval, reliability coefficient, measurement error
	Responsiveness: Name of outcome measure, Method of testing: *Hypothesis testing, Distribution based method* (ES, SRM and MDC), time to follow-up. *Anchor-based methods* (MIC or MCIC or MID).
	Interpretability: : Name of outcome measure, distribution of score in the study population, percentage of missing items, floor and ceiling effects, scores and change scores available for relevant (sub)groups , MIC Or MID, information on response shift
	Feasibility: Patient's comprehensibility, Clinician's comprehensibility, Type and ease of administration, Length of instrument, Completion time, Patient's required mental and physical ability level, Ease of standardization, Ease of score calculation, Copyright, Cost of an instrument, Required equipment, Availability in different settings, Regulatory agency's requirement for approval

ES Effects Size, MCIC Minimal Clinically Important Change, MDC Minimal Detectable Change, MIC Minimal Important Change, MID Minimal Important Difference, SRM Standardized Response Mean

Outcomes and prioritisation

The gold standard and the primary outcome measure for evaluation of body structure and function (e.g. spinal curvature), is the radiographs using the Cobb method [2]. However, no primary PROM or PBOM of physical functioning for individuals with AIS, can be identified for this review.

Risk of bias in individual studies

The COSMIN checklist for assessment of risk of bias and methodological quality in individual studies will be used. [28] It was revised and specifically designed for use in systematic reviews of PROMs to evaluate studies on measurement properties. [34] The methodological quality of each study for each measurement property will be assessed separately. [35] The items for measurement property in the standards box will be rated as either very good, adequate, doubtful or inadequate quality. [35] Then, the overall methodological quality of the measurement property will be rated based on "the worst score counts principle" i.e. that the overall quality of the study for a specific measurement property is based on the lowest rating of any items in the standards' box. [35] The result of each item and overall rating will be reported in the final results. The COSMIN group recommend researchers to adapt the checklist to other measures (i.e. PBOMs, body structure and function measure) since it was originally developed for PROMs. [35] Two independent reviewers (SA, EB) will assess the risk of bias for all included studies. Any disagreement will be resolved through discussion, and if no agreement is reached, a third reviewer (NH) will be consulted. The agreement between reviewers will be estimated with percentage agreement and the Kappa statistic using SPSS for Windows statistical software package (IBM SPSS Statistics Version 25) and will be reported in the final results.

Data synthesis

The COSMIN guidelines for systematic reviews will be followed for the synthesis of the results. [36] Characteristics of the outcome measures, sample, measurement properties results, information about interpretability and feasibility of the scores of the included outcome measures will be presented in overview tables for each outcome measure. [36] Each measurement property for each study per tool will be rated against the updated criteria for good measurement properties as either sufficient (+), insufficient (–), or indeterminate (?). [36] The result of rating of measurement property and its methodological quality rating will be added to the overview table. [36] Then, the evidence will be pooled or summarized per measurement property per tool, with the overall result will be rated against the criteria for good measurement properties, and the quality of the evidence will be graded using

the a modified Grading of Recommendations Assessment, Development and Evaluation (GRADE) approach. [36]

The results on measurement properties from different studies will be pooled in a meta-analysis if there is sufficient clinical and methodological homogeneity. [36] The data will be statistically pooled when: (1): Individuals with AIS displayed similar characteristics in terms of curve severity, intervention. (2): similar base-line score (3): Same time interval (4): Same statistical parameters. [36] If inconsistent results of measurement properties were presents due to different subgroups (i.e. mild and sever curve), the consistent results will be separately summarized per subgroup. [36] Pooled estimate of measurement properties will be obtained by calculating weighted means and 95% confidence interval. [36] If deemed not possible to pool the results, a qualitative synthesis will be conducted e.g. the percentage of confirmed hypotheses for construct validity will be provided. [36] The pooled or summarized evidence will be rated as sufficient when at least 75% of the results met the criteria. [36] For example, for structural validity, "at least 75% of the confirmatory factor analysis studies should found the same factor structure". [36]

The recommendation of an outcome measure will be depending on the measurement properties, as well as interpretability and feasibility results. [36] The tool should have sufficient content validity and at least low quality evidence for sufficient internal consistency to be recommended for use and the results of this tool is trustworthy. [36]

Confidence in cumulative evidence

The overall quality and strength of evidence will be assessed for pooled or summarised result for each measurement property per outcome measure per category by two reviewers, independently. Using a modified (GRADE) approach. [36] The GRADE approach uses five factors to determine the quality of the evidence: risk of bias (quality of the studies), inconsistency (of the results of the studies), indirectness (evidence comes from different populations, interventions or outcomes than the ones of interest in the review), imprecision (wide confidence intervals), and publication bias (negative results are less often published).[37] For evaluating measurement properties in systematic reviews of PROMs, only four factors will be assessed as recommended by COSMIN group, while the fifth factor (publication bias) was removed as there is no registry exists for measurement properties. [36]

Discussion

The AIS affects the physical functioning of individuals with AIS [7]. Measurement of its impact is important in research and clinical practice. Physical functioning considered as an important outcome domain in health-related quality of life. [12] It can be used to predict future disability as well as health and social care use.[13] Variety of tools are available for assessment of physical functioning, ranging from patient-reported to performance-based measures. However, it is essential to confirm the psychometric properties of these tools to be recommended for clinical use. The COS study for 'all spine deformities' identified the SRS-22r as the recommended PROM for assessment for physical functioning among young adults with spinal deformities. [16] However, there is still a need for a more specific review that evaluate the quality of all outcome measures used in the assessment of physical functioning in AIS including patient-reported, and performance-based as well as measures of body structure and function. This systematic review will retrieve all tools that have been used to assess physical functioning among individuals with AIS. Then, it will evaluate and synthesis the quality of studies that report psychometric properties of physical functioning outcome measures in AIS. This review will provide a comprehensive assessment of current evidence which benefits practitioners, patients as well as policymakers. Limitations of this review are a focus on individuals with AIS specifically, so recommendations cannot be generalised to other forms of scoliosis.

Patient and public involvement

The study question and systematic review protocol were informed following discussion at a patient and public involvement meeting at the Centre of Precision Rehabilitation for Spinal Pain at the University of Birmingham. The group were individuals with different musculoskeletal and spinal complaints. They actively contributed to research question and the need for systematic review. Since no patient data is needed, patients will not be involved in data collection or analysis. However, the results of the study will be shared at public engagement events.

Implications of this study

AIS is a complex deformity of the spine and causes a significant impact on physical activities of individuals' daily living such as walking and maintaining body position. [7, 17] In consequence, the quality of life is affected. Physical functioning gives an indication about the current health status and identifies people at risk of disability. [12, 13] Therefore, physical functioning is considered as one of the outcomes that should be assessed and reported in clinical trials of musculoskeletal conditions. [15] A systematic review is needed to evaluate current practice in assessment of physical functioning among individuals with AIS. The results of this review will inform clinicians and researchers on the best

available tools for assessment of physical functioning in AIS. This review could provide a research agenda that may highlight the gap in the literature around physical functioning measure and their measurement properties among individuals with AIS.

Declarations

Ethics and Dissemination

No ethics approval is required for this systematic review. The results of this systematic review will be disseminated through peer-reviewed journals as well as international and national conferences presentation. The publications will be split into different publications according to the volume of data. Each category of outcome measures will be published in a separate article.

Author Contributions

All authors conceptualized and designed the protocol. SA is a PhD student and NH (lead supervisor), AR and DF are supervisors and AG spinal surgeon. SA drafted the initial manuscript with NH, AR DF, and AG providing guidance on design, topic, methodology and analyses. All authors reviewed and commented on each draft of the protocol. All authors have approved and contributed to the final manuscript.

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Competing interests

None declared.

Patient consent for publication

Not required.

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Supplementary File 1

Example of search strategy

Stage 1

1. scoliosis.mp.
2. exp Scoliosis/
3. exp Spinal Curvatures/
4. Adolescen$.mp.
5. exp Adolescent/
6. Physical functioning.mp.
7. exp Physical Functional Performance/
8. Functional activity.mp.
9. independence.mp.
10. Functional independence.mp.
11. exp Health Status/
12. exp performance status/
13. exp Health Behavior/
14. exp Movement/
15. mobility.mp.
16. Functional limitation.mp.
17. Activity limitation.mp.
18. exp Motor Activity/
19. Recovery of function/
20. (Recover$ adj5 function$).tw.
21. exp Motor Skills/
22. exp Disability Evaluation/
23. exp Disabled Persons/
24. exp physical examination/
25. exp "Activities of Daily Living"/
26. ((daily or domestic or house or home) adj5 (activit$ or task$ or skill$ or chore$)).t
27. (Activities of daily living or adl$ or eadl$ or iadl$).tw.
28. exp Self Care/
29. ((self or personal) adj5 (Care or manage$)).tw.
30. (Dressing or feeding or eating or toilet$ or bathing or mobil$ or driving or public transport$).tw.
31. exp Lifting/
32. Bending.mp.

