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# BMJ Open

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

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3 1 **Outcome Measures Evaluating Physical Functioning and their Measurement Properties in**  
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5 2 **Adolescent Idiopathic Scoliosis: Protocol for a Systematic Review**  
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**29 ABSTRACT**

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

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

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

50 **PrOsPErO registration number:** CRD42019142335

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

53

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3 55 **ARTICLE SUMMARY**  
4

5 56 **Strengths and Limitations of This Study**  
6

- 7 57 • This will be the first systematic review to evaluate the measurement properties of all  
8 outcome measures evaluating physical functioning in AIS.  
9 58  
10 59 • This study will employ rigorous methods and using COnsensus-based Standards for the  
11 selection of health Measurement Instruments risk of bias tool and modified GRADE.  
12 60  
13  
14 61 • Although other measures of functioning such as role and social functioning are important to  
15 an AIS population, this study is focused on outcome measures of physical functioning.  
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## 65 INTRODUCTION

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

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

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

1  
2  
3 98 life which includes physical functioning. [16] However, the SRS-22r fails to fully capture important  
4  
5 99 aspects of physical functioning for AIS population e.g. self-care and mobility [17]. Where this study  
6  
7 100 included all forms of spinal deformities, the heterogeneity limits its applicability to AIS as a discrete  
8  
9 101 population.

10  
11 102 Establishment of measurement properties of outcome measures are important to avoid the risk of  
12  
13 103 bias and ensure accuracy in the evaluation of test results.[34] The COnsensus-based Standards for  
14  
15 104 the selection of health Measurement INstruments, (COSMIN) group developed a taxonomy of  
16  
17 105 measurement properties to improve the selection of outcome measures. [35] Three main domains  
18  
19 106 identified, reliability, validity and responsiveness with further subgrouping.[35] In the absence of  
20  
21 107 existing evidence, a systematic review is needed to inform and summarise the evidence on physical  
22  
23 108 functioning outcome measure in AIS population and to evaluate their measurement properties.

## 24 109 **Objective**

25  
26  
27 110 To synthesise evidence of physical functioning outcome measures in AIS population. A secondary  
28  
29 111 aim is to evaluate measurement properties of outcome measures evaluating physical functioning in  
30  
31 112 AIS.

## 32 33 113 **METHODS**

34  
35  
36 114 This protocol has been informed by expertise in the field including a surgeon, musculoskeletal  
37  
38 115 rehabilitation experts including physiotherapists and individuals with methodological expertise. It  
39  
40 116 has been designed in line with the Cochrane handbook [36] and is reported in line with the Preferred  
41  
42 117 Reporting Items for Systematic Reviews and Meta-Analysis-P (PRISMA-P) (Supplementary File  
43  
44 118 1).[37]

## 45 119 **Eligibility criteria**

46  
47  
48 120 Inclusion criteria

### 49 50 51 121 *Participants*

52  
53  
54 122 Participants aged between the age of 10 years until the end of bone growth with a diagnosis of  
55  
56 123 idiopathic scoliosis and  $\geq 10^\circ$  Cobb angle will be considered. The end of bone growth is estimated by  
57  
58 124 the Risser classification, which grades the level of ossification and fusion of the iliac crest apophyses  
59  
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1  
2  
3 125 into six stages [38] where stage 0 describes no ossification, and Stage 5 represents complete  
4  
5 126 ossification and fusion of the iliac apophysis and end of bone growth. [38]  
6  
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### *Outcome measures*

11  
12  
13 129 Any study that includes assessments of the physical functioning of AIS using specific outcome  
14  
15 130 measure will be included. Physical functioning defined according to the Core Outcome Measures in  
16  
17 131 Effectiveness Trials (COMET) taxonomy [15] as any physical activities of daily living such as the  
18  
19 132 ability to walk, independence, self-care, performance status, disability index. [15, 39] The outcome  
20  
21 133 measures being defined as any one of the following:

- 22  
23 134 1. Patient Reported Outcome Measures (PROMs) in the form of questionnaires or scales  
24  
25 135 designed for AIS to evaluate physical functioning (e.g. SRS-22r) and/or
- 26  
27 136 2. Performance-based outcome measures (PBOMs); a measure of physical functioning by  
28  
29 137 clinician while the individual is performing a functional task (e.g. standing balance, walking  
30  
31 138 speed and/or
- 31  
32 139 3. Impairment measure (IMs) which means any dysfunction in a specific body part or system  
33  
34 140 which limits function, such as muscle performance and range of motion. [23]  
35

### *Measurement properties*

36 141  
37  
38 142 Any study that has evaluated one or more measurement properties of the above-mentioned  
39  
40 143 outcome measures in AIS. This will include all measurement properties in the three main domains of  
41  
42 144 the COSMIN Taxonomy (i.e. reliability, validity, and responsiveness). [35] The definitions of  
43  
44 145 measurement properties according to COSMIN taxonomy are summarised in the online  
45  
46 146 supplementary file 2.  
47

### *Study design*

48 147  
49  
50  
51 148 In the first stage, all study designs including; randomised clinical trials, cohort, observational studies  
52  
53 149 and case studies will be included to identify all outcome measure of physical functioning being used  
54  
55 150 within AIS population. The second stage will include the validation studies of the physical functioning  
56  
57 151 measures identified in the first stage.  
58

### **Information sources**

1  
2  
3 153 A search strategy has been developed using medical subject headings, where available and relevant  
4  
5 154 text words relating to AIS, physical functioning, outcome measures and measurement properties. An  
6  
7 155 electronic search of databases will be conducted including MEDLINE (OVID interface), PsycINFO  
8  
9 156 (OVID interface), EMBASE (OVID interface), SPORT discus (EBSCO interface), CINHALL (EBSCO  
10  
11 157 interface), Web of Science and PubMed. A hand search in the key journals including Spine, The Spine  
12  
13 158 Journal, Spine Deformity, Scoliosis and Spinal Disorder, Scoliosis and European Spine Journal as well  
14  
15 159 as contacting relevant leading researchers in the field. Further, searching of the Grey literature,  
16  
17 160 including conference proceedings, British National bibliography for report literature, open-Grey,  
18  
19 161 dissertation abstracts and ETHOS will be conducted.

### 19 162 **Search strategy**

21  
22 163 Two reviewers (SA, EB) will independently complete searches and identify potential studies for  
23  
24 164 eligibility. To ensure that all relevant data are included, no limitations will be applied. Initial search  
25  
26 165 terms will be developed for MEDLINE and then adapted with relevant syntax and subject headings  
27  
28 166 for other databases. Recommended search filters specifically designed for retrieving articles on  
29  
30 167 measurement properties will be used where appropriate.[40] An example of the search strategy of  
31  
32 168 both stages is available as an online supplementary file 3.

### 33 169 **Data management**

34  
35  
36 170 Search records will be imported into Endnote Version X9 (Clarivate Analytics). Using Endnote, the  
37  
38 171 abstracts and full texts will be stored, and any duplicates will be identified and removed.

### 40 172 **Selection Process**

41  
42  
43 173 Two reviewers (SA, EB) will independently search information sources and assess study eligibility  
44  
45 174 according to inclusion/exclusion criteria by grading each study as eligible/not eligible/might be  
46  
47 175 eligible. [41] If a study cannot be clearly excluded based on the inclusion criteria, the full text will be  
48  
49 176 retrieved. Study selection (included and excluded studies) with the reasons for exclusion, will be  
50  
51 177 summarised in a PRISMA flow diagram (Supplementary file 1 ).[37] Articles will be included if both  
52  
53 178 reviewers agreed that the eligibility criteria were met. Any disagreement will be first discussed and  
54  
55 179 the third reviewer (NH) will mediate situations of disagreement. The percentage of agreement  
56  
57 180 between reviewers on the extracted data will be reported.

### 57 181 **Data collection process**

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

186

187

### 188 **Data items**

189 The data that will be extracted from each study at each stage is summarised in Table 1. In the case of  
 190 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

### 191 **Outcomes and prioritisation**

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

### 194 **Risk of bias in individual studies**

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

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2  
3 203 reached, a third reviewer (NH) will be consulted. The percentage of agreement between reviewers  
4  
5 204 will be reported in the final results.  
6

### 7 8 205 **Data synthesis**

9  
10 206 Scoping searches of the currently available literature indicate heterogeneity of the outcome  
11  
12 207 measures used for assessment of physical functioning in AIS. Thus, a meta-analysis may not be  
13  
14 208 possible. If deemed not possible, a narrative synthesis will be conducted. The COSMIN guidelines for  
15  
16 209 systematic reviews will be used for the synthesis of the results.[43] The methodological quality of  
17  
18 210 each single study on a measurement property will be assessed using the COSMIN Risk of Bias  
19  
20 211 checklist (Online supplementary file 2).[42] The result will be rated against the predefined criteria for  
21  
22 212 good measurement properties as sufficient (+), insufficient (–), inconsistent ( $\pm$ ), or indeterminate (?).  
23  
24 213 [43] The evidence will be summarised for each measurement property for PROM, PBOM or IM and  
25  
26 214 the overall result is rated against the criteria for good measurement properties. [43]

### 27 215 **Confidence in cumulative evidence**

28  
29  
30 216 The overall quality and strength of evidence will be assessed using a modified Grading of  
31  
32 217 Recommendations Assessment, Development and Evaluation (GRADE) approach for systematic  
33  
34 218 reviews of clinical trials. [43] The GRADE approach uses five factors to determine the quality of the  
35  
36 219 evidence: risk of bias (quality of the studies), inconsistency (of the results of the studies),  
37  
38 220 indirectness (evidence comes from different populations, interventions or outcomes than the ones  
39  
40 221 of interest in the review), imprecision (wide confidence intervals), and publication bias (negative  
41  
42 222 results are less often published).[44] For evaluating measurement properties in systematic reviews  
43  
44 223 of PROMs, only four factors will be assessed as recommended by COSMIN, while the fifth factor  
45  
46 224 (publication bias) was removed as there is no registry exists for measurement properties. [43]  
47

### 48 225 49 226 **Patient and public involvement**

50 227  
51 228 The study question and systematic review protocol were informed following discussion at a patient  
52  
53 229 and public involvement meeting at the Centre of Precision Rehabilitation for Spinal Pain at the  
54  
55 230 University of Birmingham. Since no patient data is needed, patients will not be involved in data  
56  
57 231 collection or analysis. However, the results of the study will be shared at public engagement events.  
58

### 59 232 60 233 **Implications of this study**

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3 235 The AIS is a complex deformity of the spine and causes a significant impact on physical activities of  
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5 236 individuals' daily living such as lifting objects and maintaining body position. [17, 19] In  
6  
7 237 consequence, the quality of life is affected. Physical functioning gives an indication about the current  
8  
9 238 health status and identifies people at risk of disability.[12, 13] Therefore, physical functioning is  
10  
11 239 considered as one of the outcomes that should be assessed and reported in clinical trials of  
12  
13 240 musculoskeletal conditions. [15] Within the AIS population, little attention has been paid to the  
14  
15 241 physical functioning measures and their measurement properties. This systematic review will be the  
16  
17 242 first assessing the measurement properties of physical functioning outcome measures among the  
18  
19 243 AIS population. This will inform clinicians and researchers of the best available tools for assessment  
20  
21 244 of physical functioning in AIS.  
22

245

#### 246 **Declarations**

#### 247 **Ethics and Dissemination**

248 No ethics approval is required for this systematic review. The results will be disseminated through a  
27  
28 249 peer-reviewed publication and conference presentation.

#### 250 **Author Contributions**

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

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#### 258 **Competing interests**

259 None declared.

#### 260 **Patient consent for publication**

261 Not required.

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## 264 REFERENCES

- 265
- 266 1. Reamy, B.V. and J.B. Slakey, Adolescent idiopathic scoliosis: review and current  
267 concepts. *Am Fam Physician*, 2001. **64**(1):111-6.
- 268 2. Cobb, J.R., The problem of the primary curve. *J Bone Joint Surg Am*, 1960. **42-**  
269 **a**:1413-25.
- 270 3. Konieczny, M.R., H. Senyurt, and R. Krauspe, Epidemiology of adolescent idiopathic  
271 scoliosis. *Journal of Children's Orthopaedics*, 2013. **7**(1):3-9.
- 272 4. Hamad, A., E.B. Ahmed, and A.I. Tsirikos, Adolescent idiopathic scoliosis: a  
273 comprehensive approach to aetiology, diagnostic assessment and treatment.  
274 *Orthopaedics and Trauma*, 2017. **31**(6):343-9.
- 275 5. Horne, J.P., R. Flannery, and S. Usman, Adolescent idiopathic scoliosis: diagnosis  
276 and management. *Am Fam Physician*, 2014. **89**(3):193-8.
- 277 6. Lonstein, J.E., Adolescent idiopathic scoliosis. *The Lancet*, 1994. **344**(8934):1407-12.
- 278 7. Du, C., et al., Relevant areas of functioning in patients with adolescent idiopathic  
279 scoliosis on the International Classification of Functioning, Disability and Health: The  
280 patients' perspective. *J Rehabil Med*, 2016. **48**(9):806-14.
- 281 8. Makino, T., et al., Low back pain and patient-reported QOL outcomes in patients with  
282 adolescent idiopathic scoliosis without corrective surgery. *SpringerPlus*, 2015. **4**(1).
- 283 9. Durmala, J., W. Tomalak, and T. Kotwicki, Function of the respiratory system in  
284 patients with idiopathic scoliosis: reasons for impairment and methods of evaluation.  
285 *Stud Health Technol Inform*, 2008. **135**:237-45.
- 286 10. Leszczewska, J., et al., Evaluation of the Stress Level of Children with Idiopathic  
287 Scoliosis in relation to the Method of Treatment and Parameters of the Deformity.  
288 *The Scientific World Journal*, 2012. **2012**:1-5.
- 289 11. Malmqvist, M., et al., Patients With Idiopathic Scoliosis Run an Increased Risk of  
290 Schizophrenia. *Spine Deform*, 2019. **7**(2):262-6.

