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QUALITATIVE REVIEW

Body Image and Body Schema in Adolescents with Idiopathic Scoliosis: A Scoping Review

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Abstract

Alterations in body representations (i.e., body image and body schema) are increasingly getting attention in clinical practice. Adolescents affected by idiopathic scoliosis experience body image dissatisfaction, and alterations in body schema have been suggested to be a consequence of the disease development. Although research has recognized the predisposing role of body representation disorders to psychopathologies, these aspects have been largely overlooked in this clinical population. This scoping review aims to establish the state of the art on the widely neglected aspects of body image and body schema disorders in adolescents affected by idiopathic scoliosis. PubMed, Scopus, PsycInfo, and MEDLINE were consulted to select articles published between 2000 and 2021. Three independent reviewers identified 27 articles by following the Preferred Reporting Items for Systematic Reviews and Meta-Analyses extension for scoping review guidelines. Body image was assessed in 24 of the 27 studies. Body image disorders were reported, with more severe scoliosis cases showing higher body image dissatisfaction. Surgery seems to be the best approach to improve body image outcomes, but studies did not reveal clear associations between clinical measures of scoliosis severity (e.g., Cobb angle, hump height) and body image. Disorders of body schema have been reported, but the finding might have been biased by the paucity of studies on this aspect of body representations (4/27). This review highlighted the wide prevalence of psychological distress and body schema alterations among adolescents affected by idiopathic scoliosis; but it also revealed that both are disregarded and not properly evaluated.

Keywords Body representation · Body image · Body schema · Adolescents · Idiopathic scoliosis

Introduction

Alterations of body representations are commonly encountered conditions among different clinical and non-clinical settings. Many psychiatric diseases such as eating disorders or body dysmorphic disorders are characterized by body image misperceptions and dysfunctional beliefs about the body. Neurological conditions affecting somatosensation also can eventually lead to disorders of body representations (e.g., somatoparaphrenia, personal neglect, phantom limbs) (Case et al., 2019). However, alterations of body representations can be observed even in unlooked-for clinical conditions. Among them, the adolescent girls affected by idiopathic scoliosis (Carrasco & Ruiz, 2014). Because of adolescent idiopathic scoliosis disease characteristics, this condition can be used as a model to study the concurrent impairment of dual aspects of body representation: body image and body schema. To date, these facets of idiopathic scoliosis have been largely neglected compared to their clinical and genetic determinants. This study addresses the need

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to systematically review literature on body representation disorders (i.e., body image and body schema) in adolescents affected by idiopathic scoliosis to better understand its multifactorial nature and improve these patients' care. Moreover, it seeks to advance a better theoretical comprehension of body image and body schema' interaction.

Literature disagrees on how many brain representations of the human body exists, but at least two are always mentioned: body image and body schema. Body image is a complex psychological construct involving the subjective perception of body appearance and its associated feelings, beliefs and thoughts (de Vignemont, 2010). Body image disorders can manifest as alterations of each of these components, resulting in different pathological conditions. Thus, perceptual and cognitive disorders of body image are associated with eating disorders (Sattler et al., 2020), while dysfunctional feelings on physical appearance can lead to depression (Soares Filho et al., 2020). Body schema is a different aspect of body representation. It is defined as a continuously updated and largely unaware sensorimotor representation of the body in the space and of the configurations of all its parts in relation between each other and the world (de Vignemont, 2010; Head & Holmes, 1912). Hence, body schema has a fundamental role in setting body movements and in posture control mechanisms (Picelli et al., 2016).

Adolescents affected by idiopathic scoliosis display concurrent alterations of both of these aspects of body representation. Indeed, adolescent idiopathic scoliosis presents as a noticeable three-dimensional spinal deformity, characterized by frontal plane deviation, rotation of the affected vertebral bodies and reduction of the physiological curves on the sagittal plane (Negrini et al., 2018). The disfiguring appearance caused by the scoliotic curve and the related brace treatments has notable effects on adolescents' mental health. Not surprisingly, body image dissatisfaction in this clinical population has been largely described as having detrimental effects on both quality of life and adherence to treatment (Auerbach et al., 2014; Tones et al., 2006).

Adolescent idiopathic scoliosis etiopathogenesis is largely unknown, but a central nervous system (CNS) involvement has been postulated. Many studies have reported visual, vestibular, and proprioceptive system deficits, postural control alterations and brain structural abnormalities (Burwell & Dangerfield, 2002; Burwell et al., 2008; Chu et al., 2011). Starting from these findings, some authors have suggested the so-called "double neuro-osseous theory" of scoliosis, positing an imbalance between peripheral skeletal/body maturation and posterior parietal cortex (PPC) sensory-motor integration processes (Assaiante et al., 2014; Burwell et al., 2006). These abnormalities in sensory-motor adaptation of the brain to the growing bodies of adolescents corroborate the hypothesis of an altered body schema in this clinical population.