33. exp sitting/
34. exp Walking/
35. exp Walking Speed/
36. exp Postural Balance/
37. Standing balance.mp.
38. exp Hand Strength/ or Grip strength.mp.
39. 1- 3/OR
40. 4 OR 5
41. 6-38/OR
42. 39 and 40 and 41
43. Limit 42 to humans

Stage 2

44. Name of the Identified outcome measure
45. validity.mp
46. exp validation studies/
47. reliability.mp
48. exp reproducibility of results/
49. interpretability.mp
50. internal consistency.mp
51. exp sensitivity and Specificity/
52. clinical sensitivity.mp
53. exp psychometrics/
54. responsiveness.mp
55. exp Evaluation studies/
56. measurement error.mp
57. measurement properties.mp
58. 46-58/OR
59. 44 AND 58
60. Limit 59 to humans

PRISMA-P (Preferred Reporting Items for Systematic review and Meta-Analysis Protocols) 2015 checklist: recommended items to address in a systematic review protocol*

Section and topic	Item No	Checklist item	Reported on Page
ADMINISTRATIVE INFORMATION			
Title:			
Identification	1a	Identify the report as a protocol of a systematic review	Page 1 line 2
Update	1b	If the protocol is for an update of a previous systematic review, identify as such	NA
Registration	2	If registered, provide the name of the registry (such as PROSPERO) and registration number	Page 1 line 21
Authors:			
Contact	3a	Provide name, institutional affiliation, e-mail address of all protocol authors; provide physical mailing address of corresponding author	Page 1 line 4-19
Contributions	3b	Describe contributions of protocol authors and identify the guarantor of the review	Page13 line 350-354
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	NA
Support:			
Sources	5a	Indicate sources of financial or other support for the review	NA
Sponsor	5b	Provide name for the review funder and/or sponsor	Page13 line 356-357
Role of sponsor or funder	5c	Describe roles of funder(s), sponsor(s), and/or institution(s), if any, in developing the protocol	NA
INTRODUCTION			
Rationale	6	Describe the rationale for the review in the context of what is already known	Page 4 line 104-108 Page 5 line 130-134
Objectives	7	Provide an explicit statement of the question(s) the review will address with reference to participants, interventions, comparators, and outcomes (PICO)	Page 5 line 136-137
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	Page 6-7

Information sources	9	Describe all intended information sources (such as electronic databases, contact with study authors, trial registers or other grey literature sources) with planned dates of coverage	Page 7 line 199-210
Search strategy	10	Present draft of search strategy to be used for at least one electronic database, including planned limits, such that it could be repeated	Page 8 line 212-218 Supplementary file 1
Study records:			
Data management	11a	Describe the mechanism(s) that will be used to manage records and data throughout the review	Page 8 line 220-222
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)	Page 8 line 224-232
Data collection process	11c	Describe planned method of extracting data from reports (such as piloting forms, done independently, in duplicate), any processes for obtaining and confirming data from investigators	Page 8,9 line 234-238
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	Page 9, Table 1
Outcomes and prioritization	13	List and define all outcomes for which data will be sought, including prioritization of main and additional outcomes, with rationale	Page 10 line 250-252
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	Page 10 line 274-261
Data synthesis	15a	Describe criteria under which study data will be quantitatively synthesised	Page 10, 11line 275-277
	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 I_2, Kendall's τ)	Page 11 line 279-281
	15c	Describe any proposed additional analyses (such as sensitivity or subgroup analyses, meta-regression)	Page 11 line 277-279
	15d	If quantitative synthesis is not appropriate, describe the type of summary planned	Page 11 line 281-282
Meta-bias(es)	16	Specify any planned assessment of meta-bias(es) (such as publication bias across studies, selective reporting within studies)	NA
Confidence in cumulative evidence	17	Describe how the strength of the body of evidence will be assessed (such as GRADE)	Page 11 line 291-299

*** It is strongly recommended that this checklist be read in conjunction with the PRISMA-P Explanation and Elaboration (cite when available) for important clarification on the items. Amendments to a review protocol should be tracked and dated. The copyright for PRISMA-P (including checklist) is held by the PRISMA-P Group and is distributed under a Creative Commons Attribution Licence 4.0.**

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Outcome Measures Evaluating Physical Functioning and their Measurement Properties in Adolescent Idiopathic Scoliosis: Protocol for a Systematic Review

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Keywords:	Adolescent Idiopathic Scoliosis, Measurement properties, Physical function, Scoliosis < ORTHOPAEDIC & TRAUMA SURGERY

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Outcome Measures Evaluating Physical Functioning and their Measurement Properties in Adolescent Idiopathic Scoliosis: Protocol for a Systematic Review

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ABSTRACT

Introduction Physical functioning (PF) is the ability to carry out physical activity of daily living. It is an important outcome that provide meaningful evaluation of individuals' life. PF can be assessed using Patient-Reported outcome measures, Performance-Based Outcome Measures or Body Structure and Function Measure. Measures need to be valid, reliable and responsive to change to evaluate effects of an intervention. Adolescent Idiopathic Scoliosis (AIS) is the most common deformity among the paediatric population. It causes significant impact on individuals' life. This systematic review will appraise evidence on the measurement properties of PF tools in individuals with AIS.

Methods/analysis A protocol for systematic review and meta-analysis informed by Cochrane guidelines is reported in line with Preferred Reporting Items for Systematic Reviews and Meta-Analysis-P. MEDLINE, PsycINFO, EMBASE, CINHAL, SPORTdiscus, Web of science and PubMed will be searched in two stages, from inception until December 2019. Search one will inventory all studies that assessed PF in participants with AIS, without any limitations. The Search terms will be Scoliosis, Adolescent and PF related terms. Search two will include studies which investigated instrument measurement properties in the same population for measures identified in search one. Two reviewers will independently perform study selection, data extraction, risk of bias, and overall quality assessment. The Consensus-based Standards for the selection of Health Measurement Instruments risk of bias and a modified Grading of Recommendations, Assessment, Development and Evaluation guidelines will be used. Meta-analysis will be conducted if possible, or the evidence will be synthesised and summarized per measurement property per outcome measure per measurement type.

Ethics and dissemination: This review will provide recommendations for practice and future research, considering psychometric properties of outcome measures of PF in AIS. The results of this study will be disseminated through a peer-reviewed publication and conference presentation.

Prospero registration number: CRD42019142335

Keywords: Adolescent Idiopathic Scoliosis, measurement properties, physical function, systematic review

ARTICLE SUMMARY

Strengths and Limitations of this Study

1. This review will synthesise evidence of patient-reported, performance-based or, body structure and function outcome measures of physical functioning, for use in practice or research involving individuals with AIS.
2. The search strategy of this review comprises two stages. The first stage will retrieve all studies that assessed physical functioning in individuals with AIS, while the Second stage will retrieve studies that investigated measurement properties of the instrument identified in the first search.
3. This study will employ rigorous methods and uses COnsensus-based Standards for the selection of health Measurement Instruments risk of bias tool and modified Grading of Recommendations, Assessment, Development and Evaluation approach.
4. This review will be limited to studies of the English language that assess measurement properties among adolescents with idiopathic scoliosis.

INTRODUCTION

Scoliosis is a complex three-dimensional deformity of the spine, including lateral curvature and rotation of the vertebrae, [1] and characterised by a curve angle ≥10°. [2] There are two main types of scoliosis, idiopathic and non-idiopathic with the latter arising from congenital, neuromuscular or mesenchymal causes. [3] While the aetiology of idiopathic scoliosis remains unknown; genetic, hormonal, and mechanical factors are involved. [4] Adolescent Idiopathic Scoliosis (AIS) often develops between 10 and 16 years of age and represents ~85% of cases. [5] AIS is the most common spinal deformity among the pediatric population, with a prevalence ranging from 2% to 3%. [6] Nearly 80% of those affected presents with a curvature of the thoracic or thoracolumbar/lumbar region.[3] Whilst males and females are equally affected, females are reported to be at 10 times greater risk of curve progression. [1]

A number of health-related problems are reported among individuals with AIS including; lower quality of life, [7] back pain, [8] pulmonary dysfunction, [9] stress, [10] mental health disorders. [11] A major component of health status and health-related quality of life is physical functioning, [12] which can be used to identify individuals at risk of disability and to predict health and social care use. [13, 14] Accordingly, physical functioning is included in the Core Outcome Set (COS) for use within clinical trials for many musculoskeletal conditions, [12, 15, 16] including adolescents with spinal deformity.[16] Where the COS study includes all types of spinal deformity, there is a now need for a more specific systematic review of physical functioning outcome measures for this unique population subset. Limitations in physical functioning are reported by individuals with AIS, e.g. walking, moving around, and maintaining body position. [7, 17] Additionally, pain is often reported in individuals with AIS which may cause functional limitations. [8, 18, 19]

Physical functioning can be assessed with Patient-Reported Outcome Measures (PROMs), Performance-Based Outcome Measures (PBOMs) [20] or a measure of body structure and function. The most widely used PROM for assessment of the quality of life as well as physical functioning of individuals with spinal deformity is the Scoliosis Research Society (SRS) questionnaire, [21] and its variants. [22-25] The SRS is mostly used among surgically treated individuals with AIS, [21, 24, 25] but may not be applicable to those treated conservatively. [25] Although relevant, PROMs should be used cautiously; as it influenced by patients' perception of their abilities to perform activities and lack sensitivity to change. [26] Measures such as PBOMs have the potential to provide unbiased and reproducible assessments of physical functioning during the performance of activities of daily living, [26, 27] such as walking speed and trunk endurance testing. [26] Within individuals with AIS, little is known about the available PBOMs for evaluating physical functioning. The body structure and

function measures such as radiographs can give an indication about dysfunctions in structure but fail to fully capture functional limitations. [26]

The SRS-22r questionnaire is the gold standard outcome measure of quality of life, which include physical functioning items as recommended by the recent COS study for adolescents and young adults with spinal deformity. [16] However, the SRS-22r fails to fully capture important aspects of physical functioning for individuals with AIS e.g. self-care and mobility. [7] The COS study included all forms of spinal deformities, the heterogeneity limits applicability to individuals with AIS as a discrete population.