- 1  
2  
3 291 12. Taylor, A.M., et al., Assessment of physical function and participation in chronic pain  
4 292 clinical trials: IMMPACT/OMERACT recommendations. *Pain*, 2016. **157**(9):1836-  
5 293 50.  
6  
7  
8  
9 294 13. Tomey, K.M. and M.R. Sowers, Assessment of physical functioning: a conceptual  
10 295 model encompassing environmental factors and individual compensation strategies.  
11 296 *Physical therapy*, 2009. **89**(7):705-14.  
12  
13  
14  
15 297 14. Cooper, R., et al., Objective measures of physical capability and subsequent health: a  
16 298 systematic review. *Age and ageing*, 2011. **40**(1):14-23.  
17  
18  
19  
20 299 15. Dodd, S., et al., A taxonomy has been developed for outcomes in medical research to  
21 300 help improve knowledge discovery. *Journal of clinical epidemiology*, 2018. **96**:84-92.  
22  
23  
24 301 16. de Kleuver, M., et al., Defining a core outcome set for adolescent and young adult  
25 302 patients with a spinal deformity: A collaborative effort for the Nordic Spine Surgery  
26 303 Registries. *Acta Orthopaedica*, 2017. **88**(6):612-8.  
27  
28  
29  
30 304 17. Du, C., et al., Relevant areas of functioning in patients with adolescent idiopathic  
31 305 scoliosis on the International Classification of Functioning, Disability and Health: The  
32 306 patientsâ€™ perspective. *Journal of Rehabilitation Medicine*, 2016. **48**(9):806-14.  
33  
34  
35  
36 307 18. Kibsgård, T., J.I. Brox, and O.J.J.o.O.S. Reikerås, Physical and mental health in  
37 308 young adults operated on for idiopathic scoliosis. 2004. **9**(4):360-3.  
38  
39  
40  
41 309 19. Lerman, J.A., E. Sullivan, and R.J. Haynes, The Pediatric Outcomes Data Collection  
42 310 Instrument (PODCI) and functional assessment in patients with adolescent or juvenile  
43 311 idiopathic scoliosis and congenital scoliosis or kyphosis. *Spine*, 2002. **27**(18):2052-7;  
44 312 discussion 7-8.  
45  
46  
47  
48  
49 313 20. Bastrom, T.P., et al., Prevalence of Postoperative Pain in Adolescent Idiopathic  
50 314 Scoliosis and the Association With Preoperative Pain. *Spine*, 2013. **38**(21):1848-52.  
51  
52  
53  
54 315 21. Seki, H., et al., Postoperative pain management in patients undergoing posterior  
55 316 spinal fusion for adolescent idiopathic scoliosis: a narrative review. *Scoliosis and*  
56 317 *Spinal Disorders*, 2018. **13**(1):17.  
57  
58  
59  
60

- 1  
2  
3 318 22. Ward, M.M., Interpreting measurements of physical function in clinical trials. *Annals*  
4 319 *of the Rheumatic Diseases*, 2007. **66**(Supplement 3):iii32-iii4.  
5  
6  
7 320 23. Reiman, M.P. and R.C. Manske, The assessment of function: How is it measured? A  
8 321 clinical perspective. *The Journal of manual & manipulative therapy*, 2011. **19**(2):91-  
9 322 9.  
10  
11  
12  
13 323 24. Negrini, S., et al., Why do we treat adolescent idiopathic scoliosis? What we want to  
14 324 obtain and to avoid for our patients. SOSORT 2005 Consensus paper. *Scoliosis*, 2006.  
15 325 **1**:4.  
16  
17  
18  
19 326 25. Haheer, T.R., et al., Results of the Scoliosis Research Society instrument for evaluation  
20 327 of surgical outcome in adolescent idiopathic scoliosis. A multicenter study of 244  
21 328 patients. *Spine (Phila Pa 1976)*, 1999. **24**(14):1435-40.  
22  
23  
24  
25 329 26. Baldus, C., et al., The Scoliosis Research Society Health-Related Quality of Life  
26 330 (SRS-30) age-gender normative data: an analysis of 1346 adult subjects unaffected by  
27 331 scoliosis. *Spine*, 2011. **36**(14):1154-62.  
28  
29  
30  
31 332 27. Chen, A.F., et al., Converting Scoliosis Research Society-24 to Scoliosis Research  
32 333 Society-22r in a Surgical-Range, Medical/Interventional Adolescent Idiopathic  
33 334 Scoliosis Patient Cohort. *Spine Deform*, 2013. **1**(2):108-14.  
34  
35  
36  
37 335 28. Asher, M., et al., Discrimination validity of the scoliosis research society-22 patient  
38 336 questionnaire: relationship to idiopathic scoliosis curve pattern and curve size. *Spine*  
39 337 *(Phila Pa 1976)*, 2003. **28**(1):74-8.  
40  
41  
42  
43 338 29. Asher, M.A., S.M. Lai, and D.C. Burton, Further development and validation of the  
44 339 Scoliosis Research Society (SRS) outcomes instrument. *Spine*, 2000. **25**(18):2381-6.  
45  
46  
47  
48 340 30. Negrini, S., et al., Recommendations for research studies on treatment of idiopathic  
49 341 scoliosis: Consensus 2014 between SOSORT and SRS non-operative management  
50 342 committee. *Scoliosis*, 2015. **10**:8.  
51  
52  
53  
54 343 31. Wang, S., et al., Evaluation of Performance-Based Outcome Measures for the Upper  
55 344 Limb: A Comprehensive Narrative Review. *PM&R*, 2018. **10**(9):951-62.e3.  
56  
57  
58  
59  
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- 1  
2  
3 345 32. Mizner, R.L., et al., Measuring Functional Improvement After Total Knee  
4 346 Arthroplasty Requires Both Performance-Based and Patient-Report Assessments: A  
5 347 Longitudinal Analysis of Outcomes. *The Journal of Arthroplasty*, 2011. **26**(5):728-37.  
6  
7  
8  
9 348 33. Bean, J.F., et al., Performance-based versus patient-reported physical function: what  
10 349 are the underlying predictors? *Phys Ther*, 2011. **91**(12):1804-11.  
11  
12  
13 350 34. Mokkink, L.B., et al., The COSMIN checklist for assessing the methodological  
14 351 quality of studies on measurement properties of health status measurement  
15 352 instruments: an international Delphi study. *Quality of Life Research*, 2010. **19**(4):539-  
16 353 49.  
17  
18  
19 354 35. Mokkink, L.B., et al., The COSMIN study reached international consensus on  
20 355 taxonomy, terminology, and definitions of measurement properties for health-related  
21 356 patient-reported outcomes. *Journal of Clinical Epidemiology*, 2010. **63**(7):737-45.  
22  
23  
24 357 36. Higgins, J.P.T. and S. Green, *Cochrane handbook for systematic reviews of*  
25 358 *interventions version 5.1.0 [updated March 2011]*. 2011, The Cochrane  
26 359 Collaboration.  
27  
28 360 37. Moher, D., et al., Preferred reporting items for systematic review and meta-analysis  
29 361 protocols (PRISMA-P) 2015 statement. *Systematic reviews*, 2015. **4**:1.  
30  
31  
32 362 38. Hacquebord, J.H. and S.S. Leopold, In brief: The Risser classification: a classic tool  
33 363 for the clinician treating adolescent idiopathic scoliosis. *Clinical orthopaedics and*  
34 364 *related research*, 2012. **470**(8):2335-8.  
35  
36  
37 365 39. Williamson, P.R., et al., The COMET Handbook: version 1.0. *Trials*, 2017. **18**(S3).  
38  
39  
40 366 40. Terwee, C.B., et al., Development of a methodological PubMed search filter for  
41 367 finding studies on measurement properties of measurement instruments. *Quality of*  
42 368 *Life Research*, 2009. **18**(8):1115-23.  
43  
44  
45 369 41. Furlan, A.D., et al., 2015 Updated Method Guideline for Systematic Reviews in the  
46 370 Cochrane Back and Neck Group. *Spine*, 2015. **40**(21):1660-73.  
47  
48  
49 371 42. Mokkink, L.B., et al., COSMIN Risk of Bias checklist for systematic reviews of  
50 372 Patient-Reported Outcome Measures. *Quality of Life Research*, 2018. **27**(5):1171-9.

- 1  
2  
3 373 43. Prinsen, C.A.C., et al., COSMIN guideline for systematic reviews of patient-reported  
4 374 outcome measures. *Quality of life research : an international journal of quality of life*  
5 375 *aspects of treatment, care and rehabilitation*, 2018. **27**(5):1147-57.  
6  
7  
8  
9 376 44. Balshem, H., et al., GRADE guidelines: 3. Rating the quality of evidence. *Journal of*  
10 377 *Clinical Epidemiology*, 2011. **64**(4):401-6.  
11  
12 378  
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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
<b>ADMINISTRATIVE INFORMATION</b>		
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
<b>INTRODUCTION</b>		
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)
<b>METHODS</b>		
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 $\tau$ )
	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 (test-retest) 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” <sup>a</sup> 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”
<b>Responsiveness</b>			The ability of an HR-PRO instrument to detect change over time in the construct to be measured
	Responsiveness		Idem responsiveness
<b>Interpretability<sup>b</sup></b>			The degree to which one can assign qualitative meaning <sup>d</sup> 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
<b>Structural validity</b>	+	<p><b>CTT:CFA:</b> CFI or TLI or comparable measure <math>&gt; 0.95</math> OR RMSEA <math>&lt; 0.06</math> OR SRMR <math>&lt; 0.08</math><sup>a</sup></p> <p><b>IRT/Rasch:</b> No violation of unidimensionality <sup>b</sup>: CFI or TLI or comparable measure <math>&gt; 0.95</math> OR RMSEA <math>&lt; 0.06</math> OR SRMR <math>&lt; 0.08</math></p> <p>AND</p> <p>no violation of local independence: residual correlations among the items after controlling for the dominant factor <math>&lt; 0.20</math> OR Q3's <math>&lt; 0.37</math></p> <p>AND</p> <p>no violation of monotonicity: adequate looking graphs OR item scalability <math>&gt; 0.30</math></p> <p>AND</p> <p>adequate model fit</p> <p>IRT: <math>\chi^2 &gt; 0.001</math></p> <p>Rasch: infit and outfit mean squares <math>\geq 0.5</math> and <math>\leq 1.5</math> OR Z-standardized values <math>&gt; -2</math> and <math>&lt; 2</math></p>
	?	CTT: not all information for '+' reported IRT/Rasch: model fit not reported
	-	Criteria for '+' not met
<b>Internal consistency</b>	+	At least low evidence for sufficient structural validity AND Cronbach's alpha(s) $\geq 0.70$ for each unidimensional scale or subscale
	?	Criteria for "At least low evidence <sup>c</sup> for sufficient structural validity <sup>d</sup> " not met
	-	At least low evidence <sup>c</sup> for sufficient structural validity <sup>d</sup> AND Cronbach's alpha(s) $< 0.70$ for each unidimensional scale or subscale <sup>e</sup>
<b>Reliability</b>	+	ICC or weighted Kappa $\geq 0.70$
	?	ICC or weighted Kappa not reported
	-	ICC or weighted Kappa $< 0.70$
<b>Measurement error</b>	+	SDC or LoA $< MIC$ <sup>d</sup>
	?	MIC not defined
	-	SDC or LoA $> MIC$ <sup>d</sup>
<b>Hypotheses testing for construct validity</b>	+	The result is in accordance with the hypothesis <sup>f</sup>
	?	No hypothesis defined (by the review team)
	-	The result is not in accordance with the hypothesis <sup>f</sup>
<b>Cross-cultural validity\measurement invariance</b>	+	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
<b>Criterion validity</b>	+	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
<b>Responsiveness</b>	+	The result is in accordance with the hypothesis <sup>f</sup> OR AUC ≥ 0.70
	?	No hypothesis defined (by the review team)
	–	The result is not in accordance with the hypothesis <sup>f</sup> 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

<sup>a</sup> To rate the quality of the summary score, the factor structures should be equal across studies

<sup>b</sup> Unidimensionality refers to a factor analysis per subscale, while structural validity refers to a factor analysis of a (multidimensional) patient reported outcome measure

<sup>c</sup> As defined by grading the evidence according to the GRADE approach

<sup>d</sup> This evidence may come from different studies

<sup>e</sup> 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

<sup>f</sup> 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. Adolescenc\$.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/

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- 4 35. exp Walking/
- 5 36. exp Walking Speed/
- 6 37. exp Postural Balance/
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- 8 38. Standing balance.mp.
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- 10 39. exp Hand Strength/ or Grip strength.mp.
- 11 40. 1- 4/OR
- 12 41. 5 OR 6
- 13 42. 7-39/OR
- 14 43. 40 and 41 and 42
- 15 44. Limit 43 to humans
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## 20 Stage 2

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- 22 45. Name of the Identified outcome measure
- 23 46. validity.mp
- 24 47. exp validation studies/
- 25 48. reliability.mp
- 26 49. exp reproducibility of results/
- 27 50. interpretability.mp
- 28 51. internal consistency.mp
- 29 52. exp sensitivity and Specificity/
- 30 53. clinical sensitivity.mp
- 31 54. exp psychometrics/
- 32 55. responsiveness.mp
- 33 56. exp Evaluation studies/
- 34 57. measurement error.mp
- 35 58. measurement properties.mp
- 36 59. 46-58/OR
- 37 60. 1 AND 59
- 38 61. Limit 60 to humans
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# BMJ Open

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

Journal:	<i>BMJ Open</i>
Manuscript ID	bmjopen-2019-034286.R1
Article Type:	Protocol
Date Submitted by the Author:	13-Jan-2020
Complete List of Authors:	Alamrani, Samia; University of Birmingham, Centre of Precision Rehabilitation for Spinal Pain, School of Sport, Exercise & Rehabilitation Sciences; University of Tabouk, Physical Therapy Department, College of Applied Medical Science Rushton, Alison; University of Birmingham, Centre of Precision Rehabilitation for Spinal Pain Gardner, Adrian; The Royal Orthopaedic Hospital Birmingham Falla, Deborah; University of Birmingham, School of Sport, Exercise and Rehabilitation Sciences Heneghan, Nicola; University of Birmingham, Centre of Precision Rehabilitation for Spinal Pain, School of Sport, Exercise and Rehabilitation Sciences
<b>Primary Subject Heading</b>:	Rehabilitation medicine
Secondary Subject Heading:	Patient-centred medicine, Paediatrics
Keywords:	Adolescent Idiopathic Scoliosis, Measurement properties, Physical function, Scoliosis < ORTHOPAEDIC & TRAUMA SURGERY

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3 1 **Outcome Measures Evaluating Physical Functioning and their Measurement Properties in**  
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5 2 **Adolescent Idiopathic Scoliosis: Protocol for a Systematic Review**  
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8 3 **Authors**  
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3 29 **ABSTRACT**  
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6 30 **Introduction** Physical functioning (PF) is the ability to carry out physical activity of daily living. It is an  
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8 31 important outcome that provide meaningful evaluation of individuals' life. PF can be assessed using  
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10 32 Patient-Reported outcome measures, Performance-Based Outcome Measures or Body Structure and  
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12 33 Function Measure. Measures need to be valid, reliable and responsive to change to evaluate effects  
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14 34 of an intervention. Adolescent Idiopathic Scoliosis (AIS) is the most common deformity among  
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16 35 the paediatric population. It causes significant impact on individuals' life. In the absence of existing  
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18 36 evidence, this systematic review will appraise evidence on measurement properties of PF tools in  
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20 37 individuals with AIS.

21 38 **Methods/analysis** A protocol for systematic review and meta-analysis informed by Cochrane  
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23 39 guidelines and, reported in line with Preferred Reporting Items for Systematic Reviews and Meta-  
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25 40 Analysis-P. MEDLINE, PsycINFO, EMBASE, CINHALL, SPORTdiscus, Web of science and PubMed will be  
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27 41 searched in two stages (inception until December 2019). Search one will include all studies that  
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29 42 assessed PF in participants with AIS without any limitations. Search terms will be Scoliosis, Adolescent  
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31 43 and PF related terms. Search two will include studies which investigated instrument measurement  
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33 44 properties in the same population for measures identified in search one. Two reviewers will  
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35 45 independently perform study selection, data extraction, risk of bias, and overall quality assessment.  
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37 46 The Consensus-based Standards for the selection of Health Measurement Instruments risk of bias and  
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39 47 a modified Grading of Recommendations, Assessment, Development and Evaluation guidelines will be  
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41 48 used. Meta-analysis will be conducted if possible, or the evidence will be synthesised and summarized  
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43 49 for each outcome measure and according to specific measurement properties.