Current Study

Scoliosis-associated disorders of body representations among adolescents have been inadequately studied compared to the other determinants of this condition, regardless of their clinical relevance. The present scoping review aims to summarize literature findings on disorders of body representations in adolescents affected by idiopathic scoliosis, including studies on body schema alterations and body image distortions. Toward this aim, three research questions were formulated. Do adolescents affected by idiopathic scoliosis have altered body image and/or body schema (Question 1)? Are there any correlations between altered body representations and clinical parameters of scoliosis severity (Question 2)? Do different treatment approaches have a different impact on body representations (Question 3)?

Method

Considering the high heterogeneity of the assessment instruments adopted to evaluate body representations and different study designs, a scoping review was deemed the best-suited tool to answer the research questions.

The Preferred Reporting Items for Systematic reviews and Meta-Analysis extension for Scoping Reviews (PRISMA-ScR) guidelines were followed (Tricco et al., 2018).

Eligibility Criteria

The included articles were full-length, peer-reviewed, original research articles written in English and published between 2000 and 15th June 2020. Participants included in the studies should be diagnosed with Adolescent Idiopathic Scoliosis by an expert clinician, according to Scoliosis Research Society criteria (Negrini et al., 2018), and additionally assessed for at least one of our measures of interest: body image alterations or body schema alterations. Clinical and psychological (body image) evaluations had to be carried out using standardized assessment tools. As body schema is often indirectly assessed manipulating sensory and proprioceptive information, articles adopting innovative experimental paradigms were included as well if provided with adequate control groups. Studies not matching the inclusion criteria, single case studies, validation studies and literature reviews were excluded (See Supplementary Material).

Information Sources

A comprehensive systematic search on body representations in adolescent idiopathic scoliosis was performed

independently by two authors (MB, FC). The following keywords were chosen to search on SCOPUS, PubMed, PsycINFO and MEDLINE databases: (*Adolescent idiopathic scoliosis OR scoliosis), AND (body representation OR body schema OR body schema alterations OR body image OR body image alterations)*. See Appendix for detailed search queries in PubMed. Authors additionally screened reference lists of retrieved articles and relevant reviews.

The final search results were exported in MENDELEY where duplicates were removed by the dedicated tool.

Selection of Sources of Evidence

To increase consistency in article inclusion and data extraction, two reviewers (MB, FC) screened the same 42 publications. Eventual disagreements during the whole selection procedure were mediated by a third reviewer (SM). See Fig. 1 for the full selection procedure.

Quality Assessment

As no agreed form exist to evaluate the methodological quality of articles for scoping reviews, the three reviewers jointly established six quality criteria by adapting already published ones of a systematic (Taborri et al., 2018) and a scoping review (Rubega et al., 2021):

1. Aim/s of the study was/were clearly stated.
2. Selection Bias: inclusion and exclusion criteria were clearly described.
3. Performance Bias: the process of data collection was precisely detailed and reliable; a control group was present.
4. Detection Bias: the outcomes reported were relevant to answer the research question.
5. Results presentation: the tests and questionnaires scores were correctly and completely reported (i.e., mean/median, and standard deviation).
6. Statistical approach: statistics performed was appropriated and clearly described; enough subjects were included (power analysis).

For each criterium was assigned a score ranging from 0 to 2 (0 = not meeting, 1 = partially meeting, 2 = fully meeting the criteria). Scores were finally added for each article resulting in a final quality score ranging from 0 to 12. An arbitrary cut-off of at least 60% (> 7) of the maximum possible score (12) was established for articles inclusion (See Appendix, Table 5, Articles quality assessment).

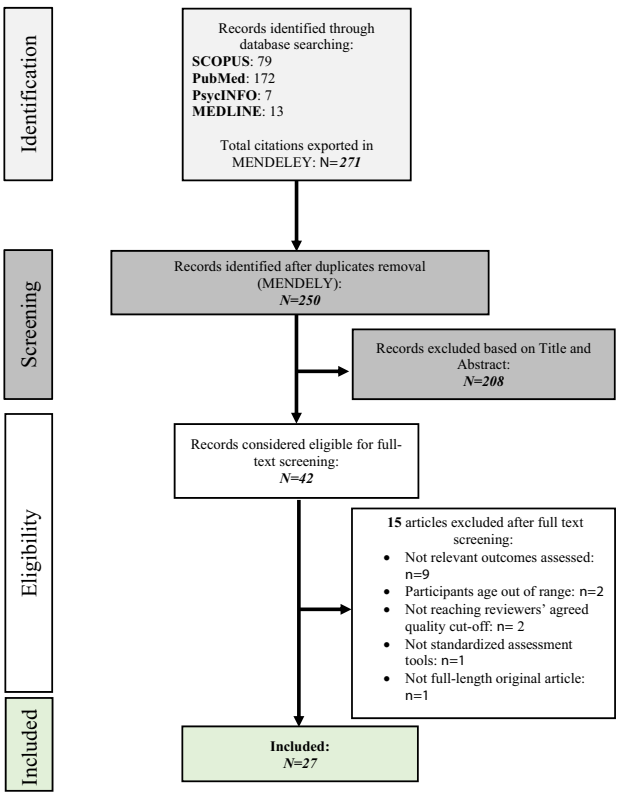

Fig. 1 Flow chart of the systematic search. Preferred Reporting Items of Systematic Review and Meta-Analysis (PRISMA) flow chart showing the process of systematic article search and selection