Adequate measurement properties of outcome measures are important to avoid the risk of bias and ensure accuracy in the evaluation of test results.[28] The COnsensus-based Standards for the selection of health Measurement INstruments, (COSMIN) group developed a taxonomy of measurement properties to improve the selection of outcome measures. [29] Three main domains identified reliability, validity and responsiveness with further subgrouping.[28] A systematic review is needed to evaluate the measurement properties, of physical functioning outcome measure for individuals with AIS. Review findings will inform clinicians and researchers on the best available tools for the assessment of physical functioning in AIS. Furthermore, findings will inform future research drawing on a range of measures of physical functioning to investigate health status in AIS.

Objective

To identify outcome measures used to assess physical functioning in individuals with AIS. A secondary aim is to evaluate the measurement properties of physical functioning outcome measures in AIS.

METHODS

This protocol has been informed by experts in musculoskeletal orthopaedics including a consultant spine surgeon, musculoskeletal rehabilitation experts including physiotherapists and individuals with review, measurement properties and research experience. It has been designed in line with the COSMIN methodology for systematic reviews of Patient-Reported Outcome Measures [30] and is reported in line with the Preferred Reporting Items for Systematic Reviews and Meta-Analysis-P (PRISMA-P) [31] The search for this systematic review will be conducted in two parts. Stage one to identify studies used an outcome measure to evaluate physical functioning in individuals with AIS. This search will allow a list of outcome measures to be generated. Stage 2 will identify studies, which

evaluated measurement properties of physical functioning outcome measure identified in the first search.

Stage one: Inventory of outcome measure

Eligibility criteria

Study design

All study designs including; randomised clinical trials, cohort, observational studies and case studies will be included to identify all outcome measure of physical functioning being used with individuals with AIS. No limitation on language or location will be applied at this stage.

Participants

Participants aged between the age of 10 years to 18 years of age, with a diagnosis of idiopathic scoliosis and ≥10° Cobb angle will be considered. No restrictions will be applied to the curve severity, evaluation settings, and the type of treatment.

Outcome

Any study that includes assessments of the physical functioning of AIS using a specific outcome measure will be included. Physical functioning is defined according to the Core Outcome Measures in Effectiveness Trials (COMET) taxonomy, [15] as any physical activities of daily living such as the ability to walk, independence, self-care, performance status, disability index. [15, 32] The outcome measures are defined as any one of the following:

1. Patient-Reported Outcome Measures (PROMs) in the form of questionnaires or scales designed for AIS to evaluate physical functioning or if it is included as a sub-scale within a questionnaire.
2. Performance-based outcome measures (PBOMs); a measure of physical functioning by clinician while the individual is performing a functional task e.g walking, and/or
3. Body structure and function measures which means any dysfunction in a specific body part or system which may limits function, such as range of motion. [26]

Search strategy

A comprehensive, systematic and reproducible search strategy will be completed by one reviewer (SA). Databases will be searched to identify studies that assessed physical functioning among individuals with AIS. To ensure that all relevant studies are included, the type of the outcome measure will not be specified at this stage (Figure 1). Initial search terms will be developed for MEDLINE and then adapted with relevant syntax and subject headings for the other databases. An example of the search strategy of stage 1 is available as an online supplementary file 1. As a result of this search, a list of outcome measure for physical functioning used in AIS will be generated. Then, the outcome measures will be classified i.e. PROM, PBOM, or measure of body structure and function. The list will then be used to perform the search in stage 2.

Stage two: measurement properties

Eligibility criteria

Study design

Any study that has evaluated one or more measurement properties of the identified outcome measures in the first search will be eligible. Only full-text studies available in English will be included. Conference abstracts will be excluded due to the inability to effectively evaluate the quality of the study.

Participants

Participants aged between the age of 10 to 18 years of age, with a diagnosis of idiopathic scoliosis and ≥10° Cobb angle will be eligible. In studies with mixed cohorts, >50% of participants should be individuals with AIS for the study to be included. Authors of studies will be contacted in case of missing information about number of participants with AIS. Studies without original participant data (e.g. systematic review) will be excluded.

Outcome

The outcomes of interest are the measurement properties: Reliability including (Internal consistency, test–retest, inter-rater and intra-rater), measurement error, validity (Content validity, Structural validity or Criterion validity), hypothesis testing, and responsiveness [29] of the outcome measures identified in the search one will be eligible. Studies that provide indirect evidence on the measurement properties (by testing an alternative test against an outcome measure of interest, studies in which the

outcome measure is used to measure an outcome) will be excluded. Also, studies that only provide normative data will be excluded.

Search strategy

Using the list of outcome measures determined from the search in stage one, one reviewer (SA) will conduct the search. Each category of outcome measure will be searched separately. The search terms will be consisting of the name of the outcome measure/s, the AIS and the measurement properties (Figure 1). The recommended search filters specifically designed for retrieving articles on measurement properties will be adapted and used at this stage [33]. An example of the search strategy of stage 2 is available as an online supplementary file 1.

Information sources

The electronic search of databases will be conducted including MEDLINE (1946- November 2019), PsycINFO (1967- December 2019), and EMBASE (1974- December 2019) through OVID interface, CINHAL (1937- December 2019) and SPORTdiscus (1800- December 2019) through EBSCO interface, Web of Science (1900- December 2019) and PubMed (1997- December 2019). No language limitations will be applied in the first search; however, the second search will be limited to the full-text article in English. The Web of Science database will be searched for conference proceedings for the last 5 years for the first search only. A hand search in the key journals including Spine, The Spine Journal, Spine Deformity, Scoliosis and Spinal Disorders and European Spine Journal as well as contacting relevant leading researchers in the field. Further, searching for the Grey literature, including British National bibliography for report literature, open-Grey, dissertation abstracts and Electronic Thesis Online Service (EThOS) will be conducted.

Data management

Search records will be imported into Endnote Version X9 (Clarivate Analytics). Using Endnote, the abstracts and full texts will be stored. The duplicates will be identified through the Endnote software and exact duplicates will be removed.

Selection Process

A standardised eligibility assessment will be performed by two independent reviewers (SA, EB). All studies identified by the search strategy will assessed based on title/abstract for eligibility. If there is insufficient information to include/exclude study, full text will be retrieved and then, screened for

eligibility. The study selection (included and excluded studies) with the reasons for exclusion, will be summarised in a PRISMA flow diagram. [31] Articles will be included if both reviewers agreed that the eligibility criteria were met. Any disagreement will be first discussed and the third reviewer (NH) will mediate situations of disagreement. At each assessment stage, agreement between reviewers will be estimated with percentage of agreement and the Kappa statistic using SPSS for Windows statistical software package (IBM SPSS Statistics Version 25).

Data collection process

Two reviewers (SA, EB) will independently extract the data of eligible studies. A bespoke data extraction form will be used and piloted on 3 studies. Any disagreement between reviewers will be mediated through discussion with a third reviewer (NH) if needed. If information is not clear or unavailable in the studies, corresponding authors will be contacted to request further details. A second and final reminder will then each be sent 2 weeks apart.

Data items

The data that will be extracted from each study at each stage is summarised in Table 1. In the case of missing data, the authors of the study will be contacted.

Table 1: Summary of items to be extracted from included studies

Study & Participants Characteristics	Reference, Year, Country, Design of Study, Age, Gender, Sample Size (used in the analysis), Curve Size, Curve Type, Type of Intervention (Bracing, Physiotherapy, Exercise, Or Surgery)
Outcome measure	**PROM:** Name of outcome measure, means of scores (standard deviation), mode of administration, recall period, sub-scale, number of items, response option, response rate, missing items, setting, target population, scoring, original language, available translation
	PBOM: Name of outcome measure, equipment needed, number of assessments, outcome (e.g. time needed, ability/disability), setting, scoring
	Body structure and function measure: Name of outcome measure, equipment needed, mode of administration, setting, scoring, outcome (e.g. time needed, ability/disability)
Measurement properties	**Validity:** Name of outcome measure, type of validity, descriptive statistics, missing value, comparator outcome or predictor outcome, hypothesis, statistics method, confidence interval, validation results
	Reliability: Name of outcome measure, type of reliability, descriptive statistic, time interval, reliability coefficient, measurement error

	Responsiveness: Name of outcome measure, Method of testing: *Hypothesis testing, Distribution based method* (ES, SRM and MDC), hypothesis, time to follow-up. *Anchor-based methods* (MIC or MCIC or MID), anchor/s.
	Interpretability: Name of outcome measure, distribution of score in the study population, percentage of missing items, floor and ceiling effects, scores and change scores available for relevant (sub)groups**,** MIC Or MID, information on response shift
	Feasibility: Patient's comprehensibility, Clinician's comprehensibility, Type and ease of administration, Length of instrument, Completion time, Patient's required mental and physical ability level, Ease of standardization, Ease of score calculation, Copyright, Cost of an instrument, Required equipment, Availability in different settings, Regulatory agency's requirement for approval
ES Effects Size, MCIC Minimal Clinically Important Change, MDC Minimal Detectable Change, MIC Minimal Important Change, MID Minimal Important Difference, SRM Standardized Response Mean	

Outcomes and prioritisation

The gold standard and the primary outcome measure for evaluation of body structure and function (e.g. spinal curvature), is the radiographs using the Cobb method [2]. However, no primary PROM or PBOM of physical functioning for individuals with AIS, can be identified for this review.