44 50 **Ethics and dissemination:** This review will provide recommendations for practice and future  
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46 51 research, considering psychometric properties of outcome measures of PF in AIS. The results of this  
47  
48 52 study will be disseminated through a peer-reviewed publication and conference presentation.

49 53 **Keywords:** Adolescent Idiopathic Scoliosis, measurement properties, physical function, systematic  
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51 54 review  
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3 59 **ARTICLE SUMMARY**  
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6 60 **Strengths and Limitations of this Study**  
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- 9 62 1. This review synthesises evidence of patient-reported, performance-based or, body structure  
10 63 and function outcome measures of physical functioning, for use in practice or research  
11 64 involving individuals with AIS.  
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13 65 2. The search strategy of this review comprises two stages, to ensure that all measures of  
14 66 physical functioning of adolescent idiopathic scoliosis are included.  
15  
16 67 3. This study will employ rigorous methods and using COnsensus-based Standards for the  
17 68 selection of health Measurement Instruments risk of bias tool and modified GRADE.  
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19 69 4. This review is limited to studies of the English language that assess measurement properties  
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21 70 among adolescents with idiopathic scoliosis.  
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## 87 INTRODUCTION

88 Scoliosis is a complex three-dimensional deformity of the spine, including lateral curvature and  
89 rotation of the vertebrae, [1] and characterised by a curve angle  $\geq 10^\circ$ . [2] There are two main types  
90 of scoliosis, idiopathic and non-idiopathic with the latter arising from congenital, neuromuscular or  
91 mesenchymal causes. [3] While the aetiology of idiopathic scoliosis remains unknown; genetic,  
92 hormonal, and mechanical factors are involved. [4] Adolescent Idiopathic Scoliosis (AIS) often  
93 develops between 10 and 16 years of age and represents ~85% of cases. [5] AIS is the most common  
94 spinal deformity among the pediatric population, with a prevalence ranging from 2% to 3%. [6] Nearly  
95 80% of those affected presents with a curvature of the thoracic or thoracolumbar/lumbar region.[3]  
96 Whilst males and females are equally affected, females are reported to be at 10 times greater risk of  
97 curve progression. [1]

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

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



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3 120 with AIS, little is known about the available PBOMs for evaluating physical functioning. The SRS-22r  
4  
5 121 questionnaire is the gold standard outcome measure of quality of life, which include physical  
6  
7 122 functioning items as recommended by the recent COS study for adolescents and young adults with  
8  
9 123 spinal deformity. [16] However, the SRS-22r fails to fully capture important aspects of physical  
10  
11 124 functioning for individuals with AIS e.g. self-care and mobility. [7] This study included all forms of spinal  
12  
13 125 deformities, the heterogeneity limits applicability to individuals with AIS as a discrete population.

14  
15 126 Adequate measurement properties of outcome measures are important to avoid the risk of bias and  
16  
17 127 ensure accuracy in the evaluation of test results.[28] The COnsensus-based Standards for the selection  
18  
19 128 of health Measurement INstruments, (COSMIN) group developed a taxonomy of measurement  
20  
21 129 properties to improve the selection of outcome measures. [29] Three main domains identified  
22  
23 130 reliability, validity and responsiveness with further subgrouping.[28] A systematic review is needed to  
24  
25 131 evaluate measures of assessment and, their measurement properties, of physical functioning for  
26  
27 132 individuals with AIS. Review findings will inform clinicians and researchers on the best available tools  
28  
29 133 for assessment of physical functioning in AIS. Furthermore, findings will inform future research  
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31 134 drawing on a range of measures of physical functioning to investigate health status in AIS.

### 32 33 34 35 36 37 38 135 **Objective**

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41 136 To identify outcome measures used to assess physical functioning in individuals with AIS. A secondary  
42  
43 137 aim is to evaluate the measurement properties of physical functioning outcome measures in AIS.

### 44 45 46 47 48 49 50 51 52 53 54 55 56 57 58 59 60 138 **METHODS**

41 139 This protocol has been informed by experts in musculoskeletal orthopaedics including a consultant  
42  
43 140 spine surgeon, musculoskeletal rehabilitation experts including physiotherapists and individuals with  
44  
45 141 review, measurement properties and research experience. It has been designed in line with the  
46  
47 142 COSMIN methodology for systematic reviews of Patient-Reported Outcome Measures [30] and is  
48  
49 143 reported in line with the Preferred Reporting Items for Systematic Reviews and Meta-Analysis-P  
50  
51 144 (PRISMA-P) [31] The search for this systematic review will be conducted in two parts. Stage one to  
52  
53 145 identify studies used an outcome measure to evaluate physical functioning in individuals with AIS. This  
54  
55 146 search will allow a list of outcome measures to be generated. Stage 2 will identify studies, which  
56  
57 147 evaluated measurement properties of physical functioning outcome measure identified in the search  
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59 148 one.

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3 150 **Stage one: Outcome measure**  
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6 151 **Eligibility criteria**  
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8 152 *Study design*  
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10 153 All study designs including; randomised clinical trials, cohort, observational studies and case studies  
11  
12 154 will be included to identify all outcome measure of physical functioning being used with individuals  
13  
14 155 with AIS. No limitation on language or location will be applied at this stage.  
15

16  
17 156 *Participants*  
18

19 157 Participants aged between the age of 10 years to 18 years of age, with a diagnosis of idiopathic  
20  
21 158 scoliosis and  $\geq 10^\circ$  Cobb angle will be considered. No restrictions will be applied to the curve severity,  
22  
23 159 evaluation settings, and type of treatment as well as to the language or country.  
24

25 160 *Outcome*  
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27  
28 161 Any study that includes assessments of the physical functioning of AIS using specific outcome measure  
29  
30 162 will be included. Physical functioning defined according to the Core Outcome Measures in  
31  
32 163 Effectiveness Trials (COMET) taxonomy, [15] as any physical activities of daily living such as the ability  
33  
34 164 to walk, independence, self-care, performance status, disability index. [15, 32] The outcome measures  
35  
36 165 being defined as any one of the following:

- 37 166 1. Patient-Reported Outcome Measures (PROMs) in the form of questionnaires or scales  
38  
39 167 designed for AIS to evaluate physical functioning or if it is included as a sub-scale within a  
40  
41 168 questionnaire.  
42  
43 169 2. Performance-based outcome measures (PBOMs); a measure of physical functioning by  
44  
45 170 clinician while the individual is performing a functional task e.g walking, and/or  
46  
47 171 3. Body structure and function measure which means any dysfunction in a specific body part or  
48  
49 172 system which may limits function, such as range of motion. [21]  
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3 177 **Stage two: measurement properties**  
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6 178 **Eligibility criteria**  
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8 179 *Study design*  
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10  
11 180 Any study that has evaluated one or more measurement properties of the identified outcome  
12 181 measures in the first search will be eligible. Only full-text studies available in English will be included.  
13 182 Conference abstracts will be excluded due to the inability to effectively evaluate the quality of the  
14 183 study.  
15  
16  
17

18  
19 184 *Participants*  
20

21  
22 185 Participants aged between the age of 10 to 18 years of age, with a diagnosis of idiopathic scoliosis and  
23 186  $\geq 10^\circ$  Cobb angle will be eligible. In studies with mixed cohorts, >50% of participants should be  
24 187 individuals with AIS for the study to be included. Authors of studies will be contacted in case of missing  
25 188 information about number of participants with AIS. Studies without original participant data (e.g.  
26 189 systematic review) will be excluded.  
27  
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30  
31 190 *Outcome*  
32

33  
34 191 Outcome of interest is the measurement properties: Reliability including (Internal consistency, test-  
35 192 retest, inter-rater and intra-rater), measurement error, validity (Content validity, Structural validity or  
36 193 Criterion validity), hypothesis testing, responsiveness [29] of the outcome measures identified in the  
37 194 search one will be eligible. Studies that provide indirect evidence on the measurement properties (by  
38 195 testing an alternative test against an outcome measure of interest, studies in which the outcome  
39 196 measure is used to measure an outcome) will be excluded. Also, studies that provide normative data  
40 197 will be excluded.  
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47 198 **Information sources**  
48

49 199 The search strategy has been developed using medical subject headings, and relevant text words  
50 200 relating to AIS, physical functioning, outcome measures and measurement properties using the  
51 201 adapted search filter. [33] The electronic search of databases will be conducted including MEDLINE  
52 202 (1946- November 2019), PsycINFO (1967- December 2019), EMBASE (1974- December 2019), CINHAL  
53 203 (1937- December 2019), SPORTdiscus (1800- December 2019), Web of Science (1900- December  
54 204 2019) and PubMed (1997- December 2019). No language limitations will be applied in the first search;  
55 205 however, the second search will be limited to the full-text article in English. A hand search in the key  
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3 206 journals including Spine, The Spine Journal, Spine Deformity, Scoliosis and Spinal Disorders and  
4  
5 207 European Spine Journal as well as contacting relevant leading researchers in the field. Further,  
6  
7 208 searching for the Grey literature, including British National bibliography for report literature, open-  
8  
9 209 Grey, dissertation abstracts and Electronic Thesis Online Service (EThOS) will be conducted. Web of  
10  
11 210 Science database will be searched for conference proceedings for the last 5 years.

### 12 13 211 **Search strategy**

14  
15 212 Following discussions with the team and specialist librarian, the search be completed by one  
16  
17 213 researcher (SA). Then, the potential studies will be independently assessed for eligibility by two  
18  
19 214 researchers (SA, EB). To ensure that all relevant data are included, no limitations will be applied. Initial  
20  
21 215 search terms will be developed for MEDLINE and then adapted with relevant syntax and subject  
22  
23 216 headings for other databases. Recommended search filters specifically designed for retrieving articles  
24  
25 217 on measurement properties will be used where appropriate.[33] An example of the search strategy of  
26  
27 218 both stages is available as an online supplementary file 1.

### 28 29 219 **Data management**

30  
31 220 Search records will be imported into Endnote Version X9 (Clarivate Analytics). Using Endnote, the  
32  
33 221 abstracts and full texts will be stored. The duplicates will be identified through the Endnote software  
34  
35 222 and exact duplicates will be removed.

### 36 37 223 **Selection Process**

38  
39 224 A standardised eligibility assessment will be performed by two independent reviewers (SA, EB). All  
40  
41 225 studies identified by the search strategy will assessed based on title/abstract for eligibility. If there is  
42  
43 226 insufficient information to include/exclude study, full text will be retrieved and then, screened for  
44  
45 227 eligibility. The Study selection (included and excluded studies) with the reasons for exclusion, will be  
46  
47 228 summarised in a PRISMA flow diagram. [31] Articles will be included if both reviewers agreed that the  
48  
49 229 eligibility criteria were met. Any disagreement will be first discussed and the third reviewer (NH) will  
50  
51 230 mediate situations of disagreement. At each assessment stage, agreement between reviewers will be  
52  
53 231 estimated with percentage of agreement and the Kappa statistic using SPSS for Windows statistical  
54  
55 232 software package (IBM SPSS Statistics Version 25).

### 56 57 233 **Data collection process**

58  
59 234 Two reviewers (SA, EB) will independently extract the data of eligible studies. A bespoke data  
60  
235 extraction form will be used and piloted on 3 studies. Any disagreement between reviewers will be

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

### 239 Data items

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

Table 1: Summary of items to be extracted from included studies	
<b>Study &amp; Participants Characteristics</b>	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)
<b>Outcome measure</b>	<b>PROM:</b> 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
	<b>PBOM:</b> Name of outcome measure, equipment needed, number of assessments, outcome (e.g. time needed, ability/disability), setting, scoring
	<b>Body structure and function measure:</b> Name of outcome measure, equipment needed, mode of administration, setting, scoring, outcome (e.g. time needed, ability/disability)
<b>Measurement properties</b>	<b>Validity:</b> Name of outcome measure, type of validity, descriptive statistics, missing value, comparator outcome or predictor outcome, statistics method, confidence interval, validation results
	<b>Reliability:</b> Name of outcome measure, type of reliability, descriptive statistic, time interval, reliability coefficient, measurement error
	<b>Responsiveness:</b> Name of outcome measure, Method of testing: <i>Hypothesis testing, Distribution based method</i> (ES, SRM and MDC), time to follow-up. <i>Anchor-based methods</i> (MIC or MCIC or MID).
	<b>Interpretability:</b> : 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
	<b>Feasibility:</b> 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	

## 242 **Outcomes and prioritisation**

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

## 246 **Risk of bias in individual studies**

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

## 262 **Data synthesis**

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

1  
2  
3 272 the a modified Grading of Recommendations Assessment, Development and Evaluation (GRADE)  
4  
5 273 approach. [36]  
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7

8 274 The results on measurement properties from different studies will be pooled in a meta-analysis if  
9  
10 275 there is sufficient clinical and methodological homogeneity. [36] The data will be statistically pooled  
11  
12 276 when: (1): Individuals with AIS displayed similar characteristics in terms of curve severity, intervention.  
13  
14 277 (2): similar base-line score (3): Same time interval (4): Same statistical parameters. [36] If inconsistent  
15  
16 278 results of measurement properties were presents due to different subgroups (i.e. mild and sever  
17  
18 279 curve), the consistent results will be separately summarized per subgroup. [36] Pooled estimate of  
19  
20 280 measurement properties will be obtained by calculating weighted means and 95% confidence interval.  
21  
22 281 [36] If deemed not possible to pool the results, a qualitative synthesis will be conducted e.g. the  
23  
24 282 percentage of confirmed hypotheses for construct validity will be provided. [36] The pooled or  
25  
26 283 summarized evidence will be rated as sufficient when at least 75% of the results met the criteria. [36]  
27  
28 284 For example, for structural validity, “at least 75% of the confirmatory factor analysis studies should  
29  
30 285 found the same factor structure”. [36]  
31  
32

33  
34 286 The recommendation of an outcome measure will be depending on the measurement properties, as  
35  
36 287 well as interpretability and feasibility results. [36] The tool should have sufficient content validity and  
37  
38 288 at least low quality evidence for sufficient internal consistency to be recommended for use and the  
39  
40 289 results of this tool is trustworthy. [36]  
41  
42

### 43 290 **Confidence in cumulative evidence**

44  
45 291 The overall quality and strength of evidence will be assessed for pooled or summarised result for each  
46  
47 292 measurement property per outcome measure per category by two reviewers, independently. Using a  
48  
49 293 modified (GRADE) approach. [36] The GRADE approach uses five factors to determine the quality of  
50  
51 294 the evidence: risk of bias (quality of the studies), inconsistency (of the results of the studies),  
52  
53 295 indirectness (evidence comes from different populations, interventions or outcomes than the ones of  
54  
55 296 interest in the review), imprecision (wide confidence intervals), and publication bias (negative results  
56  
57 297 are less often published).[37] For evaluating measurement properties in systematic reviews of PROMs,  
58  
59 298 only four factors will be assessed as recommended by COSMIN group, while the fifth factor  
60  
61 299 (publication bias) was removed as there is no registry exists for measurement properties. [36]  
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## 304 **Discussion**

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

## 322 **Patient and public involvement**

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

## 330 **Implications of this study**

331 AIS is a complex deformity of the spine and causes a significant impact on physical activities of  
332 individuals' daily living such as walking and maintaining body position. [7, 17] In consequence, the  
333 quality of life is affected. Physical functioning gives an indication about the current health status and  
334 identifies people at risk of disability. [12, 13] Therefore, physical functioning is considered as one of  
335 the outcomes that should be assessed and reported in clinical trials of musculoskeletal conditions.  
336 [15] A systematic review is needed to evaluate current practice in assessment of physical functioning  
337 among individuals with AIS. The results of this review will inform clinicians and researchers on the best



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2  
3 338 available tools for assessment of physical functioning in AIS. This review could provide a research  
4  
5 339 agenda that may highlight the gap in the literature around physical functioning measure and their  
6  
7 340 measurement properties among individuals with AIS.