Data Charting Process and Data Items

A data charting form was jointly designed by the three reviewers adapting the one proposed by the Cochrane handbook for systematic reviews (Higgins & Green, 2011), (see Supplementary Material). Two reviewers (MB, FC) independently charted the data, discussed the results, and cross-checked their forms. The third reviewer (SM) revised articles in cases of disagreement on data extraction or inclusion. The charting form comprises an initial part including general articles' information (title, first author name, Journal, year of publication, type of publication, short article description) and organizational information (date of revision and reviewer identity). The second part includes the eligibility form based on articles' general characteristics (e.g., date of publication, language) participants' characteristics (age, diagnosis), assessment instruments and outcomes. In the last part reports study-specific information (study design, sample characteristics, assessment tools) and the findings of interest. Data on body image and body schema alterations in adolescent idiopathic scoliosis were summarized and placed in relation to clinical indices of scoliosis and treatment approaches.

Results

An initial literature screening based on titles and abstracts led to the identification of 42 articles, which underwent full-text screening. Of these, a total of 15 were excluded for the following reasons: 9 didn't report outcomes of interest to answer the research question; in two of them, subjects age was out of the inclusion range; two did not reach the estimated cut-off of quality; one used non-standardized assessment tools; one was not a full-length original article.

The twenty-seven articles eventually included were grouped according to the type of body representation assessed: body image and body schema. For each domain, the assessment tools, the main findings related to body representational alterations, their correlation with clinically relevant variables, and the effects of different scoliosis treatment approaches on body representation changes were reported.

Study Characteristics

Studies were heterogeneous in terms of study design with a higher prevalence of cross-sectional (10/27) and longitudinal prospective trials (8/27). A minor number of articles were retrospective (4/27), case-controlled (3/27) and randomized controlled (2/27) trials (see Table 1).

Most of the included studies assessed body image alterations (24/27) while body schema was assessed in just four articles (See Table 2).

The Scoliosis research society questionnaire (SRS), (Asher et al., 2003) emerged as the preferred assessment tool to evaluate body image alterations (19/24), specifically the SRS-22 (15/19), SRS-23 (Asher et al., 2003), (2/19), and SRS-24, (2/19), (Haher et al., 1999).

The SRS-22 questionnaire is composed of 22 items, each one scored from 1(worst) to 5 (best), and sorted in different domains (Pain, Self-Image, Function, Mental Health, Satisfaction with Management, and total score).

The other body image assessment scales were: the Spinal Appearance Questionnaire (SAQ) (Sanders et al., 2007), (4/24); the Trunk Appearance Perception Scale (TAPS), (Bago et al., 2010), (3/24); the Walter Reed Visual Assessment Scale (WRVAS)(Pineda et al., 2006) (2/24); the Body Image Disturbance Questionnaire-Scoliosis (BIDQ-S)(Auerbach et al., 2014) (2/24); the Body Shape Questionnaire (BSQ-14), (Dowson & Henderson, 2001), and the Body Cathexis Scale (BCS), (Secord & Jourard, 1953);

Body schema alterations were assessed indirectly employing the following experimental paradigms: a laser line projection task (J. Cheung et al., 2002) specifically designed to test perception of body posture; a motorized version of the wheel paradigm described by Pérennou to assess subjective

Table 1 Study designs

Study Designs	Articles	Total number
Cross-sectional	Babaee et al. (2014) Cantele et al. (2020) Cheung et al. (2002) Çolak et al (2017) Lee et al. (2016) Lendzion et al. (2018) Misterska et al. (2014) Picelli et al. (2016) Soliman (2018) Watanabe et al. (2005)	10
Longitudinal prospective	Duramaz et al. (2018) Lonner et al. (2019) Mariconda et al. (2016) Misterska et al. (2011) Misterska et al. (2013) Pérez-Prieto et al. (2014) (Multicenter) Schwieger et al. (2016) (Multicenter) Watanabe et al. (2007)	8
Retrospective studies	Asher et al. (2004) Brewer et al. (2013) Cheshire et al. (2017) Wang et al. (2014a, 2014b)	4
Case–control studies	Le Berre et al. (2019) (Multicenter) Paolucci et al. (2017) Yagci et al. (2020)	3
Randomized Controlled trials	Schwieger et al. (2017) (Multicenter) Yagci et al. (2018)	2