Risk of bias in individual studies

The COSMIN checklist for assessment of risk of bias and methodological quality in individual studies will be used. [28] It was revised and specifically designed for use in systematic reviews of PROMs to evaluate studies on measurement properties. [34] The methodological quality of each study for each measurement property will be assessed separately. [30] The items for each measurement property in the relevant standards box will be rated as either very good, adequate, doubtful or inadequate quality. [30] Then, the overall methodological quality of the measurement property will be rated based on "the worst score counts principle" i.e. that the overall quality of the study for a specific measurement property is based on the lowest rating of any items in the standards' box. [30] The result of each item and overall rating will be reported in the final results. The COSMIN group recommend researchers to adapt the checklist to other measures (i.e. PBOMs, body structure and function measure) since it was originally developed for PROMs. [30] Two independent reviewers (SA, EB) will assess the risk of bias for all included studies. Any disagreement will be resolved through discussion, and if no agreement is reached, a third reviewer (NH) will be consulted. The agreement between reviewers will be estimated with percentage agreement and the Kappa statistic using SPSS for Windows statistical software package (IBM SPSS Statistics Version 25) and will be reported in the final results.

Data synthesis

The COSMIN guidelines for systematic reviews will be followed for the synthesis of the results. [30] Characteristics of the outcome measures, sample, measurement properties results, information about interpretability and feasibility of the scores of the included outcome measures will be presented in overview tables for each outcome measure. [30] Each measurement property for each study per tool will be rated against the updated criteria for good measurement properties as either sufficient (+), insufficient (–), or indeterminate (?).[30] The result of rating of measurement property and its methodological quality rating will be added to the overview table. [30] Then, the evidence will be pooled or summarized per measurement property per tool, with the overall result will be rated against the criteria for good measurement properties, and the quality of the evidence will be graded using a modified Grading of Recommendations Assessment, Development and Evaluation (GRADE) approach. [30]

The results on measurement properties from different studies will be pooled in a meta-analysis if there is enough clinical and methodological homogeneity. The data will be statistically pooled when: (1): Individuals with AIS displayed similar characteristics in terms of curve severity, intervention. (2): similar base-line score (3): Same time interval (4): Same statistical parameters. If inconsistent results of measurement properties were presents due to different subgroups (i.e. mild and sever curve), the consistent results will be separately summarized per subgroup. [30] Pooled estimate of measurement properties will be obtained by calculating weighted means and 95% confidence interval. If deemed not possible to pool the results, a qualitative synthesis will be conducted e.g. the percentage of confirmed hypotheses for construct validity will be provided. [30] The pooled or summarized evidence will be rated as sufficient when at least 75% of the results met the criteria. For example, for structural validity, “at least 75% of the confirmatory factor analysis studies should found the same factor structure”. [30]

The recommendation of an outcome measure will be depending on the measurement properties, as well as interpretability and feasibility results. The tool should have sufficient content validity and at least low-quality evidence for sufficient internal consistency to be recommended for use and the results of this tool is trustworthy. [30]

Confidence in cumulative evidence

Two independent reviewers will assess the quality and strength of evidence for the pooled or summarised result. Using the modified (GRADE) approach, each measurement property per outcome

measure in each category will be evaluated. The GRADE approach uses five factors to determine the quality of the evidence: risk of bias (quality of the studies), inconsistency (of the results of the studies), indirectness (evidence comes from different populations, interventions or outcomes than the ones of interest in the review), imprecision (wide confidence intervals), and publication bias (negative results are less often published).[35] For evaluating measurement properties in systematic reviews of PROMs, only four factors will be assessed as recommended by COSMIN group, while the fifth factor (publication bias) will be removed as there is no registry exists for measurement properties.

Discussion

Physical functioning is considered as an important outcome domain in health-related quality of life. [12] It can be used to predict future disability as well as health and social care use.[13] Individuals with AIS reported a limitation in their physical functioning [7]. Thus, measurement of its impact is important in research and clinical practice. Numerous of tools are available for the assessment of physical functioning, ranging from patient-reported to performance-based measures. However, it is essential to confirm the psychometric properties of these tools before recommending for clinical use. The COS study for 'all spine deformities' identified the SRS-22r as the recommended PROM for assessment for physical functioning among young adults with spinal deformities. [16] However, there is still a need for a more specific review that evaluate the quality of all outcome measures used in the assessment of physical functioning in AIS including patient-reported, and performance-based as well as measures of body structure and function. This systematic review will retrieve all tools that have been used to assess physical functioning among individuals with AIS. Then, it will evaluate and synthesise the quality of studies that report psychometric properties of physical functioning outcome measures in AIS. This review will provide a comprehensive assessment of current evidence which may benefit: (1) health practitioners in selection of the most suitable tools to assess physical functioning in AIS (2) patients who need a good outcome measures that reflect their actual health status (3) researchers and policy maker who can use the recommend measures in designing research trials and defining the COS for individuals with AIS, which in turn will improve health assessment and patient care. Limitations of this review are a focus on individuals with AIS specifically, so recommendations cannot be generalised to other forms of scoliosis.

Patient and public involvement

The study question and systematic review protocol were informed following discussion at a patient and public involvement meeting at the Centre of Precision Rehabilitation for Spinal Pain at the University of Birmingham. The group consisted of individuals with different musculoskeletal and spinal complaints. They actively contributed to research question and to establish the need for systematic review. Since no patient data is needed, patients will not be involved in data collection or analysis. However, the results of the study will be shared at public engagement events.

Implications of this study

AIS is a complex deformity of the spine and causes a significant impact on physical activities of individuals' daily living such as walking and maintaining body position. [7, 17] In consequence, the quality of life is affected. Physical functioning gives an indication about the current health status and identifies people at risk of disability. [12, 13] Therefore, physical functioning is considered as one of the outcomes that should be assessed and reported in clinical trials of musculoskeletal conditions. [15] A systematic review is needed to evaluate current practice in the assessment of physical functioning among individuals with AIS. The results of this review will inform clinicians and researchers on the best available tools for assessment of physical functioning in AIS. This review could provide a research agenda that may highlight the gap in the literature around physical functioning measure and their measurement properties among individuals with AIS.

Declarations

Ethics and Dissemination

No ethics approval is required for this systematic review. The results of this systematic review will be disseminated through peer-reviewed journals as well as international and national conferences presentation. The publications will be split into different publications according to the volume of data. Each category of outcome measures will be published in a separate article.

Author Contributions

All authors conceptualized and designed the protocol. SA is a PhD student and NH (lead supervisor), AR and DF are supervisors and AG is a spinal surgeon. SA drafted the initial manuscript with NH, AR DF, and AG providing guidance on design, topic, methodology and analyses. All authors reviewed and

commented on each draft of the protocol. All authors have approved and contributed to the final manuscript.

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Competing interests

None declared.

Patient consent for publication

Not required.

Figure 1:

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Figure Captions

Figure 1: Flow diagram of search strategy (Search One and Two) and selection process. PROM=Patient Reported Outcome Measure; PBOM= Performance-Based Outcome Measure.

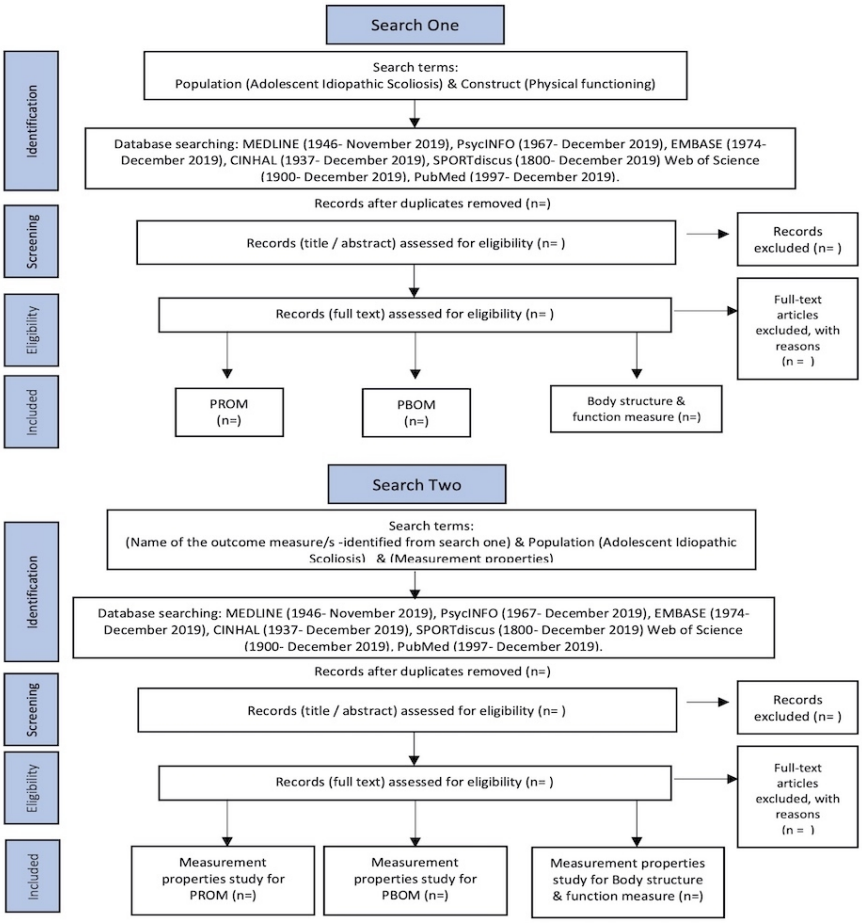

Figure 1: Flow diagram of search strategy (Search One and Two) and selection process. PROM=Patient Reported Outcome Measure; PBOM= Performance-Based Outcome Measure.