8 341

9  
10 342 **Declarations**

11  
12 343 **Ethics and Dissemination**

13  
14 344 No ethics approval is required for this systematic review. The results of this systematic review will be  
15  
16 345 disseminated through peer-reviewed journals as well as international and national conferences  
17  
18 346 presentation. The publications will be split into different publications according to the volume of data.  
19  
20 347 Each category of outcome measures will be published in a separate article.

21 348

22  
23 349 **Author Contributions**

24  
25 350 All authors conceptualized and designed the protocol. SA is a PhD student and NH (lead supervisor),  
26  
27 351 AR and DF are supervisors and AG spinal surgeon. SA drafted the initial manuscript with NH, AR DF,  
28  
29 352 and AG providing guidance on design, topic, methodology and analyses. All authors reviewed and  
30  
31 353 commented on each draft of the protocol. All authors have approved and contributed to the final  
32  
33 354 manuscript.

34  
35  
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39  
40 357 form University of Tabuk, Tabuk, Saudi Arabia.

41  
42  
43 358 **Competing interests**

44  
45 359 None declared.

46  
47 360 **Patient consent for publication**

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49 361 Not required.

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## 365 REFERENCES

- 366 1 Reamy BV, Slakey JB. Adolescent idiopathic scoliosis: review and current concepts. *Am*  
367 *Fam Physician* 2001;**64**:111-6.
- 368 2 Cobb JR. The problem of the primary curve. *J Bone Joint Surg Am* 1960;**42-A**:1413-25.
- 369 3 Konieczny MR, Senyurt H, Krauspe R. Epidemiology of adolescent idiopathic scoliosis.  
370 *J Child Orthop* 2013;**7**:3-9.
- 371 4 Hamad A, Ahmed EB, Tsirikos AI. Adolescent idiopathic scoliosis: a comprehensive  
372 approach to aetiology, diagnostic assessment and treatment. *Orthopaedics and Trauma*  
373 2017;**31**:343-9.
- 374 5 Horne JP, Flannery R, Usman S. Adolescent idiopathic scoliosis: diagnosis and  
375 management. *Am Fam Physician* 2014;**89**:193-8.
- 376 6 Lonstein D. Adolescent idiopathic scoliosis. *The Lancet* 1994;**344**:1407-12.
- 377 7 Du C, Yu J, Zhang J, *et al*. Relevant areas of functioning in patients with adolescent  
378 idiopathic scoliosis on the International Classification of Functioning, Disability and Health:  
379 The patients' perspective. *J Rehabil Med* 2016;**48**:806-14.
- 380 8 Makino T, Kaito T, Kashii M, *et al*. Low back pain and patient-reported QOL outcomes  
381 in patients with adolescent idiopathic scoliosis without corrective surgery. *Springerplus*  
382 2015;**4**:397.
- 383 9 Durmala J, Tomalak W, Kotwicki T. Function of the respiratory system in patients with  
384 idiopathic scoliosis: reasons for impairment and methods of evaluation. *Stud Health Technol*  
385 *Inform* 2008;**135**:237-45.
- 386 10 Leszczewska J, Czaprowski D, Pawlowska P, *et al*. Evaluation of the stress level of  
387 children with idiopathic scoliosis in relation to the method of treatment and parameters of  
388 the deformity. *ScientificWorldJournal* 2012;**2012**:538409.

- 1  
2  
3 389 11 Malmqvist M, Tropp H, Lyth J, *et al.* Patients With Idiopathic Scoliosis Run an Increased  
4  
5 390 Risk of Schizophrenia. *Spine Deform* 2019;**7**:262-6.  
6  
7  
8 391 12 Taylor AM, Phillips K, Patel KV, *et al.* Assessment of physical function and participation  
9  
10 392 in chronic pain clinical trials: IMMPACT/OMERACT recommendations. *Pain* 2016;**157**:1836-  
11  
12 393 50.  
13  
14  
15 394 13 Tomey KM, Sowers MR. Assessment of physical functioning: a conceptual model  
16  
17 395 encompassing environmental factors and individual compensation strategies. *Phys Ther*  
18  
19 396 2009;**89**:705-14.  
20  
21  
22 397 14 Cooper R, Kuh D, Cooper C, *et al.* Objective measures of physical capability and  
23  
24 398 subsequent health: a systematic review. *Age Ageing* 2011;**40**:14-23.  
25  
26  
27 399 15 Dodd S, Clarke M, Becker L, *et al.* A taxonomy has been developed for outcomes in  
28  
29 400 medical research to help improve knowledge discovery. *J Clin Epidemiol* 2018;**96**:84-92.  
30  
31  
32 401 16 de Kleuver M, Faraj SSA, Holewijn RM, *et al.* Defining a core outcome set for  
33  
34 402 adolescent and young adult patients with a spinal deformity. *Acta Orthop* 2017;**88**:612-8.  
35  
36  
37 403 17 Kibsgard T, Brox JI, Reikeras O. Physical and mental health in young adults operated  
38  
39 404 on for idiopathic scoliosis. *J Orthop Sci* 2004;**9**:360-3.  
40  
41  
42 405 18 Bastrom TP, Marks MC, Yaszay B, *et al.* Prevalence of postoperative pain in adolescent  
43  
44 406 idiopathic scoliosis and the association with preoperative pain. *Spine (Phila Pa 1976)*  
45  
46 407 2013;**38**:1848-52.  
47  
48  
49 408 19 Seki H, Ideno S, Ishihara T, *et al.* Postoperative pain management in patients  
50  
51 409 undergoing posterior spinal fusion for adolescent idiopathic scoliosis: a narrative review.  
52  
53 410 *Scoliosis Spinal Disord* 2018;**13**:17.  
54  
55  
56 411 20 Ward MM. Interpreting measurements of physical function in clinical trials. *Ann*  
57  
58 412 *Rheum Dis* 2007;**66 Suppl 3**:iii32-4.  
59  
60

- 1  
2  
3 413 21 Reiman MP, Manske RC. The assessment of function: How is it measured? A clinical  
4  
5  
6 414 perspective. *J Man Manip Ther* 2011;**19**:91-9.
- 7  
8 415 22 Haher TR, Gorup JM, Shin TM, *et al.* Results of the Scoliosis Research Society  
9  
10 416 instrument for evaluation of surgical outcome in adolescent idiopathic scoliosis. A multicenter  
11  
12 417 study of 244 patients. *Spine (Phila Pa 1976)* 1999;**24**:1435-40.
- 13  
14  
15 418 23 Baldus C, Bridwell K, Harrast J, *et al.* The Scoliosis Research Society Health-Related  
16  
17 419 Quality of Life (SRS-30) age-gender normative data: an analysis of 1346 adult subjects  
18  
19 420 unaffected by scoliosis. *Spine (Phila Pa 1976)* 2011;**36**:1154-62.
- 20  
21  
22 421 24 Chen AF, Bi W, Singhabahu D, *et al.* Converting Scoliosis Research Society-24 to  
23  
24 422 Scoliosis Research Society-22r in a Surgical-Range, Medical/Interventional Adolescent  
25  
26 423 Idiopathic Scoliosis Patient Cohort. *Spine Deform* 2013;**1**:108-14.
- 27  
28  
29 424 25 Asher M, Min Lai S, Burton D, *et al.* Discrimination validity of the scoliosis research  
30  
31 425 society-22 patient questionnaire: relationship to idiopathic scoliosis curve pattern and curve  
32  
33 426 size. *Spine (Phila Pa 1976)* 2003;**28**:74-8.
- 34  
35  
36 427 26 Asher MA, Min Lai S, Burton DC. Further development and validation of the Scoliosis  
37  
38 428 Research Society (SRS) outcomes instrument. *Spine (Phila Pa 1976)* 2000;**25**:2381-6.
- 39  
40  
41 429 27 Bean JF, Olveczky DD, Kiely DK, *et al.* Performance-based versus patient-reported  
42  
43 430 physical function: what are the underlying predictors? *Phys Ther* 2011;**91**:1804-11.
- 44  
45  
46 431 28 Mokkink LB, de Vet HCW, Prinsen CAC, *et al.* COSMIN Risk of Bias checklist for  
47  
48 432 systematic reviews of Patient-Reported Outcome Measures. *Qual Life Res* 2018;**27**:1171-9.
- 49  
50  
51 433 29 Mokkink LB, Terwee CB, Patrick DL, *et al.* The COSMIN study reached international  
52  
53 434 consensus on taxonomy, terminology, and definitions of measurement properties for health-  
54  
55 435 related patient-reported outcomes. *J Clin Epidemiol* 2010;**63**:737-45.
- 56  
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2  
3 436 30 Mokkink LB, Prinsen C, Patrick DL, *et al.* COSMIN methodology for systematic reviews  
4  
5  
6 437 of Patient-Reported outcome measures (PROMs).  
7  
8 438 31 Moher D, Shamseer L, Clarke M, *et al.* Preferred reporting items for systematic review  
9  
10 439 and meta-analysis protocols (PRISMA-P) 2015 statement. *Syst Rev* 2015;**4**:1.  
11  
12  
13 440 32 Williamson PR, Altman DG, Bagley H, *et al.* The COMET Handbook: version 1.0. *Trials*  
14  
15 441 2017;**18**:280.  
16  
17  
18 442 33 Terwee CB, Jansma EP, Riphagen, II, *et al.* Development of a methodological PubMed  
19  
20 443 search filter for finding studies on measurement properties of measurement instruments.  
21  
22 444 *Qual Life Res* 2009;**18**:1115-23.  
23  
24  
25 445 34 Mokkink LB, Terwee CB, Patrick DL, *et al.* The COSMIN checklist for assessing the  
26  
27 446 methodological quality of studies on measurement properties of health status measurement  
28  
29 447 instruments: an international Delphi study. *Qual Life Res* 2010;**19**:539-49.  
30  
31  
32 448 35 Mokkink LB, Prinsen CA, Patrick DL, *et al.* COSMIN methodology for systematic reviews  
33  
34 449 of Patient-Reported Outcome Measures (PROMs).  
35  
36  
37 450 36 Prinsen CAC, Mokkink LB, Bouter LM, *et al.* COSMIN guideline for systematic reviews  
38  
39 451 of patient-reported outcome measures. *Qual Life Res* 2018;**27**:1147-57.  
40  
41  
42 452 37 Balshem H, Helfand M, Schunemann HJ, *et al.* GRADE guidelines: 3. Rating the quality  
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44 453 of evidence. *J Clin Epidemiol* 2011;**64**:401-6.  
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For peer review only

**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.

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- 4 33. exp sitting/
- 5 34. exp Walking/
- 6 35. exp Walking Speed/
- 7 36. exp Postural Balance/
- 8 37. Standing balance.mp.
- 9
- 10 38. exp Hand Strength/ or Grip strength.mp.
- 11
- 12 39. 1- 3/OR
- 13 40. 4 OR 5
- 14 41. 6-38/OR
- 15 42. 39 and 40 and 41
- 16 43. Limit 42 to humans
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## 21 **Stage 2**

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- 24 44. Name of the Identified outcome measure
- 25 45. validity.mp
- 26 46. exp validation studies/
- 27 47. reliability.mp
- 28 48. exp reproducibility of results/
- 29 49. interpretability.mp
- 30 50. internal consistency.mp
- 31 51. exp sensitivity and Specificity/
- 32 52. clinical sensitivity.mp
- 33 53. exp psychometrics/
- 34 54. responsiveness.mp
- 35 55. exp Evaluation studies/
- 36 56. measurement error.mp
- 37 57. measurement properties.mp
- 38 58. 46-58/OR
- 39 59. 44 AND 58
- 40 60. Limit 59 to humans
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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	Reported on Page
<b>ADMINISTRATIVE INFORMATION</b>			
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	Page 13 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	Page 13 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
<b>INTRODUCTION</b>			
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
<b>METHODS</b>			
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 $\tau$ )	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.*

# BMJ Open

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

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<b>Primary Subject Heading</b>:	Rehabilitation medicine
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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3 1 **Outcome Measures Evaluating Physical Functioning and their Measurement Properties in**  
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5 2 **Adolescent Idiopathic Scoliosis: Protocol for a Systematic Review**  
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8 3 **Authors**  
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3 29 **ABSTRACT**  
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6 30 **Introduction** Physical functioning (PF) is the ability to carry out physical activity of daily living. It is an  
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8 31 important outcome that provide meaningful evaluation of individuals' life. PF can be assessed using  
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10 32 Patient-Reported outcome measures, Performance-Based Outcome Measures or Body Structure and  
11  
12 33 Function Measure. Measures need to be valid, reliable and responsive to change to evaluate effects  
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14 34 of an intervention. Adolescent Idiopathic Scoliosis (AIS) is the most common deformity among  
15  
16 35 the paediatric population. It causes significant impact on individuals' life. This systematic review will  
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18 36 appraise evidence on the measurement properties of PF tools in individuals with AIS.

19 37 **Methods/analysis** A protocol for systematic review and meta-analysis informed by Cochrane  
20  
21 38 guidelines is reported in line with Preferred Reporting Items for Systematic Reviews and Meta-  
22  
23 39 Analysis-P. MEDLINE, PsycINFO, EMBASE, CINAHL, SPORTdiscus, Web of science and PubMed will be  
24  
25 40 searched in two stages, from inception until December 2019. Search one will inventory all studies that  
26  
27 41 assessed PF in participants with AIS, without any limitations. The Search terms will be Scoliosis,  
28  
29 42 Adolescent and PF related terms. Search two will include studies which investigated instrument  
30  
31 43 measurement properties in the same population for measures identified in search one. Two reviewers  
32  
33 44 will independently perform study selection, data extraction, risk of bias, and overall quality  
34  
35 45 assessment. The Consensus-based Standards for the selection of Health Measurement Instruments  
36  
37 46 risk of bias and a modified Grading of Recommendations, Assessment, Development and Evaluation  
38  
39 47 guidelines will be used. Meta-analysis will be conducted if possible, or the evidence will be synthesised  
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41 48 and summarized per measurement property per outcome measure per measurement type.

42 49 **Ethics and dissemination:** This review will provide recommendations for practice and future  
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44 50 research, considering psychometric properties of outcome measures of PF in AIS. The results of this  
45  
46 51 study will be disseminated through a peer-reviewed publication and conference presentation.