Table 2 Assessment tools

Assessed domain	Article	Assessment tools
Body image	Asher et al (2004)	SRS22
	Babaee et al (2014)	SRS-22 (Persian version)
	Brewer et al (2013)	SRS-22
	Cantele et al (2020)	SRS-22
	Cheshire et al (2017)	SRS-22
	Çolak et al (2017)	SRS-23, WRVAS
	Duramaz et al (2018)	BCS
	Lee et al. (2016)	SRS-22
	Lendzion et al (2018)	SRS-22 (Polish version), TAPS
	Lonner et al (2019)	SRS-22, BIDQ-S
	Mariconda et al (2016)	SRS-23
	Misterska et al (2011)	SAQ
	Misterska et al (2013)	SRS-22, TAPS
	Misterska et al. (2014)	SAQ (Poland version)
	Paolucci et al. (2017)	SRS-22, TAPS, Drawing test
	Pérez-Prieto et al. (2014)	SRS-22 (Spanish version), BSQ-14
	Schwieger et al (2016)	SAQ
	Schwieger et al (2017)	SAQ
	Soliman (2018)	SRS-22r (Arabic version), BIDQ-S
	Wang et al. (2014a, 2014b)	SRS-22 (Chinese version)
	Watanabe et al (2005)	SRS-24 (Japanese version)
	Watanabe et al (2007)	SRS-24 (Japanese version)
	Yagci et al (2018)	SRS-22, WRVAS
	Yagci et al (2020)	SRS-22
Body schema	Cheung et al (2002)	Laser line projection task
	Le Berre et al (2019)	Optokinetic stimulation (SVV), Motorized version of the wheel paradigm described by Pérennou (SPV)
	Picelli et al (2016)	Graphic table displaying pictures of progressively increasing scoliotic curves
	Yagci et al (2020)	ABC

SRS Scoliosis Research Society questionnaire, *WRVAS* Walter Reed Visual Assessment Scale, *BCS* Body Cathexis Scale, *TAPS* Trunk Appearance Perception Scale, *BIDQ-S* Body Image Disturbance Questionnaire-Scoliosis, *BSQ* Body Shape Questionnaire, *SAQ* Spinal Appearance Questionnaire, *SVV* Subjective Visual Vertical, *SPV* Subjective Postural Vertical, *ABC* Awareness-Body-Chart

postural verticality (Pérennou et al., 2008) and optokinetic visual stimulation to assess subjective visual verticality (Le Berre et al., 2019); a graphic table displaying pictures of progressively increasing scoliotic curves designed to test awareness of trunk misalignment (Picelli et al., 2016) and a measure of subjective body awareness, namely the Awareness-Body-Chart test (ABC), (Danner et al., 2017).

Adolescent Idiopathic Scoliosis and Body Image

Body image alterations were consistently reported in AIS. Tables 3 and 4 report respectively the self-image subitems median and/or mean scores assessed by the SRS questionnaires and by the other assessment instruments.

Nineteen articles report SRS self-image subitems scores (Table 3). Eight of these reported scores differences within AIS sub-groups of different scoliosis severity (3/8), (Lee et al., 2016; Soliman, 2018; Watanabe et al., 2005), under different types of treatment (3/8) (Çolak et al., 2017; Lonner et al., 2019; Mariconda et al., 2016), or compared with healthy control groups (4/8), (Cantele et al., 2020; Pérez-Prieto et al., 2014; Soliman, 2018; Watanabe et al., 2005):

Lower body image subitems scores were found in more severe scoliosis cases, compared with the mild severity cases, (Lee et al., 2016) $p = 0.031$, (Soliman, 2018) $p < 0.001$ and (Watanabe et al., 2005) $p < 0.05$. Two works did not find significant differences (Asher et al., 2004; Wang et al., 2014a, 2014b). The effects of different treatment approaches in influencing SRS self-image subitem scores are conflicting, with two studies finding improvement after surgery, namely (Lonner et al., 2019) $p < 0.001$ and (Mariconda et al., 2016) $p < 0.01$, one finding better scores in those patients treated