90x90mm (300 x 300 DPI)

Supplementary File 1

Example of search strategy (MEDLINE)

Stage 1

1. scoliosis.mp.
2. exp Scoliosis/
3. exp Spinal Curvatures/
4. Adolescen$.mp.
5. exp Adolescent/
6. Physical functioning.mp.
7. exp Physical Functional Performance/
8. Functional activity.mp.
9. independence.mp.
10. Functional independence.mp.
11. exp Health Status/
12. exp performance status/
13. exp Health Behavior/
14. exp Movement/
15. mobility.mp.
16. Functional limitation.mp.
17. Activity limitation.mp.
18. exp Motor Activity/
19. Recovery of function/
20. (Recover$ adj5 function$).tw.
21. exp Motor Skills/
22. exp Disability Evaluation/
23. exp Disabled Persons/
24. exp physical examination/
25. exp "Activities of Daily Living"/
26. ((daily or domestic or house or home) adj5 (activit$ or task$ or skill$ or chore$)).t
27. (Activities of daily living or adl$ or eadl$ or iadl$).tw.
28. exp Self Care/
29. ((self or personal) adj5 (Care or manage$)).tw.
30. (Dressing or feeding or eating or toilet$ or bathing or mobil$ or driving or public transport$).tw.
31. exp Lifting/
32. Bending.mp.

33. exp sitting/
34. exp Walking/
35. exp Walking Speed/
36. exp Postural Balance/
37. Standing balance.mp.
38. exp Hand Strength/ or Grip strength.mp.
39. 1- 3/OR
40. 4 OR 5
41. 6-38/OR
42. 39 and 40 and 41
43. Limit 42 to humans

Stage 2

44. Name of the Identified outcome measure/s
45. scoliosis.mp.
46. exp Scoliosis/
47. exp Spinal Curvatures/
48. Adolescen$.mp.
49. exp Adolescent/
50. validity.mp
51. exp validation studies/
52. reliability.mp
53. exp reproducibility of results/
54. interpretability.mp
55. internal consistency.mp
56. exp sensitivity and Specificity/
57. clinical sensitivity.mp
58. exp psychometrics/
59. responsiveness.mp
60. exp Evaluation studies/
61. measurement error.mp
62. measurement properties.mp
63. 45-47/OR
64. 48-49/OR
65. 50-62/OR
66. 44 and 63 and 64 and 65
67. Limit 66 to humans

PRISMA-P (Preferred Reporting Items for Systematic review and Meta-Analysis Protocols) 2015 checklist: recommended items to address in a systematic review protocol*

Section and topic	Item No	Checklist item	Reported on Page
ADMINISTRATIVE INFORMATION			
Title:			
Identification	1a	Identify the report as a protocol of a systematic review	Page 1 line 2
Update	1b	If the protocol is for an update of a previous systematic review, identify as such	NA
Registration	2	If registered, provide the name of the registry (such as PROSPERO) and registration number	Page 1 line 21
Authors:			
Contact	3a	Provide name, institutional affiliation, e-mail address of all protocol authors; provide physical mailing address of corresponding author	Page 1 line 4-19
Contributions	3b	Describe contributions of protocol authors and identify the guarantor of the review	Page13 line 350-354
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	NA
Support:			
Sources	5a	Indicate sources of financial or other support for the review	NA
Sponsor	5b	Provide name for the review funder and/or sponsor	Page13 line 356-357
Role of sponsor or funder	5c	Describe roles of funder(s), sponsor(s), and/or institution(s), if any, in developing the protocol	NA
INTRODUCTION			
Rationale	6	Describe the rationale for the review in the context of what is already known	Page 4 line 104-108 Page 5 line 130-134
Objectives	7	Provide an explicit statement of the question(s) the review will address with reference to participants, interventions, comparators, and outcomes (PICO)	Page 5 line 136-137
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	Page 6-7

Information sources	9	Describe all intended information sources (such as electronic databases, contact with study authors, trial registers or other grey literature sources) with planned dates of coverage	Page 7 line 199-210
Search strategy	10	Present draft of search strategy to be used for at least one electronic database, including planned limits, such that it could be repeated	Page 8 line 212-218 Supplementary file 1
Study records:			
Data management	11a	Describe the mechanism(s) that will be used to manage records and data throughout the review	Page 8 line 220-222
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)	Page 8 line 224-232
Data collection process	11c	Describe planned method of extracting data from reports (such as piloting forms, done independently, in duplicate), any processes for obtaining and confirming data from investigators	Page 8,9 line 234-238
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	Page 9, Table 1
Outcomes and prioritization	13	List and define all outcomes for which data will be sought, including prioritization of main and additional outcomes, with rationale	Page 10 line 250-252
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	Page 10 line 274-261
Data synthesis	15a	Describe criteria under which study data will be quantitatively synthesised	Page 10, 11line 275-277
	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 I_2, Kendall's τ)	Page 11 line 279-281
	15c	Describe any proposed additional analyses (such as sensitivity or subgroup analyses, meta-regression)	Page 11 line 277-279
	15d	If quantitative synthesis is not appropriate, describe the type of summary planned	Page 11 line 281-282
Meta-bias(es)	16	Specify any planned assessment of meta-bias(es) (such as publication bias across studies, selective reporting within studies)	NA
Confidence in cumulative evidence	17	Describe how the strength of the body of evidence will be assessed (such as GRADE)	Page 11 line 291-299

*** It is strongly recommended that this checklist be read in conjunction with the PRISMA-P Explanation and Elaboration (cite when available) for important clarification on the items. Amendments to a review protocol should be tracked and dated. The copyright for PRISMA-P (including checklist) is held by the PRISMA-P Group and is distributed under a Creative Commons Attribution Licence 4.0.**

From: Shamseer L, Moher D, Clarke M, Ghersi D, Liberati A, Petticrew M, Shekelle P, Stewart L, PRISMA-P Group. Preferred reporting items for systematic review and meta-analysis protocols (PRISMA-P) 2015: elaboration and explanation. BMJ. 2015 Jan 2;349(jan02 1):g7647.

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Outcome Measures Evaluating Physical Functioning and their Measurement Properties in Adolescent Idiopathic Scoliosis: Protocol for a Systematic Review

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Secondary Subject Heading:	Patient-centred medicine, Paediatrics, Rehabilitation medicine
Keywords:	Adolescent Idiopathic Scoliosis, Measurement properties, Physical function, Scoliosis < ORTHOPAEDIC & TRAUMA SURGERY

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Outcome Measures Evaluating Physical Functioning and their Measurement Properties in Adolescent Idiopathic Scoliosis: Protocol for a Systematic Review

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ABSTRACT

Introduction Physical functioning (PF) is the ability to carry out physical activity of daily living. It is an important outcome that provide meaningful evaluation of individuals' life. PF can be assessed using Patient-Reported outcome measures, Performance-Based Outcome Measures or Body Structure and Function Measure. Measures need to be valid, reliable and responsive to change to evaluate effects of an intervention. Adolescent Idiopathic Scoliosis (AIS) is the most common deformity among the paediatric population. It causes significant impact on individuals' life. This systematic review will appraise evidence on the measurement properties of PF tools in individuals with AIS.

Methods/analysis A protocol for systematic review and meta-analysis informed by Cochrane guidelines is reported in line with Preferred Reporting Items for Systematic Reviews and Meta-Analysis-P. MEDLINE, PsycINFO, EMBASE, CINAHL, SPORTdiscus, Web of science and PubMed will be searched in two stages, from inception until December 2019. Search one will inventory all studies that assessed PF in participants with AIS, without any limitations. The Search terms will be Scoliosis, Adolescent and PF related terms. Search two will include studies which investigated instrument measurement properties in the same population for measures identified in search one. Two reviewers will independently perform study selection, data extraction, risk of bias, and overall quality assessment. The Consensus-based Standards for the selection of Health Measurement Instruments risk of bias and a modified Grading of Recommendations, Assessment, Development and Evaluation guidelines will be used. Meta-analysis will be conducted if possible, or the evidence will be synthesised and summarized per measurement property per outcome measure per measurement type.

Ethics and dissemination: This review will provide recommendations for practice and future research, considering psychometric properties of outcome measures of PF in AIS. The results of this study will be disseminated through a peer-reviewed publication and conference presentation.

Prospero registration number: CRD42019142335

Keywords: Adolescent Idiopathic Scoliosis, measurement properties, physical function, systematic review

ARTICLE SUMMARY

Strengths and Limitations of this Study

1. This review will synthesise evidence of patient-reported, performance-based or, body structure and function outcome measures of physical functioning, for use in practice or research involving individuals with AIS.
2. The search strategy of this review comprises two stages. The first stage will retrieve all studies that assessed physical functioning in individuals with AIS, while the Second stage will retrieve studies that investigated measurement properties of the instrument identified in the first search.
3. This study will employ rigorous methods and uses COnsensus-based Standards for the selection of health Measurement Instruments risk of bias tool and modified Grading of Recommendations, Assessment, Development and Evaluation approach.
4. This review will be limited to studies of the English language that assess measurement properties among adolescents with idiopathic scoliosis.