47 52 **Prospero registration number:** CRD42019142335

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49 53 **Keywords:** Adolescent Idiopathic Scoliosis, measurement properties, physical function, systematic  
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51 54 review  
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3 58 **ARTICLE SUMMARY**  
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6 59 **Strengths and Limitations of this Study**  
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- 9 61 1. This review will synthesise evidence of patient-reported, performance-based or, body  
10 62 structure and function outcome measures of physical functioning, for use in practice or  
11 63 research involving individuals with AIS.  
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13 64 2. The search strategy of this review comprises two stages. The first stage will retrieve all studies  
14 65 that assessed physical functioning in individuals with AIS, while the Second stage will retrieve  
15 66 studies that investigated measurement properties of the instrument identified in the first  
16 67 search.  
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18 68 3. This study will employ rigorous methods and uses COnsensus-based Standards for the  
19 69 selection of health Measurement Instruments risk of bias tool and modified Grading of  
20 70 Recommendations, Assessment, Development and Evaluation approach.  
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22 71 4. This review will be limited to studies of the English language that assess measurement  
23 72 properties among adolescents with idiopathic scoliosis.  
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## 87 INTRODUCTION

88 Scoliosis is a complex three-dimensional deformity of the spine, including lateral curvature and  
89 rotation of the vertebrae, [1] and characterised by a curve angle  $\geq 10^\circ$ . [2] There are two main types  
90 of scoliosis, idiopathic and non-idiopathic with the latter arising from congenital, neuromuscular or  
91 mesenchymal causes. [3] While the aetiology of idiopathic scoliosis remains unknown; genetic,  
92 hormonal, and mechanical factors are involved. [4] Adolescent Idiopathic Scoliosis (AIS) often  
93 develops between 10 and 16 years of age and represents ~85% of cases. [5] AIS is the most common  
94 spinal deformity among the pediatric population, with a prevalence ranging from 2% to 3%. [6] Nearly  
95 80% of those affected presents with a curvature of the thoracic or thoracolumbar/lumbar region.[3]  
96 Whilst males and females are equally affected, females are reported to be at 10 times greater risk of  
97 curve progression. [1]

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

109 Physical functioning can be assessed with Patient-Reported Outcome Measures (PROMs),  
110 Performance-Based Outcome Measures (PBOMs) [20] or a measure of body structure and function.  
111 The most widely used PROM for assessment of the quality of life as well as physical functioning of  
112 individuals with spinal deformity is the Scoliosis Research Society (SRS) questionnaire, [21] and its  
113 variants. [22-25] The SRS is mostly used among surgically treated individuals with AIS, [21, 24, 25] but  
114 may not be applicable to those treated conservatively. [25] Although relevant, PROMs should be used  
115 cautiously; as it influenced by patients' perception of their abilities to perform activities and lack  
116 sensitivity to change. [26] Measures such as PBOMs have the potential to provide unbiased and  
117 reproducible assessments of physical functioning during the performance of activities of daily living,  
118 [26, 27] such as walking speed and trunk endurance testing. [26] Within individuals with AIS, little is  
119 known about the available PBOMs for evaluating physical functioning. The body structure and



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3 120 function measures such as radiographs can give an indication about dysfunctions in structure but fail  
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5 121 to fully capture functional limitations. [26]  
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8 122 The SRS-22r questionnaire is the gold standard outcome measure of quality of life, which include  
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10 123 physical functioning items as recommended by the recent COS study for adolescents and young adults  
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12 124 with spinal deformity. [16] However, the SRS-22r fails to fully capture important aspects of physical  
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14 125 functioning for individuals with AIS e.g. self-care and mobility. [7] The COS study included all forms of  
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16 126 spinal deformities, the heterogeneity limits applicability to individuals with AIS as a discrete  
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18 127 population.

19 128 Adequate measurement properties of outcome measures are important to avoid the risk of bias and  
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21 129 ensure accuracy in the evaluation of test results.[28] The COnsensus-based Standards for the selection  
22  
23 130 of health Measurement INstruments, (COSMIN) group developed a taxonomy of measurement  
24  
25 131 properties to improve the selection of outcome measures. [29] Three main domains identified  
26  
27 132 reliability, validity and responsiveness with further subgrouping.[28] A systematic review is needed to  
28  
29 133 evaluate the measurement properties, of physical functioning outcome measure for individuals with  
30  
31 134 AIS. Review findings will inform clinicians and researchers on the best available tools for the  
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33 135 assessment of physical functioning in AIS. Furthermore, findings will inform future research drawing  
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35 136 on a range of measures of physical functioning to investigate health status in AIS.

### 35 137 **Objective**

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38 138 To identify outcome measures used to assess physical functioning in individuals with AIS. A secondary  
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40 139 aim is to evaluate the measurement properties of physical functioning outcome measures in AIS.

### 41 42 140 **METHODS**

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45 141 This protocol has been informed by experts in musculoskeletal orthopaedics including a consultant  
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47 142 spine surgeon, musculoskeletal rehabilitation experts including physiotherapists and individuals with  
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49 143 review, measurement properties and research experience. It has been designed in line with the  
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51 144 COSMIN methodology for systematic reviews of Patient-Reported Outcome Measures [30] and is  
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53 145 reported in line with the Preferred Reporting Items for Systematic Reviews and Meta-Analysis-P  
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55 146 (PRISMA-P) [31] The search for this systematic review will be conducted in two parts. Stage one to  
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57 147 identify studies used an outcome measure to evaluate physical functioning in individuals with AIS. This  
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59 148 search will allow a list of outcome measures to be generated. Stage 2 will identify studies, which  
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3 149 evaluated measurement properties of physical functioning outcome measure identified in the first  
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5 150 search.

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8 151 **Stage one: Inventory of outcome measure**

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10 152 **Eligibility criteria**

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13 153 *Study design*

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15 154 All study designs including; randomised clinical trials, cohort, observational studies and case studies  
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17 155 will be included to identify all outcome measure of physical functioning being used with individuals  
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19 156 with AIS. No limitation on language or location will be applied at this stage.

20  
21 157 *Participants*

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24 158 Participants aged between the age of 10 years to 18 years of age, with a diagnosis of idiopathic  
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26 159 scoliosis and  $\geq 10^\circ$  Cobb angle will be considered. No restrictions will be applied to the curve severity,  
27  
28 160 evaluation settings, and the type of treatment.

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30 161 *Outcome*

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32 162 Any study that includes assessments of the physical functioning of AIS using a specific outcome  
33  
34 163 measure will be included. Physical functioning is defined according to the Core Outcome Measures in  
35  
36 164 Effectiveness Trials (COMET) taxonomy, [15] as any physical activities of daily living such as the ability  
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38 165 to walk, independence, self-care, performance status, disability index. [15, 32] The outcome measures  
39  
40 166 are defined as any one of the following:

- 41  
42 167 1. Patient-Reported Outcome Measures (PROMs) in the form of questionnaires or scales  
43  
44 168 designed for AIS to evaluate physical functioning or if it is included as a sub-scale within a  
45  
46 169 questionnaire.
- 47 170 2. Performance-based outcome measures (PBOMs); a measure of physical functioning by  
48  
49 171 clinician while the individual is performing a functional task e.g walking, and/or
- 50 172 3. Body structure and function measures which means any dysfunction in a specific body part or  
51  
52 173 system which may limits function, such as range of motion. [26]

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## 176 **Search strategy**

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

## 186 **Stage two: measurement properties**

### 187 **Eligibility criteria**

#### 188 *Study design*

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

#### 193 *Participants*

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

#### 199 *Outcome*

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

1  
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3 205 outcome measure is used to measure an outcome) will be excluded. Also, studies that only provide  
4  
5 206 normative data will be excluded.  
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### 8 207 **Search strategy**

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10 208 Using the list of outcome measures determined from the search in stage one, one reviewer (SA) will  
11 209 conduct the search. Each category of outcome measure will be searched separately. The search terms  
12 210 will be consisting of the name of the outcome measure/s, the AIS and the measurement properties  
13  
14 211 (Figure 1). The recommended search filters specifically designed for retrieving articles on  
15  
16 212 measurement properties will be adapted and used at this stage [33]. An example of the search strategy  
17  
18 213 of stage 2 is available as an online supplementary file 1.  
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### 21 214 **Information sources**

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24 215 The electronic search of databases will be conducted including MEDLINE (1946- November 2019),  
25 216 PsycINFO (1967- December 2019), and EMBASE (1974- December 2019) through OVID interface,  
26 217 CINHAL (1937- December 2019) and SPORTdiscus (1800- December 2019) through EBSCO interface,  
27 218 Web of Science (1900- December 2019) and PubMed (1997- December 2019). No language limitations  
28 219 will be applied in the first search; however, the second search will be limited to the full-text article in  
29 220 English. The Web of Science database will be searched for conference proceedings for the last 5 years  
30 221 for the first search only. A hand search in the key journals including Spine, The Spine Journal, Spine  
31 222 Deformity, Scoliosis and Spinal Disorders and European Spine Journal as well as contacting relevant  
32 223 leading researchers in the field. Further, searching for the Grey literature, including British National  
33 224 bibliography for report literature, open-Grey, dissertation abstracts and Electronic Thesis Online  
34 225 Service (ETHOS) will be conducted.  
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### 44 226 **Data management**

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47 227 Search records will be imported into Endnote Version X9 (Clarivate Analytics). Using Endnote, the  
48 228 abstracts and full texts will be stored. The duplicates will be identified through the Endnote software  
49 229 and exact duplicates will be removed.  
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### 53 230 **Selection Process**

54  
55 231 A standardised eligibility assessment will be performed by two independent reviewers (SA, EB). All  
56 232 studies identified by the search strategy will assessed based on title/abstract for eligibility. If there is  
57 233 insufficient information to include/exclude study, full text will be retrieved and then, screened for  
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234 eligibility. The study selection (included and excluded studies) with the reasons for exclusion, will be  
 235 summarised in a PRISMA flow diagram. [31] Articles will be included if both reviewers agreed that the  
 236 eligibility criteria were met. Any disagreement will be first discussed and the third reviewer (NH) will  
 237 mediate situations of disagreement. At each assessment stage, agreement between reviewers will be  
 238 estimated with percentage of agreement and the Kappa statistic using SPSS for Windows statistical  
 239 software package (IBM SPSS Statistics Version 25).

#### 240 **Data collection process**

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

#### 246 **Data items**

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

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

<b>Study &amp; Participants Characteristics</b>	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)
<b>Outcome measure</b>	<b>PROM:</b> 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
	<b>PBOM:</b> Name of outcome measure, equipment needed, number of assessments, outcome (e.g. time needed, ability/disability), setting, scoring
	<b>Body structure and function measure:</b> Name of outcome measure, equipment needed, mode of administration, setting, scoring, outcome (e.g. time needed, ability/disability)
<b>Measurement properties</b>	<b>Validity:</b> Name of outcome measure, type of validity, descriptive statistics, missing value, comparator outcome or predictor outcome, hypothesis, statistics method, confidence interval, validation results
	<b>Reliability:</b> Name of outcome measure, type of reliability, descriptive statistic, time interval, reliability coefficient, measurement error

	<b>Responsiveness:</b> Name of outcome measure, Method of testing: <i>Hypothesis testing, Distribution based method</i> (ES, SRM and MDC), hypothesis, time to follow-up. <i>Anchor-based methods</i> (MIC or MCIC or MID), anchor/s.
	<b>Interpretability:</b> 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
	<b>Feasibility:</b> 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	

## 249 Outcomes and prioritisation

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

## 253 Risk of bias in individual studies

254 The COSMIN checklist for assessment of risk of bias and methodological quality in individual studies  
 255 will be used. [28] It was revised and specifically designed for use in systematic reviews of PROMs to  
 256 evaluate studies on measurement properties. [34] The methodological quality of each study for each  
 257 measurement property will be assessed separately. [30] The items for each measurement property in  
 258 the relevant standards box will be rated as either very good, adequate, doubtful or inadequate quality.  
 259 [30] Then, the overall methodological quality of the measurement property will be rated based on  
 260 "the worst score counts principle" i.e. that the overall quality of the study for a specific measurement  
 261 property is based on the lowest rating of any items in the standards' box. [30] The result of each item  
 262 and overall rating will be reported in the final results. The COSMIN group recommend researchers to  
 263 adapt the checklist to other measures (i.e. PBOMs, body structure and function measure) since it was  
 264 originally developed for PROMs. [30] Two independent reviewers (SA, EB) will assess the risk of bias  
 265 for all included studies. Any disagreement will be resolved through discussion, and if no agreement is  
 266 reached, a third reviewer (NH) will be consulted. The agreement between reviewers will be estimated  
 267 with percentage agreement and the Kappa statistic using SPSS for Windows statistical software  
 268 package (IBM SPSS Statistics Version 25) and will be reported in the final results.

269

## 270 **Data synthesis**

271 The COSMIN guidelines for systematic reviews will be followed for the synthesis of the results. [30]  
272 Characteristics of the outcome measures, sample, measurement properties results, information about  
273 interpretability and feasibility of the scores of the included outcome measures will be presented in  
274 overview tables for each outcome measure. [30] Each measurement property for each study per tool  
275 will be rated against the updated criteria for good measurement properties as either sufficient (+),  
276 insufficient (-), or indeterminate (?).[30] The result of rating of measurement property and its  
277 methodological quality rating will be added to the overview table. [30] Then, the evidence will be  
278 pooled or summarized per measurement property per tool, with the overall result will be rated against  
279 the criteria for good measurement properties, and the quality of the evidence will be graded using a  
280 modified Grading of Recommendations Assessment, Development and Evaluation (GRADE) approach.  
281 [30]

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

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

## 298 **Confidence in cumulative evidence**

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

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3 301 measure in each category will be evaluated. The GRADE approach uses five factors to determine the  
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5 302 quality of the evidence: risk of bias (quality of the studies), inconsistency (of the results of the studies),  
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7 303 indirectness (evidence comes from different populations, interventions or outcomes than the ones of  
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9 304 interest in the review), imprecision (wide confidence intervals), and publication bias (negative results  
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11 305 are less often published).[35] For evaluating measurement properties in systematic reviews of PROMs,  
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13 306 only four factors will be assessed as recommended by COSMIN group, while the fifth factor  
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15 307 (publication bias) will be removed as there is no registry exists for measurement properties.  
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## 309 **Discussion**

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311 Physical functioning is considered as an important outcome domain in health-related quality of life.  
312 [12] It can be used to predict future disability as well as health and social care use.[13] Individuals with  
313 AIS reported a limitation in their physical functioning [7]. Thus, measurement of its impact is important  
314 in research and clinical practice. Numerous of tools are available for the assessment of physical  
315 functioning, ranging from patient-reported to performance-based measures. However, it is essential  
316 to confirm the psychometric properties of these tools before recommending for clinical use. The COS  
317 study for 'all spine deformities' identified the SRS-22r as the recommended PROM for assessment for  
318 physical functioning among young adults with spinal deformities. [16] However, there is still a need  
319 for a more specific review that evaluate the quality of all outcome measures used in the assessment  
320 of physical functioning in AIS including patient-reported, and performance-based as well as measures  
321 of body structure and function. This systematic review will retrieve all tools that have been used to  
322 assess physical functioning among individuals with AIS. Then, it will evaluate and synthesise the quality  
323 of studies that report psychometric properties of physical functioning outcome measures in AIS. This  
324 review will provide a comprehensive assessment of current evidence which may benefit: (1) health  
325 practitioners in selection of the most suitable tools to assess physical functioning in AIS (2) patients  
326 who need a good outcome measures that reflect their actual health status (3) researchers and policy  
327 maker who can use the recommend measures in designing research trials and defining the COS for  
328 individuals with AIS, which in turn will improve health assessment and patient care. Limitations of this  
329 review are a focus on individuals with AIS specifically, so recommendations cannot be generalised to  
330 other forms of scoliosis.