Table 3 Scoliosis Research Society (SRS) self-image sub-scores

First Author	Year	N° subjects by group	Age (mean years ± SD)	SRS	Self- Image Subitem (Median or mean ± SD; range = 1–5)	Group differences
Asher, M	2004	**Thoracic:** 35 **Double:** 14 **Thoracolumbar:** 11	15 ± 6	SRS-22	Mean = 3.3 ± 0.7	NS
Babaee, T	2014	**Milwaukee:**30 **TLSO:**30	**Milwaukee:** 16.6 ± 2.07 **TLSO**: 16.5 ± 2.3	SRS-22	**Milwaukee mean** = 3.04 ± 0.97 **TLSO mean** = 3.05 ± 0.58	NS
Brewer, P	2013	**Untreated patients with AIS:** 93	14.6 ± 1.8	SRS-22	Mean = 3.2 ± 0.8	NA
Cantele, F	2020	**AIS:** 139 **HC:** 134	**AIS:** 14.28 ± 1.82 **HC:** 14.75 (1.52)	SRS-22	**AIS mean** = 3.4 ± 0.72 **HC mean** = 3.8 ± 0.62	P = 0.000
Cheshire, J	2017	**Pre-operative patients:** 54	14.3 ± 1.29	SRS-22	Median = 2.65 (2.20–3.15)	NA
Çolak, T. K	2016	**Physiotherapy (P):**17 **Brace + Physiotherapy (BP):**25 **Surgery (S):**26	**P:** 14.1 ± 1.2 **BP:**13.7 ± 1.1 **S:**15.8 ± 1.2	SRS-23	**P mean** = 3.5 ± 0.5 **BP mean** = 3.7 ± 0.8 **S mean** = 4.3 ± 0.4	**P vs. S:** P = 0.000 **BP vs. S:** P = 0.000
Lee, H	2016	**Mild severity:** 52 **Moderate severity:** 46 **Severe:**12	14.2 ± 2.17 (Age by group not reported)	SRS-22	**Mild severity, median (IQR)** = 3.80 (2.20) **Moderate severity, median (IQR)** = 3.80 (3.00) **Severe, median (IQR)** = 3.00 (2.80)	**Severe vs. mild and moderate:** P = 0.031
Lendzion, M	2018	**Bracing and physiotherapy (BP):** 35 **Physiotherapy alone (P):** 55	**BP:** 16.5 ± 2.3 **P:** 16.6 ± 2.07	SRS-22	**BP mean** = 3.5 ± 0.6 **P mean** = 3.6 ± 0.6	NS
Lonner, B.S	2019	75	14.4 ± 1.6	SRS-22	**Pre-operation, mean** = 3.62 ± 0.59 **Post-operation, mean** = 4.18 ± 0.48	P < 0.001
Mariconda, M	2016	**AIS:** 87	14.8 ± 2.3	SRS-23	**Pre-operation, mean(females)** = 3.6 ± 0.3 **Post operation, mean (females)** = 3.9 ± 0.6 **Pre-operation, mean (males)** = 3.5 ± 0.2 **Post operation, mean (males)** = 4.0 ± 0.4	**Females and Males pre vs. post:** P < 0.01
Misterska, E	2012	**Conservative Treatment:** 36	13.4 ± 1.7	SRS-22	**1st evaluation, mean** = 3.7 ± 0.5 **2nd evaluation, mean** = 3.7 ± 0.5 **3rd evaluation, mean:** 3.9 ± 0.4	NS
Paolucci, T	2017	**Brace Group:** 16 **No-Brace Group:** 16	**Brace:** 14 ± 3.16 **No-Brace:** 14.14 ± 2.89	SRS-22	**Brace Group, mean** = 17.14 ± 2.93 **No-Brace mean** = 17.95 ± 2.96	NS
Pérez-Prieto, D	2014	**BMI < 18:**13 **BMI > 18:**26	**BMI < 18:** 16.38 ± 3.55 **BMI > 18:** 15.92 ± 2.24	SRS-22	**BMI < 18, mean** = 16.36 ± 1.96 **BMI > 18, mean** = 19.73 ± 3.17	P < 0.001

Table 3 (continued)

First Author	Year	N° subjects by group	Age (mean years ± SD)	SRS	Self- Image Subitem (Median or mean ± SD; range = 1–5)	Group differences
Soliman, H	2018	**Group 1 (Major curve angles of 90°-120°):** 76 **Group 2 (Major curve angles > 120°):** 61 **Group 3 (HC):** 50	**Group 1:** 15.3 ± 2.1 **Group 2:** 15.8 ± 1.8 **Group 3:** 16.1 ± 2.3	SRS-22r	**Group 1, mean** = 1.93 ± 0.3 **Group 2, mean** = 1.45 ± 0.3 **Group 3, mean** = 4.1 ± 0.6	**Overall group differences:** P < 0.001
Wang, L	2014	**One curve group:** 42 **Double curve group:** 102 **Three curve group:** 58	14.18 ± 1.42	SRS-22	**One curve group, mean** = 3.08 ± 0.52 **Double curve group, mean** = 3.07 ± 0.53 **Three curve group, mean** = 3.12 ± 0.51	NS
Watanabe, K	2005	**Mild scoliosis:** 43 **Moderate scoliosis:** 72 **Severe scoliosis:** 26 **HC:** 72	13.6	SRS-24	**Mild scoliosis, mean** = 10.5 ± 1.4 **Moderate scoliosis, mean** = 9.8 ± 1.9 **Severe scoliosis, mean** = 9.2 ± 1.8 **HC, mean** = 9.8 ± 1.7	**HC vs. AIS:** P < 0.05
Watanabe, K	2007	**Post-surgery patients:** 81	14.1	SRS-24	Mean = 3.1 ± 0.7	NA
Yagci, G	2018	**BBAT group:** 10 **TE group:** 10	**BBAT group** 14.20 ± 2.04 **TE group** 13.60 ± 1.65	SRS-22	**BBAT Pre-treatment, mean** = 3.46 ± 0.45 **BBAT Post-treatment, mean** = 3.62 ± 0.82 **TE Pre-treatment, mean** = 3.52 ± 0.53 **TE Post-treatment, mean** = 3.51 ± 0.71	NA
Yagci, G	2020	**AIS:** 96	15.9 ± 3.6	SRS-22	Self-image subitem: 3.3 ± 0.8	NA

HC healthy controls, *BMI* body mass index, *TLSO* thoraco−lumbosacral orthosis, *SRS* Scoliosis Research Society questionnaire, *IQR* interquartile range, *BBAT* basic body awareness therapy, *TE* traditional exercises, *NS* not significant, *NA* not applicable. Groups names are in bold

with surgery compared with traditional physiotherapy and brace treatments, p = 0.000 (Çolak et al., 2017) and five studies not reporting significant differences between different treatment approaches (Babaee et al., 2014; Lendzion et al., 2018; Misterska et al., 2013; Paolucci et al., 2017; Yagci et al., 2018).