INTRODUCTION

Scoliosis is a complex three-dimensional deformity of the spine, including lateral curvature and rotation of the vertebrae, [1] and characterised by a curve angle ≥10°. [2] There are two main types of scoliosis, idiopathic and non-idiopathic with the latter arising from congenital, neuromuscular or mesenchymal causes. [3] While the aetiology of idiopathic scoliosis remains unknown; genetic, hormonal, and mechanical factors are involved. [4] Adolescent Idiopathic Scoliosis (AIS) often develops between 10 and 16 years of age and represents ~85% of cases. [5] AIS is the most common spinal deformity among the pediatric population, with a prevalence ranging from 2% to 3%. [6] Nearly 80% of those affected presents with a curvature of the thoracic or thoracolumbar/lumbar region.[3] Whilst males and females are equally affected, females are reported to be at 10 times greater risk of curve progression. [1]

A number of health-related problems are reported among individuals with AIS including; lower quality of life, [7] back pain, [8] pulmonary dysfunction, [9] stress, [10] mental health disorders. [11] A major component of health status and health-related quality of life is physical functioning, [12] which can be used to identify individuals at risk of disability and to predict health and social care use. [13, 14] Accordingly, physical functioning is included in the Core Outcome Set (COS) for use within clinical trials for many musculoskeletal conditions, [12, 15, 16] including adolescents with spinal deformity.[16] Where the COS study includes all types of spinal deformity, there is a now need for a more specific systematic review of physical functioning outcome measures for this unique population subset. Limitations in physical functioning are reported by individuals with AIS, e.g. walking, moving around, and maintaining body position. [7, 17] Additionally, pain is often reported in individuals with AIS which may cause functional limitations. [8, 18, 19]

Physical functioning can be assessed with Patient-Reported Outcome Measures (PROMs), Performance-Based Outcome Measures (PBOMs) [20] or a measure of body structure and function. The most widely used PROM for assessment of the quality of life as well as physical functioning of individuals with spinal deformity is the Scoliosis Research Society (SRS) questionnaire, [21] and its variants. [22-25] The SRS is mostly used among surgically treated individuals with AIS, [21, 24, 25] but may not be applicable to those treated conservatively. [25] Although relevant, PROMs should be used cautiously; as it influenced by patients' perception of their abilities to perform activities and lack sensitivity to change. [26] Measures such as PBOMs have the potential to provide unbiased and reproducible assessments of physical functioning during the performance of activities of daily living, [26, 27] such as walking speed and trunk endurance testing. [26] Within individuals with AIS, little is known about the available PBOMs for evaluating physical functioning. The body structure and

function measures such as radiographs can give an indication about dysfunctions in structure but fail to fully capture functional limitations. [26]

The SRS-22r questionnaire is the reference standard outcome measure of quality of life, which include physical functioning items as recommended by the recent COS study for adolescents and young adults with spinal deformity. [16] However, the SRS-22r fails to fully capture important aspects of physical functioning for individuals with AIS e.g. self-care and mobility. [7] The COS study included all forms of spinal deformities, the heterogeneity limits applicability to individuals with AIS as a discrete population.

Adequate measurement properties of outcome measures are important to avoid the risk of bias and ensure accuracy in the evaluation of test results.[28] The COnsensus-based Standards for the selection of health Measurement INstruments, (COSMIN) group developed a taxonomy of measurement properties to improve the selection of outcome measures. [29] Three main domains identified reliability, validity and responsiveness with further subgrouping.[28] A systematic review is needed to evaluate the measurement properties, of physical functioning outcome measure for individuals with AIS. Review findings will inform clinicians and researchers on the best available tools for the assessment of physical functioning in AIS. Furthermore, findings will inform future research drawing on a range of measures of physical functioning to investigate health status in AIS.

Objective

To identify outcome measures used to assess physical functioning in individuals with AIS. A secondary aim is to evaluate the measurement properties of physical functioning outcome measures in AIS.

METHODS

This protocol has been informed by experts in musculoskeletal orthopaedics including a consultant spine surgeon, musculoskeletal rehabilitation experts including physiotherapists and individuals with review, measurement properties and research experience. It has been designed in line with the COSMIN methodology for systematic reviews of Patient-Reported Outcome Measures [30] and is reported in line with the Preferred Reporting Items for Systematic Reviews and Meta-Analysis-P (PRISMA-P) [31] The search for this systematic review will be conducted in two parts. Stage one to identify studies used an outcome measure to evaluate physical functioning in individuals with AIS. This search will allow a list of outcome measures to be generated. Stage 2 will identify studies, which

evaluated measurement properties of physical functioning outcome measure identified in the first search.

Stage one: Inventory of outcome measure

Eligibility criteria

Study design

All study designs including; randomised clinical trials, cohort, observational studies and case studies will be included to identify all outcome measure of physical functioning being used with individuals with AIS. No limitation on language or location will be applied at this stage.

Participants

Participants aged between the age of 10 years to 18 years of age, with a diagnosis of idiopathic scoliosis and ≥10° Cobb angle will be considered. No restrictions will be applied to the curve severity, evaluation settings, and the type of treatment.

Outcome

Any study that includes assessments of the physical functioning of AIS using a specific outcome measure will be included. Physical functioning is defined according to the Core Outcome Measures in Effectiveness Trials (COMET) taxonomy, [15] as any physical activities of daily living such as the ability to walk, independence, self-care, performance status, disability index. [15, 32] The outcome measures are defined as any one of the following:

1. Patient-Reported Outcome Measures (PROMs) in the form of questionnaires or scales designed for AIS to evaluate physical functioning or if it is included as a sub-scale within a questionnaire.
2. Performance-based outcome measures (PBOMs); a measure of physical functioning by clinician while the individual is performing a functional task e.g walking, and/or
3. Body structure and function measures which means any dysfunction in a specific body part or system which may limits function, such as range of motion. [26]

Search strategy

A comprehensive, systematic and reproducible search strategy will be completed by one reviewer (SA). Databases will be searched to identify studies that assessed physical functioning among individuals with AIS. To ensure that all relevant studies are included, the type of the outcome measure will not be specified at this stage (Figure 1). Initial search terms will be developed for MEDLINE and then adapted with relevant syntax and subject headings for the other databases. An example of the search strategy of stage 1 is available as an online supplementary file 1. As a result of this search, a list of outcome measure for physical functioning used in AIS will be generated. Then, the outcome measures will be classified i.e. PROM, PBOM, or measure of body structure and function. The list will then be used to perform the search in stage 2.

Stage two: measurement properties

Eligibility criteria

Study design

Any study that has evaluated one or more measurement properties of the identified outcome measures in the first search will be eligible. Only full-text studies available in English will be included. Conference abstracts will be excluded due to the inability to effectively evaluate the quality of the study.

Participants

Participants aged between the age of 10 to 18 years of age, with a diagnosis of idiopathic scoliosis and ≥10° Cobb angle will be eligible. In studies with mixed cohorts, >50% of participants should be individuals with AIS for the study to be included. Authors of studies will be contacted in case of missing information about number of participants with AIS. Studies without original participant data (e.g. systematic review) will be excluded.

Outcome

The outcomes of interest are the measurement properties: Reliability including (Internal consistency, test–retest, inter-rater and intra-rater), measurement error, validity (Content validity, Structural validity or Criterion validity), hypothesis testing, and responsiveness [29] of the outcome measures identified in the search one will be eligible. Studies that provide indirect evidence on the measurement properties (by testing an alternative test against an outcome measure of interest, studies in which the

outcome measure is used to measure an outcome) will be excluded. Also, studies that only provide normative data will be excluded.

Search strategy

Using the list of outcome measures determined from the search in stage one, one reviewer (SA) will conduct the search. Each category of outcome measure will be searched separately. The search terms will be consisting of the name of the outcome measure/s, the AIS and the measurement properties (Figure 1). The recommended search filters specifically designed for retrieving articles on measurement properties will be adapted and used at this stage [33]. An example of the search strategy of stage 2 is available as an online supplementary file 1.

Information sources

The electronic search of databases will be conducted including MEDLINE (1946- November 2019), PsycINFO (1967- December 2019), and EMBASE (1974- December 2019) through OVID interface, CINAHL (1937- December 2019) and SPORTdiscus (1800- December 2019) through EBSCO interface, Web of Science (1900- December 2019) and PubMed (1997- December 2019). No language limitations will be applied in the first search; however, the second search will be limited to the full-text article in English. The Web of Science database will be searched for conference proceedings for the last 5 years for the first search only. A hand search in the key journals including Spine, The Spine Journal, Spine Deformity, Scoliosis and Spinal Disorders and European Spine Journal as well as contacting relevant leading researchers in the field. Further, searching for the Grey literature, including British National bibliography for report literature, open-Grey, dissertation abstracts and Electronic Thesis Online Service (EThOS) will be conducted.

Data management

Search records will be imported into Endnote Version X9 (Clarivate Analytics). Using Endnote, the abstracts and full texts will be stored. The duplicates will be identified through the Endnote software and exact duplicates will be removed.