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### 335 **Patient and public involvement**

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

343

### 344 **Implications of this study**

345

346 AIS is a complex deformity of the spine and causes a significant impact on physical activities of  
347 individuals' daily living such as walking and maintaining body position. [7, 17] In consequence, the  
348 quality of life is affected. Physical functioning gives an indication about the current health status and  
349 identifies people at risk of disability. [12, 13] Therefore, physical functioning is considered as one of  
350 the outcomes that should be assessed and reported in clinical trials of musculoskeletal conditions.  
351 [15] A systematic review is needed to evaluate current practice in the assessment of physical  
352 functioning among individuals with AIS. The results of this review will inform clinicians and researchers  
353 on the best available tools for assessment of physical functioning in AIS. This review could provide a  
354 research agenda that may highlight the gap in the literature around physical functioning measure and  
355 their measurement properties among individuals with AIS.

356

### 357 **Declarations**

### 358 **Ethics and Dissemination**

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

363

### 364 **Author Contributions**

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

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3 368 commented on each draft of the protocol. All authors have approved and contributed to the final  
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5 369 manuscript.

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9  
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11  
12 372 form University of Tabuk, Tabuk, Saudi Arabia.

13  
14 373 **Competing interests**

15  
16 374 None declared.

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19 375 **Patient consent for publication**

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21 376 Not required.

22 377  
23 378 Figure 1:

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25 379 **REFERENCES**

- 26  
27  
28 380 1 Reamy BV, Slakey JB. Adolescent idiopathic scoliosis: review and current concepts. *Am*  
29  
30 381 *Fam Physician* 2001;**64**:111-6.
- 31  
32  
33 382 2 Cobb JR. The problem of the primary curve. *J Bone Joint Surg Am* 1960;**42-A**:1413-25.
- 34  
35  
36 383 3 Konieczny MR, Senyurt H, Krauspe R. Epidemiology of adolescent idiopathic scoliosis.  
37  
38 384 *J Child Orthop* 2013;**7**:3-9.
- 39  
40  
41 385 4 Hamad A, Ahmed EB, Tsirikos AI. Adolescent idiopathic scoliosis: a comprehensive  
42  
43 386 approach to aetiology, diagnostic assessment and treatment. *Orthopaedics and Trauma*  
44  
45 387 2017;**31**:343-9.
- 46  
47  
48 388 5 Horne JP, Flannery R, Usman S. Adolescent idiopathic scoliosis: diagnosis and  
49  
50 389 management. *Am Fam Physician* 2014;**89**:193-8.
- 51  
52  
53 390 6 Lonstein D. Adolescent idiopathic scoliosis. *The Lancet* 1994;**344**:1407-12.
- 54  
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- 1  
2  
3 391 7 Du C, Yu J, Zhang J, *et al.* Relevant areas of functioning in patients with adolescent  
4  
5  
6 392 idiopathic scoliosis on the International Classification of Functioning, Disability and Health:  
7  
8 393 The patients' perspective. *J Rehabil Med* 2016;**48**:806-14.  
9  
10 394 8 Makino T, Kaito T, Kashii M, *et al.* Low back pain and patient-reported QOL outcomes  
11  
12  
13 395 in patients with adolescent idiopathic scoliosis without corrective surgery. *Springerplus*  
14  
15 396 2015;**4**:397.  
16  
17  
18 397 9 Durmala J, Tomalak W, Kotwicki T. Function of the respiratory system in patients with  
19  
20 398 idiopathic scoliosis: reasons for impairment and methods of evaluation. *Stud Health Technol*  
21  
22 399 *Inform* 2008;**135**:237-45.  
23  
24  
25 400 10 Leszczewska J, Czaprowski D, Pawlowska P, *et al.* Evaluation of the stress level of  
26  
27 401 children with idiopathic scoliosis in relation to the method of treatment and parameters of  
28  
29 402 the deformity. *Scientific World Journal* 2012;**2012**:538409.  
30  
31  
32 403 11 Malmqvist M, Tropp H, Lyth J, *et al.* Patients With Idiopathic Scoliosis Run an Increased  
33  
34 404 Risk of Schizophrenia. *Spine Deform* 2019;**7**:262-6.  
35  
36  
37 405 12 Taylor AM, Phillips K, Patel KV, *et al.* Assessment of physical function and participation  
38  
39 406 in chronic pain clinical trials: IMMPACT/OMERACT recommendations. *Pain* 2016;**157**:1836-  
40  
41 407 50.  
42  
43  
44 408 13 Tomey KM, Sowers MR. Assessment of physical functioning: a conceptual model  
45  
46 409 encompassing environmental factors and individual compensation strategies. *Phys Ther*  
47  
48 410 2009;**89**:705-14.  
49  
50  
51  
52 411 14 Cooper R, Kuh D, Cooper C, *et al.* Objective measures of physical capability and  
53  
54 412 subsequent health: a systematic review. *Age Ageing* 2011;**40**:14-23.  
55  
56  
57 413 15 Dodd S, Clarke M, Becker L, *et al.* A taxonomy has been developed for outcomes in  
58  
59 414 medical research to help improve knowledge discovery. *J Clin Epidemiol* 2018;**96**:84-92.  
60

- 1  
2  
3 415 16 de Kleuver M, Faraj SSA, Holewijn RM, *et al.* Defining a core outcome set for  
4  
5  
6 416 adolescent and young adult patients with a spinal deformity. *Acta Orthop* 2017;**88**:612-8.  
7  
8 417 17 Kibsgard T, Brox JI, Reikeras O. Physical and mental health in young adults operated  
9  
10 418 on for idiopathic scoliosis. *J Orthop Sci* 2004;**9**:360-3.  
11  
12  
13 419 18 Bastrom TP, Marks MC, Yaszay B, *et al.* Prevalence of postoperative pain in adolescent  
14  
15 420 idiopathic scoliosis and the association with preoperative pain. *Spine (Phila Pa 1976)*  
16  
17 421 2013;**38**:1848-52.  
18  
19  
20 422 19 Seki H, Ideno S, Ishihara T, *et al.* Postoperative pain management in patients  
21  
22 423 undergoing posterior spinal fusion for adolescent idiopathic scoliosis: a narrative review.  
23  
24 424 *Scoliosis Spinal Disord* 2018;**13**:17.  
25  
26  
27 425 20 Ward MM. Interpreting measurements of physical function in clinical trials. *Ann*  
28  
29 426 *Rheum Dis* 2007;**66 Suppl 3**:iii32-4.  
30  
31  
32 427 21 Haheer TR, Gorup JM, Shin TM, *et al.* Results of the Scoliosis Research Society  
33  
34 428 instrument for evaluation of surgical outcome in adolescent idiopathic scoliosis. A multicenter  
35  
36 429 study of 244 patients. *Spine (Phila Pa 1976)* 1999;**24**:1435-40.  
37  
38  
39 430 22 Baldus C, Bridwell K, Harrast J, *et al.* The Scoliosis Research Society Health-Related  
40  
41 431 Quality of Life (SRS-30) age-gender normative data: an analysis of 1346 adult subjects  
42  
43 432 unaffected by scoliosis. *Spine (Phila Pa 1976)* 2011;**36**:1154-62.  
44  
45  
46 433 23 Chen AF, Bi W, Singhabahu D, *et al.* Converting Scoliosis Research Society-24 to  
47  
48 434 Scoliosis Research Society-22r in a Surgical-Range, Medical/Interventional Adolescent  
49  
50 435 Idiopathic Scoliosis Patient Cohort. *Spine Deform* 2013;**1**:108-14.  
51  
52  
53 436 24 Asher M, Min Lai S, Burton D, *et al.* Discrimination validity of the scoliosis research  
54  
55 437 society-22 patient questionnaire: relationship to idiopathic scoliosis curve pattern and curve  
56  
57 438 size. *Spine (Phila Pa 1976)* 2003;**28**:74-8.  
58  
59  
60

- 1  
2  
3 439 25 Asher MA, Min Lai S, Burton DC. Further development and validation of the Scoliosis  
4  
5  
6 440 Research Society (SRS) outcomes instrument. *Spine (Phila Pa 1976)* 2000;**25**:2381-6.  
7  
8 441 26 Reiman MP, Manske RC. The assessment of function: How is it measured? A clinical  
9  
10 442 perspective. *J Man Manip Ther* 2011;**19**:91-9.  
11  
12  
13 443 27 Bean JF, Olveczky DD, Kiely DK, *et al.* Performance-based versus patient-reported  
14  
15 444 physical function: what are the underlying predictors? *Phys Ther* 2011;**91**:1804-11.  
16  
17  
18 445 28 Mokkink LB, de Vet HCW, Prinsen CAC, *et al.* COSMIN Risk of Bias checklist for  
19  
20 446 systematic reviews of Patient-Reported Outcome Measures. *Qual Life Res* 2018;**27**:1171-9.  
21  
22  
23 447 29 Mokkink LB, Terwee CB, Patrick DL, *et al.* The COSMIN study reached international  
24  
25 448 consensus on taxonomy, terminology, and definitions of measurement properties for health-  
26  
27 449 related patient-reported outcomes. *J Clin Epidemiol* 2010;**63**:737-45.  
28  
29  
30 450 30 Mokkink LB, Prinsen C, Patrick DL, *et al.* COSMIN methodology for systematic reviews  
31  
32 451 of Patient-Reported outcome measures (PROMs). *User manual* 2018.  
33  
34  
35 452 31 Moher D, Shamseer L, Clarke M, *et al.* Preferred reporting items for systematic review  
36  
37 453 and meta-analysis protocols (PRISMA-P) 2015 statement. *Syst Rev* 2015;**4**:1.  
38  
39  
40 454 32 Williamson PR, Altman DG, Bagley H, *et al.* The COMET Handbook: version 1.0. *Trials*  
41  
42 455 2017;**18**:280.  
43  
44  
45 456 33 Terwee CB, Jansma EP, Riphagen, II, *et al.* Development of a methodological PubMed  
46  
47 457 search filter for finding studies on measurement properties of measurement instruments.  
48  
49 458 *Qual Life Res* 2009;**18**:1115-23.  
50  
51  
52 459 34 Mokkink LB, Terwee CB, Patrick DL, *et al.* The COSMIN checklist for assessing the  
53  
54 460 methodological quality of studies on measurement properties of health status measurement  
55  
56 461 instruments: an international Delphi study. *Qual Life Res* 2010;**19**:539-49.  
57  
58  
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60

1  
2  
3 462 35 Balshem H, Helfand M, Schunemann HJ, *et al.* GRADE guidelines: 3. Rating the quality  
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6 463 of evidence. *J Clin Epidemiol* 2011;**64**:401-6.  
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12 465 **Figure Captions**

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14 466 **Figure 1:** Flow diagram of search strategy (Search One and Two) and selection process.  
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16 467 PROM=Patient Reported Outcome Measure; PBOM= Performance-Based Outcome  
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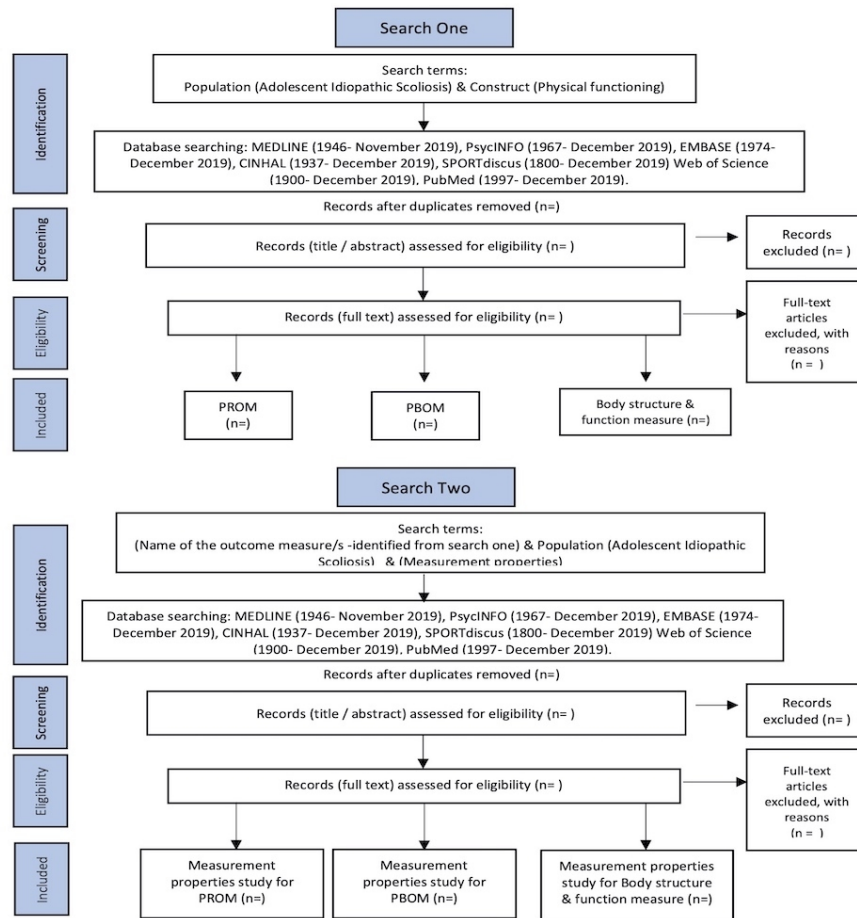


Figure 1: Flow diagram of search strategy (Search One and Two) and selection process. PROM=Patient Reported Outcome Measure; PBOM= Performance-Based Outcome Measure.

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For peer review only

**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.

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- 3
- 4 33. exp sitting/
- 5 34. exp Walking/
- 6 35. exp Walking Speed/
- 7 36. exp Postural Balance/
- 8 37. Standing balance.mp.
- 9
- 10 38. exp Hand Strength/ or Grip strength.mp.
- 11
- 12 39. 1- 3/OR
- 13 40. 4 OR 5
- 14 41. 6-38/OR
- 15 42. 39 and 40 and 41
- 16 43. Limit 42 to humans
- 17
- 18
- 19
- 20

## 21 Stage 2

- 22
- 23
- 24 44. Name of the Identified outcome measure/s
- 25 45. scoliosis.mp.
- 26 46. exp Scoliosis/
- 27 47. exp Spinal Curvatures/
- 28 48. Adolescen\$.mp.
- 29 49. exp Adolescent/
- 30 50. validity.mp
- 31 51. exp validation studies/
- 32 52. reliability.mp
- 33 53. exp reproducibility of results/
- 34 54. interpretability.mp
- 35 55. internal consistency.mp
- 36 56. exp sensitivity and Specificity/
- 37 57. clinical sensitivity.mp
- 38 58. exp psychometrics/
- 39 59. responsiveness.mp
- 40 60. exp Evaluation studies/
- 41 61. measurement error.mp
- 42 62. measurement properties.mp
- 43 63. 45-47/OR
- 44 64. 48-49/OR
- 45 65. 50-62/OR
- 46 66. 44 and 63 and 64 and 65
- 47 67. Limit 66 to humans
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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	Reported on Page
<b>ADMINISTRATIVE INFORMATION</b>			
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	Page 13 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	Page 13 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
<b>INTRODUCTION</b>			
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
<b>METHODS</b>			
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 $\tau$ )	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.*

# BMJ Open

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

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<b>Primary Subject Heading</b>:	Rehabilitation medicine
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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3 1 **Outcome Measures Evaluating Physical Functioning and their Measurement Properties in**  
4  
5 2 **Adolescent Idiopathic Scoliosis: Protocol for a Systematic Review**  
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8 3 **Authors**  
9

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3 29 **ABSTRACT**  
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6 30 **Introduction** Physical functioning (PF) is the ability to carry out physical activity of daily living. It is an  
7  
8 31 important outcome that provide meaningful evaluation of individuals' life. PF can be assessed using  
9  
10 32 Patient-Reported outcome measures, Performance-Based Outcome Measures or Body Structure and  
11  
12 33 Function Measure. Measures need to be valid, reliable and responsive to change to evaluate effects  
13  
14 34 of an intervention. Adolescent Idiopathic Scoliosis (AIS) is the most common deformity among  
15  
16 35 the paediatric population. It causes significant impact on individuals' life. This systematic review will  
17  
18 36 appraise evidence on the measurement properties of PF tools in individuals with AIS.