Among the three articles comparing AIS and healthy control groups, lower body image subitem scores were always found in the AIS population, respectively (Cantele et al., 2020) p = 0.000, (Soliman, 2018) p < 0.00, and (Watanabe et al., 2005) p < 0.05.

In both of the two studies assessing self-image subitem scores contrasting AIS girls having normative Body Mass Index (BMI) and those with BMI values below normality-threshold score, lower values were found in the underweight girls (Cantele et al., 2020; Pérez-Prieto et al., 2014).

Body image outcomes assessed by instruments other than SRS questionnaires highlighted general body image alterations as well (See Table 4).

Two articles assessing body image perception by comparing milder and more severe scoliosis cases, found lower body image scores in the latter group, specifically: one study evaluating a group of patients with the Cobb angle of more than 40 degrees reported lower self-body, ideal-body and self-ideal subitems scores of SAQ compared with the less severe group at different evaluation intervals (see Table 4) (Schwieger et al., 2016); another study assessing body image disturbance through the BIDQ-S, found significantly higher scores (higher body image disturbance), in the group having the major curve angle (p < 0.001) (Soliman, 2018).

Table 4 Other measures of body image alterations

First author	Year	N° subjects by group	Age (mean years ± SD)	SRS	Self- image subitem (Mean ± SD/ Median)	P values
Çolak, T. K	2016	**Physiotherapy (P):**17 **Brace + Physiotherapy (BP):**25 **Surgery (S):**26	**P:** 14.1 ± 1.2 **BP:**13.7 ± 1.1 **S:**15.8 ± 1.2	WRVAS	**P mean =** 14.0 ± 5.9 **BP mean =** 14.0 ± 3.2 **S mean =** 7.0 ± 2.7	**P vs. S:** P = 0.000 **BP vs. S:** P = 0.000
Duramaz, A	2018	**AIS:** 41 **HC:** 52	**AIS:**15.3 ± 1.5 **HC:** 15.57 ± 1.46	BCS	**AIS pre-operative, mean =** 91.7 ± 24.9 **AIS post-operative, mean =** 74.2 ± 20.0 **HC:** 79.4 ± 21.3	**AIS pre vs. HC:** P = 0.010 **AIS pre vs. post:** P = 0.000
Lendzion, M	2018	**Bracing and physiotherapy (BP):** 35 **Physiotherapy alone (P):** 55	**BP:** 16.5 ± 2.3 **P:** 16.6 ± 2.07	TAPS	**Bracing and Physiotherapy mean TOT** = 3.2 ± 0.8 **Physiotherapy alone mean TOT** = 3.8 ± 0.6	**P vs. BP:** P = 0.04 (P alone better total TAPS)
Lonner, B.S	2019	75	14.4 ± 1.6	BIDQ-S	**BIDQ-S pre-operative, mean =** 1.64 ± 0.51 **BIDQ-S post-operative, mean =** 1.22 ± 0.38	**Pre vs. Post:** P < 0.0001
Misterska, E	2011	40	15 ± 31.5	SAQ	SAQ (two years post-surgery) **Median general score** = 34.48	NA
Misterska, E	2012	**Conservative Treatment:** 36	13.4 ± 1.7	TAPS	**TAPS total 1st evaluation mean** = 3.6 ± 0.6 **TAPS total 2nd evaluation mean** = 3.9 ± 0.5 **TAPS total 3rd evaluation mean** = 4.0 ± 0.4	**2nd vs. 3rd evaluations:** P = 0.005
Misterska, E	2014	41	13.60 ± 1.6	SAQ	**SAQ mean total score** = 2.7 ± 0.6	NA
Paolucci, T	2017	**Brace Group:** 16 **No-Brace Group:** 16	**Brace:** 14 ± 3.16 **No-Brace:** 14.14 ± 2.89	TAPS	**Brace Group mean** = 3.90 ± 0.44 **No-Brace mean** = 3.97 ± 0.47	NS
Perez-Prieto, D	2014	**BMI < 18:** 13 **BMI > 18:** 26	**BMI < 18:**16.38 ± 3.55 **BMI > 18:**15.92 ± 2.24	BSQ-14	**BMI < 18, mean:** 46.62 ± 8.86 **BMI > 18, mean:** 37.69 (15.15) 19 of 39: self-perception disorder (BSQ-14 > 40)	**BMI < 18 vs. BMI > 18** P = 0.05

Table 4 (continued)