Selection Process

A standardised eligibility assessment will be performed by two independent reviewers (SA, EB). All studies identified by the search strategy will assessed based on title/abstract for eligibility. If there is insufficient information to include/exclude study, full text will be retrieved and then, screened for

eligibility. The study selection (included and excluded studies) with the reasons for exclusion, will be summarised in a PRISMA flow diagram. [31] Articles will be included if both reviewers agreed that the eligibility criteria were met. Any disagreement will be first discussed and the third reviewer (NH) will mediate situations of disagreement. At each assessment stage, agreement between reviewers will be estimated with percentage of agreement and the Kappa statistic using SPSS for Windows statistical software package (IBM SPSS Statistics Version 25).

Data collection process

Two reviewers (SA, EB) will independently extract the data of eligible studies. A bespoke data extraction form will be used and piloted on 3 studies. Any disagreement between reviewers will be mediated through discussion with a third reviewer (NH) if needed. If information is not clear or unavailable in the studies, corresponding authors will be contacted to request further details. A second and final reminder will then each be sent 2 weeks apart.

Data items

The data that will be extracted from each study at each stage is summarised in Table 1. In the case of missing data, the authors of the study will be contacted.

Table 1: Summary of items to be extracted from included studies

Study & Participants Characteristics	Reference, Year, Country, Design of Study, Age, Gender, Sample Size (used in the analysis), Curve Size, Curve Type, Type of Intervention (Bracing, Physiotherapy, Exercise, Or Surgery)
Outcome measure	**PROM:** Name of outcome measure, means of scores (standard deviation), mode of administration, recall period, sub-scale, number of items, response option, response rate, missing items, setting, target population, scoring, original language, available translation
	PBOM: Name of outcome measure, equipment needed, number of assessments, outcome (e.g. time needed, ability/disability), setting, scoring
	Body structure and function measure: Name of outcome measure, equipment needed, mode of administration, setting, scoring, outcome (e.g. time needed, ability/disability)
Measurement properties	**Validity:** Name of outcome measure, type of validity, descriptive statistics, missing value, comparator outcome or predictor outcome, hypothesis, statistics method, confidence interval, validation results
	Reliability: Name of outcome measure, type of reliability, descriptive statistic, time interval, reliability coefficient, measurement error

	Responsiveness: Name of outcome measure, Method of testing: *Hypothesis testing, Distribution based method* (ES, SRM and MDC), hypothesis, time to follow-up. *Anchor-based methods* (MIC or MCIC or MID), anchor/s.
	Interpretability: Name of outcome measure, distribution of score in the study population, percentage of missing items, floor and ceiling effects, scores and change scores available for relevant (sub)groups**,** MIC Or MID, information on response shift
	Feasibility: Patient's comprehensibility, Clinician's comprehensibility, Type and ease of administration, Length of instrument, Completion time, Patient's required mental and physical ability level, Ease of standardization, Ease of score calculation, Copyright, Cost of an instrument, Required equipment, Availability in different settings, Regulatory agency's requirement for approval
ES Effects Size, MCIC Minimal Clinically Important Change, MDC Minimal Detectable Change, MIC Minimal Important Change, MID Minimal Important Difference, SRM Standardized Response Mean	

Outcomes and prioritisation

The gold standard and the primary outcome measure for evaluation of body structure and function (e.g. spinal curvature), is the radiographs using the Cobb method [2]. However, no primary PROM or PBOM of physical functioning for individuals with AIS, can be identified for this review.

Risk of bias in individual studies

The COSMIN checklist for assessment of risk of bias and methodological quality in individual studies will be used. [28] It was revised and specifically designed for use in systematic reviews of PROMs to evaluate studies on measurement properties. [34] The methodological quality of each study for each measurement property will be assessed separately. [30] The items for each measurement property in the relevant standards box will be rated as either very good, adequate, doubtful or inadequate quality. [30] Then, the overall methodological quality of the measurement property will be rated based on "the worst score counts principle" i.e. that the overall quality of the study for a specific measurement property is based on the lowest rating of any items in the standards' box. [30] The result of each item and overall rating will be reported in the final results. The COSMIN group recommend researchers to adapt the checklist to other measures (i.e. PBOMs, body structure and function measure) since it was originally developed for PROMs. [30] Two independent reviewers (SA, EB) will assess the risk of bias for all included studies. Any disagreement will be resolved through discussion, and if no agreement is reached, a third reviewer (NH) will be consulted. The agreement between reviewers will be estimated with percentage agreement and the Kappa statistic using SPSS for Windows statistical software package (IBM SPSS Statistics Version 25) and will be reported in the final results.

Data synthesis

The COSMIN guidelines for systematic reviews will be followed for the synthesis of the results. [30] Characteristics of the outcome measures, sample, measurement properties results, information about interpretability and feasibility of the scores of the included outcome measures will be presented in overview tables for each outcome measure. [30] Each measurement property for each study per tool will be rated against the updated criteria for good measurement properties as either sufficient (+), insufficient (–), or indeterminate (?).[30] The result of rating of measurement property and its methodological quality rating will be added to the overview table. [30] Then, the evidence will be pooled or summarized per measurement property per tool, with the overall result will be rated against the criteria for good measurement properties, and the quality of the evidence will be graded using a modified Grading of Recommendations Assessment, Development and Evaluation (GRADE) approach. [30]

The results on measurement properties from different studies will be pooled in a meta-analysis if there is enough clinical and methodological homogeneity. The data will be statistically pooled when: (1): Individuals with AIS displayed similar characteristics in terms of curve severity, intervention. (2): similar base-line score (3): Same time interval (4): Same statistical parameters. If inconsistent results of measurement properties were presents due to different subgroups (i.e. mild and sever curve), the consistent results will be separately summarized per subgroup. [30] Pooled estimate of measurement properties will be obtained by calculating weighted means and 95% confidence interval. If deemed not possible to pool the results, a qualitative synthesis will be conducted e.g. the percentage of confirmed hypotheses for construct validity will be provided. [30] The pooled or summarized evidence will be rated as sufficient when at least 75% of the results met the criteria. For example, for structural validity, “at least 75% of the confirmatory factor analysis studies should found the same factor structure”. [30]

The recommendation of an outcome measure will be depending on the measurement properties, as well as interpretability and feasibility results. The tool should have sufficient content validity and at least low-quality evidence for sufficient internal consistency to be recommended for use and the results of this tool is trustworthy. [30]

Confidence in cumulative evidence

Two independent reviewers will assess the quality and strength of evidence for the pooled or summarised result. Using the modified (GRADE) approach, each measurement property per outcome

measure in each category will be evaluated. The GRADE approach uses five factors to determine the quality of the evidence: risk of bias (quality of the studies), inconsistency (of the results of the studies), indirectness (evidence comes from different populations, interventions or outcomes than the ones of interest in the review), imprecision (wide confidence intervals), and publication bias (negative results are less often published).[35] For evaluating measurement properties in systematic reviews of PROMs, only four factors will be assessed as recommended by COSMIN group, while the fifth factor (publication bias) will be removed as there is no registry exists for measurement properties.

Discussion

Physical functioning is considered as an important outcome domain in health-related quality of life. [12] It can be used to predict future disability as well as health and social care use.[13] Individuals with AIS reported a limitation in their physical functioning [7]. Thus, measurement of its impact is important in research and clinical practice. Numerous of tools are available for the assessment of physical functioning, ranging from patient-reported to performance-based measures. However, it is essential to confirm the psychometric properties of these tools before recommending for clinical use. The COS study for 'all spine deformities' identified the SRS-22r as the recommended PROM for assessment for physical functioning among young adults with spinal deformities. [16] However, there is still a need for a more specific review that evaluate the quality of all outcome measures used in the assessment of physical functioning in AIS including patient-reported, and performance-based as well as measures of body structure and function. This systematic review will retrieve all tools that have been used to assess physical functioning among individuals with AIS. Then, it will evaluate and synthesise the quality of studies that report psychometric properties of physical functioning outcome measures in AIS. This review will provide a comprehensive assessment of current evidence which may benefit: (1) health practitioners in selection of the most suitable tools to assess physical functioning in AIS (2) patients who need a good outcome measures that reflect their actual health status (3) researchers and policy maker who can use the recommend measures in designing research trials and defining the COS for individuals with AIS, which in turn will improve health assessment and patient care. Limitations of this review are a focus on individuals with AIS specifically, so recommendations cannot be generalised to other forms of scoliosis.

Patient and public involvement

A study question and systematic review protocol were informed following discussion at a patient and public involvement meeting at the Centre of Precision Rehabilitation for Spinal Pain at the University of Birmingham. The group consisted of individuals with different musculoskeletal and spinal complaints. They actively contributed to research question and to establish the need for systematic review. Since no patient data is needed, patients will not be involved in data collection or analysis. However, the results of the study will be shared at public engagement events.

Implications of this study

AIS is a complex deformity of the spine and causes a significant impact on physical activities of individuals' daily living such as walking and maintaining body position. [7, 17] In consequence, the quality of life is affected. Physical functioning gives an indication about the current health status and identifies people at risk of disability. [12, 13] Therefore, physical functioning is considered as one of the outcomes that should be assessed and reported in clinical trials of musculoskeletal conditions. [15] A systematic review is needed to evaluate current practice in the assessment of physical functioning among individuals with AIS. The results of this review will inform clinicians and researchers on the best available tools for assessment of physical functioning in AIS. This review could provide a research agenda that may highlight the gap in the literature around physical functioning measure and their measurement properties among individuals with AIS.