19 37 **Methods/analysis** A protocol for systematic review and meta-analysis informed by Cochrane  
20  
21 38 guidelines is reported in line with Preferred Reporting Items for Systematic Reviews and Meta-  
22  
23 39 Analysis-P. MEDLINE, PsycINFO, EMBASE, CINAHL, SPORTdiscus, Web of science and PubMed will be  
24  
25 40 searched in two stages, from inception until December 2019. Search one will inventory all studies that  
26  
27 41 assessed PF in participants with AIS, without any limitations. The Search terms will be Scoliosis,  
28  
29 42 Adolescent and PF related terms. Search two will include studies which investigated instrument  
30  
31 43 measurement properties in the same population for measures identified in search one. Two reviewers  
32  
33 44 will independently perform study selection, data extraction, risk of bias, and overall quality  
34  
35 45 assessment. The Consensus-based Standards for the selection of Health Measurement Instruments  
36  
37 46 risk of bias and a modified Grading of Recommendations, Assessment, Development and Evaluation  
38  
39 47 guidelines will be used. Meta-analysis will be conducted if possible, or the evidence will be synthesised  
40  
41 48 and summarized per measurement property per outcome measure per measurement type.

42 49 **Ethics and dissemination:** This review will provide recommendations for practice and future  
43  
44 50 research, considering psychometric properties of outcome measures of PF in AIS. The results of this  
45  
46 51 study will be disseminated through a peer-reviewed publication and conference presentation.

47 52 **Prospero registration number:** CRD42019142335

48  
49 53 **Keywords:** Adolescent Idiopathic Scoliosis, measurement properties, physical function, systematic  
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51 54 review  
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3 58 **ARTICLE SUMMARY**  
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6 59 **Strengths and Limitations of this Study**  
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8 60

- 9 61 1. This review will synthesise evidence of patient-reported, performance-based or, body  
10 62 structure and function outcome measures of physical functioning, for use in practice or  
11 63 research involving individuals with AIS.  
12  
13 64 2. The search strategy of this review comprises two stages. The first stage will retrieve all studies  
14 65 that assessed physical functioning in individuals with AIS, while the Second stage will retrieve  
15 66 studies that investigated measurement properties of the instrument identified in the first  
16 67 search.  
17  
18 68 3. This study will employ rigorous methods and uses COnsensus-based Standards for the  
19 69 selection of health Measurement Instruments risk of bias tool and modified Grading of  
20 70 Recommendations, Assessment, Development and Evaluation approach.  
21  
22 71 4. This review will be limited to studies of the English language that assess measurement  
23 72 properties among adolescents with idiopathic scoliosis.  
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## 87 INTRODUCTION

88 Scoliosis is a complex three-dimensional deformity of the spine, including lateral curvature and  
89 rotation of the vertebrae, [1] and characterised by a curve angle  $\geq 10^\circ$ . [2] There are two main types  
90 of scoliosis, idiopathic and non-idiopathic with the latter arising from congenital, neuromuscular or  
91 mesenchymal causes. [3] While the aetiology of idiopathic scoliosis remains unknown; genetic,  
92 hormonal, and mechanical factors are involved. [4] Adolescent Idiopathic Scoliosis (AIS) often  
93 develops between 10 and 16 years of age and represents ~85% of cases. [5] AIS is the most common  
94 spinal deformity among the pediatric population, with a prevalence ranging from 2% to 3%. [6] Nearly  
95 80% of those affected presents with a curvature of the thoracic or thoracolumbar/lumbar region.[3]  
96 Whilst males and females are equally affected, females are reported to be at 10 times greater risk of  
97 curve progression. [1]

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

109 Physical functioning can be assessed with Patient-Reported Outcome Measures (PROMs),  
110 Performance-Based Outcome Measures (PBOMs) [20] or a measure of body structure and function.  
111 The most widely used PROM for assessment of the quality of life as well as physical functioning of  
112 individuals with spinal deformity is the Scoliosis Research Society (SRS) questionnaire, [21] and its  
113 variants. [22-25] The SRS is mostly used among surgically treated individuals with AIS, [21, 24, 25] but  
114 may not be applicable to those treated conservatively. [25] Although relevant, PROMs should be used  
115 cautiously; as it influenced by patients' perception of their abilities to perform activities and lack  
116 sensitivity to change. [26] Measures such as PBOMs have the potential to provide unbiased and  
117 reproducible assessments of physical functioning during the performance of activities of daily living,  
118 [26, 27] such as walking speed and trunk endurance testing. [26] Within individuals with AIS, little is  
119 known about the available PBOMs for evaluating physical functioning. The body structure and

1  
2  
3 120 function measures such as radiographs can give an indication about dysfunctions in structure but fail  
4  
5 121 to fully capture functional limitations. [26]  
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7

8 122 The SRS-22r questionnaire is the reference standard outcome measure of quality of life, which include  
9  
10 123 physical functioning items as recommended by the recent COS study for adolescents and young adults  
11 124 with spinal deformity. [16] However, the SRS-22r fails to fully capture important aspects of physical  
12  
13 125 functioning for individuals with AIS e.g. self-care and mobility. [7] The COS study included all forms of  
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15 126 spinal deformities, the heterogeneity limits applicability to individuals with AIS as a discrete  
16  
17 127 population.  
18

19 128 Adequate measurement properties of outcome measures are important to avoid the risk of bias and  
20  
21 129 ensure accuracy in the evaluation of test results.[28] The COnsensus-based Standards for the selection  
22  
23 130 of health Measurement INstruments, (COSMIN) group developed a taxonomy of measurement  
24  
25 131 properties to improve the selection of outcome measures. [29] Three main domains identified  
26  
27 132 reliability, validity and responsiveness with further subgrouping.[28] A systematic review is needed to  
28  
29 133 evaluate the measurement properties, of physical functioning outcome measure for individuals with  
30  
31 134 AIS. Review findings will inform clinicians and researchers on the best available tools for the  
32  
33 135 assessment of physical functioning in AIS. Furthermore, findings will inform future research drawing  
34  
35 136 on a range of measures of physical functioning to investigate health status in AIS.

### 35 137 **Objective**

36  
37  
38 138 To identify outcome measures used to assess physical functioning in individuals with AIS. A secondary  
39  
40 139 aim is to evaluate the measurement properties of physical functioning outcome measures in AIS.  
41

### 42 140 **METHODS**

43  
44  
45 141 This protocol has been informed by experts in musculoskeletal orthopaedics including a consultant  
46  
47 142 spine surgeon, musculoskeletal rehabilitation experts including physiotherapists and individuals with  
48  
49 143 review, measurement properties and research experience. It has been designed in line with the  
50  
51 144 COSMIN methodology for systematic reviews of Patient-Reported Outcome Measures [30] and is  
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53 145 reported in line with the Preferred Reporting Items for Systematic Reviews and Meta-Analysis-P  
54  
55 146 (PRISMA-P) [31] The search for this systematic review will be conducted in two parts. Stage one to  
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57 147 identify studies used an outcome measure to evaluate physical functioning in individuals with AIS. This  
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59 148 search will allow a list of outcome measures to be generated. Stage 2 will identify studies, which  
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3 149 evaluated measurement properties of physical functioning outcome measure identified in the first  
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5 150 search.

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8 151 **Stage one: Inventory of outcome measure**

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10 152 **Eligibility criteria**

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12  
13 153 *Study design*

14  
15 154 All study designs including; randomised clinical trials, cohort, observational studies and case studies  
16  
17 155 will be included to identify all outcome measure of physical functioning being used with individuals  
18  
19 156 with AIS. No limitation on language or location will be applied at this stage.

20  
21 157 *Participants*

22  
23  
24 158 Participants aged between the age of 10 years to 18 years of age, with a diagnosis of idiopathic  
25  
26 159 scoliosis and  $\geq 10^\circ$  Cobb angle will be considered. No restrictions will be applied to the curve severity,  
27  
28 160 evaluation settings, and the type of treatment.

29  
30 161 *Outcome*

31  
32 162 Any study that includes assessments of the physical functioning of AIS using a specific outcome  
33  
34 163 measure will be included. Physical functioning is defined according to the Core Outcome Measures in  
35  
36 164 Effectiveness Trials (COMET) taxonomy, [15] as any physical activities of daily living such as the ability  
37  
38 165 to walk, independence, self-care, performance status, disability index. [15, 32] The outcome measures  
39  
40 166 are defined as any one of the following:

- 41  
42 167 1. Patient-Reported Outcome Measures (PROMs) in the form of questionnaires or scales  
43  
44 168 designed for AIS to evaluate physical functioning or if it is included as a sub-scale within a  
45  
46 169 questionnaire.
- 47 170 2. Performance-based outcome measures (PBOMs); a measure of physical functioning by  
48  
49 171 clinician while the individual is performing a functional task e.g walking, and/or
- 50 172 3. Body structure and function measures which means any dysfunction in a specific body part or  
51  
52 173 system which may limits function, such as range of motion. [26]

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## 176 **Search strategy**

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

## 186 **Stage two: measurement properties**

### 187 **Eligibility criteria**

#### 188 *Study design*

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

#### 193 *Participants*

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

#### 199 *Outcome*

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

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

### 207 **Search strategy**

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

### 214 **Information sources**

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

### 226 **Data management**

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

### 230 **Selection Process**

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

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

#### 240 **Data collection process**

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

#### 246 **Data items**

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

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

<b>Study &amp; Participants Characteristics</b>	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)
<b>Outcome measure</b>	<b>PROM:</b> 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
	<b>PBOM:</b> Name of outcome measure, equipment needed, number of assessments, outcome (e.g. time needed, ability/disability), setting, scoring
	<b>Body structure and function measure:</b> Name of outcome measure, equipment needed, mode of administration, setting, scoring, outcome (e.g. time needed, ability/disability)
<b>Measurement properties</b>	<b>Validity:</b> Name of outcome measure, type of validity, descriptive statistics, missing value, comparator outcome or predictor outcome, hypothesis, statistics method, confidence interval, validation results
	<b>Reliability:</b> Name of outcome measure, type of reliability, descriptive statistic, time interval, reliability coefficient, measurement error

	<b>Responsiveness:</b> Name of outcome measure, Method of testing: <i>Hypothesis testing, Distribution based method</i> (ES, SRM and MDC), hypothesis, time to follow-up. <i>Anchor-based methods</i> (MIC or MCIC or MID), anchor/s.
	<b>Interpretability:</b> 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
	<b>Feasibility:</b> 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	

## 249 Outcomes and prioritisation

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

## 253 Risk of bias in individual studies

254 The COSMIN checklist for assessment of risk of bias and methodological quality in individual studies  
 255 will be used. [28] It was revised and specifically designed for use in systematic reviews of PROMs to  
 256 evaluate studies on measurement properties. [34] The methodological quality of each study for each  
 257 measurement property will be assessed separately. [30] The items for each measurement property in  
 258 the relevant standards box will be rated as either very good, adequate, doubtful or inadequate quality.  
 259 [30] Then, the overall methodological quality of the measurement property will be rated based on  
 260 "the worst score counts principle" i.e. that the overall quality of the study for a specific measurement  
 261 property is based on the lowest rating of any items in the standards' box. [30] The result of each item  
 262 and overall rating will be reported in the final results. The COSMIN group recommend researchers to  
 263 adapt the checklist to other measures (i.e. PBOMs, body structure and function measure) since it was  
 264 originally developed for PROMs. [30] Two independent reviewers (SA, EB) will assess the risk of bias  
 265 for all included studies. Any disagreement will be resolved through discussion, and if no agreement is  
 266 reached, a third reviewer (NH) will be consulted. The agreement between reviewers will be estimated  
 267 with percentage agreement and the Kappa statistic using SPSS for Windows statistical software  
 268 package (IBM SPSS Statistics Version 25) and will be reported in the final results.

269



## 270 **Data synthesis**

271 The COSMIN guidelines for systematic reviews will be followed for the synthesis of the results. [30]  
272 Characteristics of the outcome measures, sample, measurement properties results, information about  
273 interpretability and feasibility of the scores of the included outcome measures will be presented in  
274 overview tables for each outcome measure. [30] Each measurement property for each study per tool  
275 will be rated against the updated criteria for good measurement properties as either sufficient (+),  
276 insufficient (-), or indeterminate (?).[30] The result of rating of measurement property and its  
277 methodological quality rating will be added to the overview table. [30] Then, the evidence will be  
278 pooled or summarized per measurement property per tool, with the overall result will be rated against  
279 the criteria for good measurement properties, and the quality of the evidence will be graded using a  
280 modified Grading of Recommendations Assessment, Development and Evaluation (GRADE) approach.  
281 [30]

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

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

## 298 **Confidence in cumulative evidence**

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

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3 301 measure in each category will be evaluated. The GRADE approach uses five factors to determine the  
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5 302 quality of the evidence: risk of bias (quality of the studies), inconsistency (of the results of the studies),  
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7 303 indirectness (evidence comes from different populations, interventions or outcomes than the ones of  
8  
9 304 interest in the review), imprecision (wide confidence intervals), and publication bias (negative results  
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11 305 are less often published).[35] For evaluating measurement properties in systematic reviews of PROMs,  
12  
13 306 only four factors will be assessed as recommended by COSMIN group, while the fifth factor  
14  
15 307 (publication bias) will be removed as there is no registry exists for measurement properties.  
16

308

## 309 **Discussion**

310

311 Physical functioning is considered as an important outcome domain in health-related quality of life.  
312 [12] It can be used to predict future disability as well as health and social care use.[13] Individuals with  
313 AIS reported a limitation in their physical functioning [7]. Thus, measurement of its impact is important  
314 in research and clinical practice. Numerous of tools are available for the assessment of physical  
315 functioning, ranging from patient-reported to performance-based measures. However, it is essential  
316 to confirm the psychometric properties of these tools before recommending for clinical use. The COS  
317 study for 'all spine deformities' identified the SRS-22r as the recommended PROM for assessment for  
318 physical functioning among young adults with spinal deformities. [16] However, there is still a need  
319 for a more specific review that evaluate the quality of all outcome measures used in the assessment  
320 of physical functioning in AIS including patient-reported, and performance-based as well as measures  
321 of body structure and function. This systematic review will retrieve all tools that have been used to  
322 assess physical functioning among individuals with AIS. Then, it will evaluate and synthesise the quality  
323 of studies that report psychometric properties of physical functioning outcome measures in AIS. This  
324 review will provide a comprehensive assessment of current evidence which may benefit: (1) health  
325 practitioners in selection of the most suitable tools to assess physical functioning in AIS (2) patients  
326 who need a good outcome measures that reflect their actual health status (3) researchers and policy  
327 maker who can use the recommend measures in designing research trials and defining the COS for  
328 individuals with AIS, which in turn will improve health assessment and patient care. Limitations of this  
329 review are a focus on individuals with AIS specifically, so recommendations cannot be generalised to  
330 other forms of scoliosis.