First author	Year	N° subjects by group	Age (mean years ± SD)	SRS	Self- image subitem (Mean ± SD/ Median)	P values
Schwieger, T	2016	**< 40 Degree Largest Cobb Angle:** 257 **> 40 Degree Largest Cobb Angle:** 42	Range: 10–15 years	SAQ	**< 40 Degree Largest Cobb Angle:** T1 = Self-body: 16.8 (4.1) Ideal body: 19.3 (10.4) Self-ideal: 36.2 (12.7) T2 = Self-body: 17.5 (4.5) Ideal body: 19.2 (10.1) Self-ideal: 36.7 (12.6) T3 = Self-body: 17.5 (4.7) Ideal body: 19.2 (10.7) Self-ideal: 36.7 (13.2) T4 = Self-body: 17.3 (4.7) Ideal body: 19.2 (10.2) Self-ideal: 36.5 (12.7) **> 40 Degree Largest Cobb Angle:** T1 = Self-body: 19.9 (5.4) Ideal body: 21.5 (10.0) Self-ideal: 41.4 (13.0) T2 = Self-body: 21.9 (5.6) Ideal body: 22.4 (10.8) Self-ideal: 44.3 (13.9) T3 = Self-body: 20.7 (6.0) Ideal body: 19.6 (9.6) Self-ideal: 40.2 (12.9) T4 = Self-body: 21.9 (5.3) Ideal body: 22.7 (10.0) Self-ideal: 44.6 (12.9)	**< 40 Degree Largest Cobb Angle vs. > 40:** **T1:** **Self-body:** P = 0.0002 **Self -ideal:** P = 0.01 **T2** **Self-body:** P < 0.0001 **Ideal body:** P = 0.02 **Self -ideal:** P = 0.0002 **T3** **Self-body:** P = 0.0001 **Self -ideal:** P = 0.004 **T4** **Self-body:** P < 0.0001 **Self -ideal:** P < 0.0001 **Brace vs. Observation treatments:** NS
Schwieger, T	2017	**Most adherent Brace wear:** 92 **Least adherent Brace wear:** 39	**Most adherent:** 11.7 ± 1.1 **Least adherent:** 11.8 ± 1.1	SAQ	**SAQ total Baseline (Most adherent):** 36.9 ± 12.0 **SAQ total Baseline (Least adherent):** 34.8 ± 9.9	NS
Soliman, H	2018	**Group 1 (Major curve angles of 90°-120°):** 76 **Group 2 (Major curve angles > 120°):** 61	**Group 1:**15.3 ± 2.1 **Group 2:**15.8 ± 1.8	BIDQ-S	**Group 1 BIDQ-S, mean =** 3.38 ± 0.76 **Group 2 BIDQ-S, mean =** 4.5 ± 0.61	**Group 1 vs. Group2:** P < 0.001
Yagci, G	2018	**BBAT group:** 10 **TE group:** 10	**BBAT group:** 14.20 ± 2.04 **TE group:** 13.60 ± 1.65	WRVAS	**BBAT Pre-treatment, mean =** 21.00 ± 3.83 **BBAT Post-treatment, mean =** 13.30 ± 2.50 **TE Pre-treatment, mean =** 20.20 ± 2.94 **TE Post-treatment, mean =** 17.50 ± 2.68	**Pre vs. Post BBAT:** P < 0.05 **Pre vs. Post TE:** P < 0.05

WRVAS Walter Reed Visual Assessment Scale, *SAQ* Spinal Appearance Questionnaire, *BCS* Body Cathexis Scale, *TAPS* Trunk Appearance Perception Scale, *BSQ-14* Body Shape Questionnaire, *BIDQ-S* Body Image Disturbance Questionnaire-Scoliosis, *BBAT* Basic Body Awareness Therapy, *TE* Traditional Exercises, *ABC* Awareness-Body-Chart, *NS* not significant, *NA* not applicable. Groups names are in bold

Different Treatment Approaches' Impact on Body Representations

Among the eight studies evaluating the effects of different treatments on body image perception, two reported post-surgery improvements in BCS scores (Duramaz et al., 2018) $p = 0.000$ and BIDQ-S scores (Lonner et al., 2019) $p < 0.0001$ and one found a better impact of surgery over other traditional types of treatments assessed with the WRVAS $p = 0.000$ (Çolak et al., 2017). Brace effects on body image disturbances were assessed in three studies providing conflicting evidence: one reported physiotherapy-alone as a better practice than combined with brace in improving body image perception, as assessed by TAPS, ($p = 0.04$), (Lendzion et al., 2018); the other two did not find significant body image score differences between a group under orthotic treatment compared with a group not treated with brace (Paolucci et al., 2017), and comparing poor adherent and most adherent to brace treatment groups (Schwieger et al., 2017).

When compared with healthy control groups, AIS scored lower in the BCS $p = 0.010$ (Duramaz et al., 2018).

Correlations Between Altered Body Representations and Clinical Parameters of Scoliosis Severity

Fifteen of the 27 included articles considered correlations between body image scores and clinical or demographic variables (see Table S2).

Within the six studies analyzing correlations between participants' age and self-image scores (Babaee et al., 2014; Cheung et al., 2002; Çolak et al., 2017; Lendzion et al., 2018; Misterska et al., 2013; Wang et al., 2014a, 2014b), just one found a significant negative correlation, with outcomes worsening at increasing age (Lendzion et al., 2018) (TAPS, $p = 0.03$; SRS-22, $p = 0.01$).

The relation between sex and scoliosis was inquired just in two studies (Cheung et al., 2002; Lendzion et al., 2018), with one finding lower body image scores in female groups assessed with Trunk Appearance Perception Scale and SRS-22 (respectively $p = 0.027$; $p = 0.015$) (Lendzion et al., 2018).

Curve localization didn't correlate with self-image scores in any of the 8 studies investigating this relation (Asher et al., 2002; Babaee et al., 2014; Cantele et al., 2020; Cheung et al., 2002; Çolak et al., 2017; Lendzion et al., 2018; Lonner et al., 2019; Wang et al., 2014a, 2014b).