Declarations

Ethics and Dissemination

No ethics approval is required for this systematic review. The results of this systematic review will be disseminated through peer-reviewed journals as well as international and national conferences presentation. The publications will be split into different publications according to the volume of data. Each category of outcome measures will be published in a separate article.

Author Contributions

All authors conceptualized and designed the protocol. SA is a PhD student and NH (lead supervisor), AR and DF are supervisors and AG is a spinal surgeon. SA drafted the initial manuscript with NH, AR DF, and AG providing guidance on design, topic, methodology and analyses. All authors reviewed and

commented on each draft of the protocol. All authors have approved and contributed to the final manuscript.

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Competing interests

None declared.

Patient consent for publication

Not required.

Figure Captions

Figure 1: Flow diagram of search strategy (Search One and Two) and selection process. PROM=Patient Reported Outcome Measure; PBOM= Performance-Based Outcome Measure.

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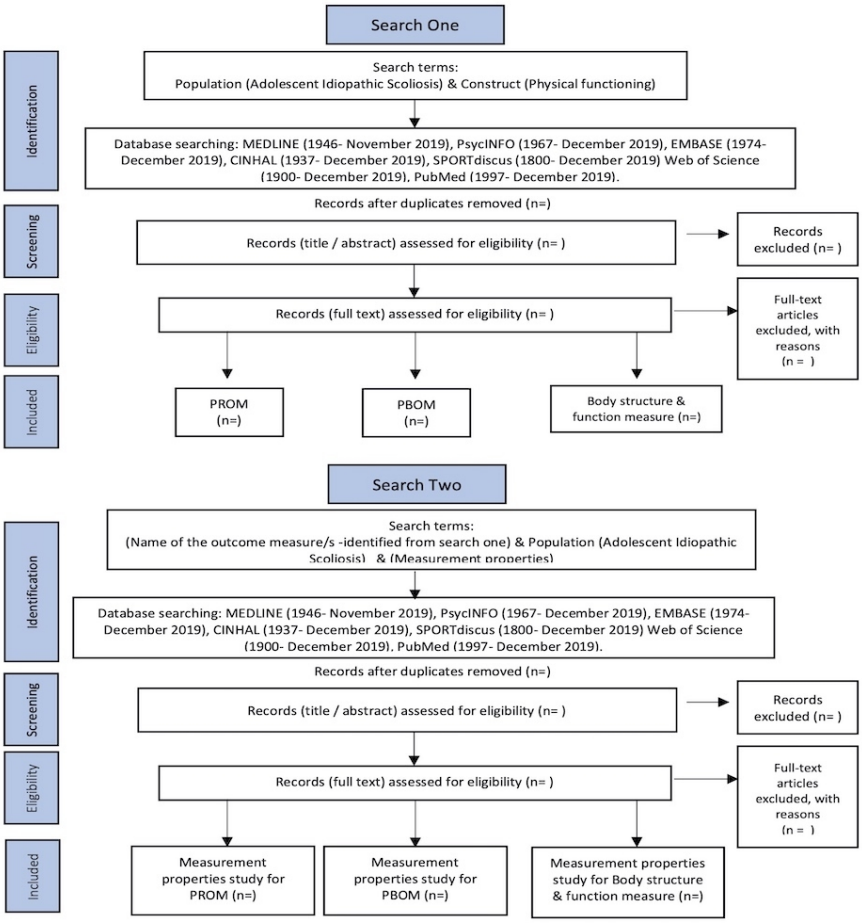

Figure 1: Flow diagram of search strategy (Search One and Two) and selection process. PROM=Patient Reported Outcome Measure; PBOM= Performance-Based Outcome Measure.

90x90mm (300 x 300 DPI)

Supplementary File 1

Example of search strategy (MEDLINE)

Stage 1

1. scoliosis.mp.
2. exp Scoliosis/
3. exp Spinal Curvatures/
4. Adolescen$.mp.
5. exp Adolescent/
6. Physical functioning.mp.
7. exp Physical Functional Performance/
8. Functional activity.mp.
9. independence.mp.
10. Functional independence.mp.
11. exp Health Status/
12. exp performance status/
13. exp Health Behavior/
14. exp Movement/
15. mobility.mp.
16. Functional limitation.mp.
17. Activity limitation.mp.
18. exp Motor Activity/
19. Recovery of function/
20. (Recover$ adj5 function$).tw.
21. exp Motor Skills/
22. exp Disability Evaluation/
23. exp Disabled Persons/
24. exp physical examination/
25. exp "Activities of Daily Living"/
26. ((daily or domestic or house or home) adj5 (activit$ or task$ or skill$ or chore$)).t
27. (Activities of daily living or adl$ or eadl$ or iadl$).tw.
28. exp Self Care/
29. ((self or personal) adj5 (Care or manage$)).tw.
30. (Dressing or feeding or eating or toilet$ or bathing or mobil$ or driving or public transport$).tw.
31. exp Lifting/
32. Bending.mp.

33. exp sitting/
34. exp Walking/
35. exp Walking Speed/
36. exp Postural Balance/
37. Standing balance.mp.
38. exp Hand Strength/ or Grip strength.mp.
39. 1- 3/OR
40. 4 OR 5
41. 6-38/OR
42. 39 and 40 and 41
43. Limit 42 to humans

Stage 2

44. Name of the Identified outcome measure/s
45. scoliosis.mp.
46. exp Scoliosis/
47. exp Spinal Curvatures/
48. Adolescen$.mp.
49. exp Adolescent/
50. validity.mp
51. exp validation studies/
52. reliability.mp
53. exp reproducibility of results/
54. interpretability.mp
55. internal consistency.mp
56. exp sensitivity and Specificity/
57. clinical sensitivity.mp
58. exp psychometrics/
59. responsiveness.mp
60. exp Evaluation studies/
61. measurement error.mp
62. measurement properties.mp
63. 45-47/OR
64. 48-49/OR
65. 50-62/OR
66. 44 and 63 and 64 and 65
67. Limit 66 to humans

PRISMA-P (Preferred Reporting Items for Systematic review and Meta-Analysis Protocols) 2015 checklist: recommended items to address in a systematic review protocol*

Section and topic	Item No	Checklist item	Reported on Page
ADMINISTRATIVE INFORMATION			
Title:			
Identification	1a	Identify the report as a protocol of a systematic review	Page 1 line 2
Update	1b	If the protocol is for an update of a previous systematic review, identify as such	NA
Registration	2	If registered, provide the name of the registry (such as PROSPERO) and registration number	Page 1 line 21
Authors:			
Contact	3a	Provide name, institutional affiliation, e-mail address of all protocol authors; provide physical mailing address of corresponding author	Page 1 line 4-19
Contributions	3b	Describe contributions of protocol authors and identify the guarantor of the review	Page13 line 350-354
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	NA
Support:			
Sources	5a	Indicate sources of financial or other support for the review	NA
Sponsor	5b	Provide name for the review funder and/or sponsor	Page13 line 356-357
Role of sponsor or funder	5c	Describe roles of funder(s), sponsor(s), and/or institution(s), if any, in developing the protocol	NA
INTRODUCTION			
Rationale	6	Describe the rationale for the review in the context of what is already known	Page 4 line 104-108 Page 5 line 130-134
Objectives	7	Provide an explicit statement of the question(s) the review will address with reference to participants, interventions, comparators, and outcomes (PICO)	Page 5 line 136-137
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	Page 6-7

Information sources	9	Describe all intended information sources (such as electronic databases, contact with study authors, trial registers or other grey literature sources) with planned dates of coverage	Page 7 line 199-210
Search strategy	10	Present draft of search strategy to be used for at least one electronic database, including planned limits, such that it could be repeated	Page 8 line 212-218 Supplementary file 1
Study records:			
Data management	11a	Describe the mechanism(s) that will be used to manage records and data throughout the review	Page 8 line 220-222
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)	Page 8 line 224-232
Data collection process	11c	Describe planned method of extracting data from reports (such as piloting forms, done independently, in duplicate), any processes for obtaining and confirming data from investigators	Page 8,9 line 234-238
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	Page 9, Table 1
Outcomes and prioritization	13	List and define all outcomes for which data will be sought, including prioritization of main and additional outcomes, with rationale	Page 10 line 250-252
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	Page 10 line 274-261
Data synthesis	15a	Describe criteria under which study data will be quantitatively synthesised	Page 10, 11line 275-277
	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 I_2, Kendall's τ)	Page 11 line 279-281
	15c	Describe any proposed additional analyses (such as sensitivity or subgroup analyses, meta-regression)	Page 11 line 277-279
	15d	If quantitative synthesis is not appropriate, describe the type of summary planned	Page 11 line 281-282
Meta-bias(es)	16	Specify any planned assessment of meta-bias(es) (such as publication bias across studies, selective reporting within studies)	NA
Confidence in cumulative evidence	17	Describe how the strength of the body of evidence will be assessed (such as GRADE)	Page 11 line 291-299

*** It is strongly recommended that this checklist be read in conjunction with the PRISMA-P Explanation and Elaboration (cite when available) for important clarification on the items. Amendments to a review protocol should be tracked and dated. The copyright for PRISMA-P (including checklist) is held by the PRISMA-P Group and is distributed under a Creative Commons Attribution Licence 4.0.**

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