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### 335 **Patient and public involvement**

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

343

### 344 **Implications of this study**

345

346 AIS is a complex deformity of the spine and causes a significant impact on physical activities of  
347 individuals' daily living such as walking and maintaining body position. [7, 17] In consequence, the  
348 quality of life is affected. Physical functioning gives an indication about the current health status and  
349 identifies people at risk of disability. [12, 13] Therefore, physical functioning is considered as one of  
350 the outcomes that should be assessed and reported in clinical trials of musculoskeletal conditions.  
351 [15] A systematic review is needed to evaluate current practice in the assessment of physical  
352 functioning among individuals with AIS. The results of this review will inform clinicians and researchers  
353 on the best available tools for assessment of physical functioning in AIS. This review could provide a  
354 research agenda that may highlight the gap in the literature around physical functioning measure and  
355 their measurement properties among individuals with AIS.

356

### 357 **Declarations**

### 358 **Ethics and Dissemination**

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

363

### 364 **Author Contributions**

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

1  
2  
3 368 commented on each draft of the protocol. All authors have approved and contributed to the final  
4  
5 369 manuscript.

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7  
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9  
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11  
12 372 form University of Tabuk, Tabuk, Saudi Arabia.

13  
14 373 **Competing interests**

15  
16 374 None declared.

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19 375 **Patient consent for publication**

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21 376 Not required.

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24 378 **Figure Captions**

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27 379 **Figure 1:** Flow diagram of search strategy (Search One and Two) and selection process.

28 380 PROM=Patient Reported Outcome Measure; PBOM= Performance-Based Outcome

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32 382 **REFERENCES**

33  
34  
35 383 1 Reamy BV, Slakey JB. Adolescent idiopathic scoliosis: review and current concepts. *Am*

36  
37 384 *Fam Physician* 2001;**64**:111-6.

38  
39  
40 385 2 Cobb JR. The problem of the primary curve. *J Bone Joint Surg Am* 1960;**42-A**:1413-25.

41  
42 386 3 Konieczny MR, Senyurt H, Krauspe R. Epidemiology of adolescent idiopathic scoliosis.

43  
44 387 *J Child Orthop* 2013;**7**:3-9.

45  
46  
47 388 4 Hamad A, Ahmed EB, Tsirikos AI. Adolescent idiopathic scoliosis: a comprehensive

48  
49 389 approach to aetiology, diagnostic assessment and treatment. *Orthopaedics and Trauma*

50  
51 390 2017;**31**:343-9.

52  
53  
54 391 5 Horne JP, Flannery R, Usman S. Adolescent idiopathic scoliosis: diagnosis and

55  
56 392 management. *Am Fam Physician* 2014;**89**:193-8.

57  
58  
59 393 6 Lonstein D. Adolescent idiopathic scoliosis. *The Lancet* 1994;**344**:1407-12.

- 1  
2  
3 394 7 Du C, Yu J, Zhang J, *et al.* Relevant areas of functioning in patients with adolescent  
4  
5  
6 395 idiopathic scoliosis on the International Classification of Functioning, Disability and Health:  
7  
8 396 The patients' perspective. *J Rehabil Med* 2016;**48**:806-14.  
9  
10 397 8 Makino T, Kaito T, Kashii M, *et al.* Low back pain and patient-reported QOL outcomes  
11  
12 398 in patients with adolescent idiopathic scoliosis without corrective surgery. *Springerplus*  
13  
14 399 2015;**4**:397.  
15  
16  
17 400 9 Durmala J, Tomalak W, Kotwicki T. Function of the respiratory system in patients with  
18  
19 401 idiopathic scoliosis: reasons for impairment and methods of evaluation. *Stud Health Technol*  
20  
21 402 *Inform* 2008;**135**:237-45.  
22  
23  
24 403 10 Leszczewska J, Czaprowski D, Pawlowska P, *et al.* Evaluation of the stress level of  
25  
26 404 children with idiopathic scoliosis in relation to the method of treatment and parameters of  
27  
28 405 the deformity. *Scientific World Journal* 2012;**2012**:538409.  
29  
30  
31 406 11 Malmqvist M, Tropp H, Lyth J, *et al.* Patients With Idiopathic Scoliosis Run an Increased  
32  
33 407 Risk of Schizophrenia. *Spine Deform* 2019;**7**:262-6.  
34  
35  
36 408 12 Taylor AM, Phillips K, Patel KV, *et al.* Assessment of physical function and participation  
37  
38 409 in chronic pain clinical trials: IMMPACT/OMERACT recommendations. *Pain* 2016;**157**:1836-  
39  
40 410 50.  
41  
42  
43 411 13 Tomey KM, Sowers MR. Assessment of physical functioning: a conceptual model  
44  
45 412 encompassing environmental factors and individual compensation strategies. *Phys Ther*  
46  
47 413 2009;**89**:705-14.  
48  
49  
50 414 14 Cooper R, Kuh D, Cooper C, *et al.* Objective measures of physical capability and  
51  
52 415 subsequent health: a systematic review. *Age Ageing* 2011;**40**:14-23.  
53  
54  
55 416 15 Dodd S, Clarke M, Becker L, *et al.* A taxonomy has been developed for outcomes in  
56  
57 417 medical research to help improve knowledge discovery. *J Clin Epidemiol* 2018;**96**:84-92.  
58  
59  
60

- 1  
2  
3 418 16 de Kleuver M, Faraj SSA, Holewijn RM, *et al.* Defining a core outcome set for  
4  
5  
6 419 adolescent and young adult patients with a spinal deformity. *Acta Orthop* 2017;**88**:612-8.  
7  
8 420 17 Kibsgard T, Brox JI, Reikeras O. Physical and mental health in young adults operated  
9  
10 421 on for idiopathic scoliosis. *J Orthop Sci* 2004;**9**:360-3.  
11  
12  
13 422 18 Bastrom TP, Marks MC, Yaszay B, *et al.* Prevalence of postoperative pain in adolescent  
14  
15 423 idiopathic scoliosis and the association with preoperative pain. *Spine (Phila Pa 1976)*  
16  
17 424 2013;**38**:1848-52.  
18  
19  
20 425 19 Seki H, Ideno S, Ishihara T, *et al.* Postoperative pain management in patients  
21  
22 426 undergoing posterior spinal fusion for adolescent idiopathic scoliosis: a narrative review.  
23  
24 427 *Scoliosis Spinal Disord* 2018;**13**:17.  
25  
26  
27 428 20 Ward MM. Interpreting measurements of physical function in clinical trials. *Ann*  
28  
29 429 *Rheum Dis* 2007;**66 Suppl 3**:iii32-4.  
30  
31  
32 430 21 Haheer TR, Gorup JM, Shin TM, *et al.* Results of the Scoliosis Research Society  
33  
34 431 instrument for evaluation of surgical outcome in adolescent idiopathic scoliosis. A multicenter  
35  
36 432 study of 244 patients. *Spine (Phila Pa 1976)* 1999;**24**:1435-40.  
37  
38  
39 433 22 Baldus C, Bridwell K, Harrast J, *et al.* The Scoliosis Research Society Health-Related  
40  
41 434 Quality of Life (SRS-30) age-gender normative data: an analysis of 1346 adult subjects  
42  
43 435 unaffected by scoliosis. *Spine (Phila Pa 1976)* 2011;**36**:1154-62.  
44  
45  
46 436 23 Chen AF, Bi W, Singhabahu D, *et al.* Converting Scoliosis Research Society-24 to  
47  
48 437 Scoliosis Research Society-22r in a Surgical-Range, Medical/Interventional Adolescent  
49  
50 438 Idiopathic Scoliosis Patient Cohort. *Spine Deform* 2013;**1**:108-14.  
51  
52  
53 439 24 Asher M, Min Lai S, Burton D, *et al.* Discrimination validity of the scoliosis research  
54  
55 440 society-22 patient questionnaire: relationship to idiopathic scoliosis curve pattern and curve  
56  
57 441 size. *Spine (Phila Pa 1976)* 2003;**28**:74-8.  
58  
59  
60

- 1  
2  
3 442 25 Asher MA, Min Lai S, Burton DC. Further development and validation of the Scoliosis  
4  
5  
6 443 Research Society (SRS) outcomes instrument. *Spine (Phila Pa 1976)* 2000;**25**:2381-6.  
7  
8 444 26 Reiman MP, Manske RC. The assessment of function: How is it measured? A clinical  
9  
10 445 perspective. *J Man Manip Ther* 2011;**19**:91-9.  
11  
12  
13 446 27 Bean JF, Olveczky DD, Kiely DK, *et al.* Performance-based versus patient-reported  
14  
15 447 physical function: what are the underlying predictors? *Phys Ther* 2011;**91**:1804-11.  
16  
17  
18 448 28 Mokkink LB, de Vet HCW, Prinsen CAC, *et al.* COSMIN Risk of Bias checklist for  
19  
20 449 systematic reviews of Patient-Reported Outcome Measures. *Qual Life Res* 2018;**27**:1171-9.  
21  
22  
23 450 29 Mokkink LB, Terwee CB, Patrick DL, *et al.* The COSMIN study reached international  
24  
25 451 consensus on taxonomy, terminology, and definitions of measurement properties for health-  
26  
27 452 related patient-reported outcomes. *J Clin Epidemiol* 2010;**63**:737-45.  
28  
29  
30 453 30 Mokkink LB, Prinsen C, Patrick DL, *et al.* COSMIN methodology for systematic reviews  
31  
32 454 of Patient-Reported outcome measures (PROMs). *User manual* 2018.  
33  
34  
35 455 31 Moher D, Shamseer L, Clarke M, *et al.* Preferred reporting items for systematic review  
36  
37 456 and meta-analysis protocols (PRISMA-P) 2015 statement. *Syst Rev* 2015;**4**:1.  
38  
39  
40 457 32 Williamson PR, Altman DG, Bagley H, *et al.* The COMET Handbook: version 1.0. *Trials*  
41  
42 458 2017;**18**:280.  
43  
44  
45 459 33 Terwee CB, Jansma EP, Riphagen, II, *et al.* Development of a methodological PubMed  
46  
47 460 search filter for finding studies on measurement properties of measurement instruments.  
48  
49 461 *Qual Life Res* 2009;**18**:1115-23.  
50  
51  
52 462 34 Mokkink LB, Terwee CB, Patrick DL, *et al.* The COSMIN checklist for assessing the  
53  
54 463 methodological quality of studies on measurement properties of health status measurement  
55  
56 464 instruments: an international Delphi study. *Qual Life Res* 2010;**19**:539-49.  
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465 35 Balshem H, Helfand M, Schunemann HJ, *et al.* GRADE guidelines: 3. Rating the quality  
466 of evidence. *J Clin Epidemiol* 2011;**64**:401-6.

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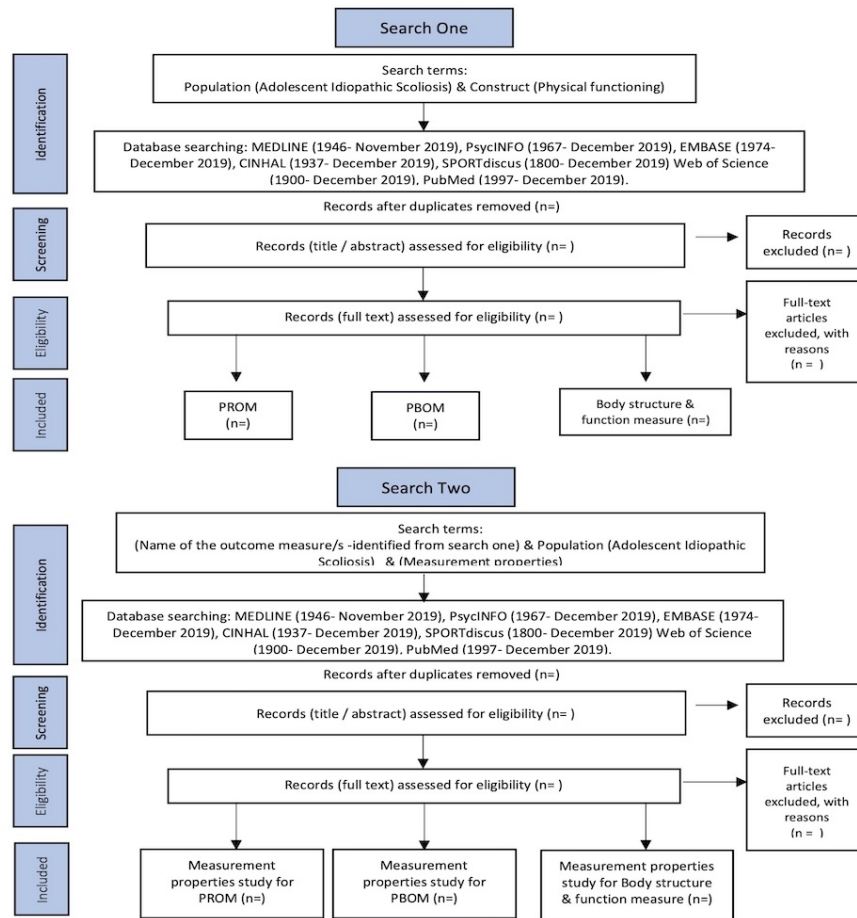


Figure 1: Flow diagram of search strategy (Search One and Two) and selection process. PROM=Patient Reported Outcome Measure; PBOM= Performance-Based Outcome Measure.

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**Supplementary File 1**

## Example of search strategy (MEDLINE)

### Stage 1

1. scoliosis.mp.
2. exp Scoliosis/
3. exp Spinal Curvatures/
4. Adolescens\$.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.

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- 4 33. exp sitting/
- 5 34. exp Walking/
- 6 35. exp Walking Speed/
- 7 36. exp Postural Balance/
- 8 37. Standing balance.mp.
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- 10 38. exp Hand Strength/ or Grip strength.mp.
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- 12 39. 1- 3/OR
- 13 40. 4 OR 5
- 14 41. 6-38/OR
- 15 42. 39 and 40 and 41
- 16 43. Limit 42 to humans
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## 21 Stage 2

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- 24 44. Name of the Identified outcome measure/s
- 25 45. scoliosis.mp.
- 26 46. exp Scoliosis/
- 27 47. exp Spinal Curvatures/
- 28 48. Adolescen\$.mp.
- 29 49. exp Adolescent/
- 30 50. validity.mp
- 31 51. exp validation studies/
- 32 52. reliability.mp
- 33 53. exp reproducibility of results/
- 34 54. interpretability.mp
- 35 55. internal consistency.mp
- 36 56. exp sensitivity and Specificity/
- 37 57. clinical sensitivity.mp
- 38 58. exp psychometrics/
- 39 59. responsiveness.mp
- 40 60. exp Evaluation studies/
- 41 61. measurement error.mp
- 42 62. measurement properties.mp
- 43 63. 45-47/OR
- 44 64. 48-49/OR
- 45 65. 50-62/OR
- 46 66. 44 and 63 and 64 and 65
- 47 67. Limit 66 to humans
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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	Reported on Page
<b>ADMINISTRATIVE INFORMATION</b>			
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	Page 13 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	Page 13 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
<b>INTRODUCTION</b>			
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
<b>METHODS</b>			
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 $\tau$ )	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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