Conversely, correlations between scoliosis severity (Cobb angle) and self-image scores were significant in 7 out of 10 studies, revealing a negative correlation between scoliosis severity and satisfaction with body image as assessed by SRS questionnaires and Trunk Appearance Perception Scale (Brewer et al., 2013; Cantele et al., 2020; Cheung et al., 2002; Lendzion et al., 2018; Misterska et al., 2013; Wang et al., 2014a, 2014b; Watanabe et al., 2007), (see Table S2 of the supplementary materials for details).

Three studies explored the correlations between self-image and other radiographic measures (Risser sign, Apical Vertebral Translation and Apical Vertical Rotation). No significant relations emerged related to the Risser sign (Babaee et al., 2014; Wang et al., 2014a, 2014b) while contrasting results are reported with regards to apical vertebral rotation and translation: one study found a significant negative correlation between the apical vertical translation and SRS self-image subitem ($r = –0.290$, $p < 0.001$) but no relation with the vertebral rotation component (Wang et al., 2014a, 2014b), while another one found a significant correlation between the latter and the SRS-24 self-image domain ($r = -0.30$, $p < 0.01$), (Watanabe et al., 2007).

Nine studies explored the impact of other clinical measures (Angle of Trunk Rotation, hump height, BMI) on self-image. Most of them did not find clear relations. However, in four studies significant correlations between body image scores and the angle of trunk rotation (Lendzion et al., 2018; Misterska et al., 2011)(respectively: $r = – 0.228$, $p = 0.03$ and $r = – 0.91$, $p = 0.002$) and hump height (Mariconda et al., 2016; Wang et al., 2014a, 2014b) (respectively: $r = -– 0.04$, $p < 0.00$ and $r = –0.277$, $p < 0.001$) is reported.

Three studies adopting surface topography measures and asymmetry parameters revealed a weak correlation with self-image scores (Asher et al., 2002; Brewer et al., 2013; Cheshire et al., 2017).

Adolescent Idiopathic Scoliosis and Body Schema

Body schema alterations were assessed in four out of the twenty-seven included studies (See Table 2).

In two of these, perception of verticality and body posture were assessed: one study (Cheung et al., 2002) aiming to assess the perception of body employing vertical and horizontal laser line projections in darkness, failed to find significant task performance differences with a control group. However, a significant correlation between the amplitude of the angle formed by the horizontal and vertical line projections and the severity of scoliosis (Cobb angle) was observed ($r = 0.16$; $p = 0.045$). In the second study(Le Berre et al., 2019), altered perception of subjective postural and visual verticality were assessed in both static and dynamic optokinetic stimulation conditions. The scoliosis group had significantly altered perception of postural verticality compared with a matched-age control group ($p = 0.00023$) which was significantly correlated with the clinical frontal tilt ($p = 0.007$), while no differences were found for the subjective visual vertical.

Two other studies, investigating trunk misalignment awareness and general body perception awareness, found

respectively significant differences between the subjective perception of trunk curvature and the objective trunk curvature (underestimation at thoracolumbar district and overestimation at thoracic and lumbar districts, $p < 0.016$) (Picelli et al., 2016) and overall lower body awareness in AIS group compared with a matched-age control group ($p < 0.001$) (Yagci et al., 2020).

Discussion

Body representation disorders are a severe and widespread phenomenon among adolescents affected by idiopathic scoliosis. Nevertheless, literature on this topic is still in its infancy, mainly focused on detecting predisposing-genetic determinants and finding treatments for scoliosis biomechanical alterations. The lack of a general framework on the scoliosis-associated disorders of body representations led to the need of this review. This study highlighted evidence of high prevalence of body representation alterations in this clinical population, which require proper assessment and treatment. The small number of articles on this topic characterized by methodological heterogeneity, mostly adopting self-reported assessment scales, stressed the need for further studies and appropriate assessment procedures.

Body Image

The first finding of this review answers the first research question, pointing to the high prevalence of body image dissatisfaction among adolescents having idiopathic scoliosis, which always emerged regardless of assessment instruments. Adolescents with scoliosis scored consistently lower in self-image compared with healthy adolescents (Cantele et al., 2020; Yagci et al., 2020) and in those presenting a more severe degree of deformity compared with milder severity cases (Asher et al., 2004; Brewer et al., 2013; Lee et al., 2016; Lendzion et al., 2018; Schwieger et al., 2016; Soliman, 2018; Watanabe et al., 2005, 2007). However, all these studies employed questionnaires to assess body image, particularly the SRS designed to assess general aspects of quality of life more than body image. This should be considered as a general bias of all these works, giving the intrinsic limitations characterizing self-reported scales. Proper assessment procedures should be considered, such as those used in other conditions characterized by body image disorders, like in eating disorders (Caspi et al., 2017), (e.g., computerized tasks, structured clinical interviews, etc.).

The second research question aimed to explore the impact of different treatment options on body image, which was assessed in a fair number of studies (8/27). The strongest evidence of improvement in self-image is given by surgery. When interpreting these data, should be considered that patients undergoing surgery present more severe scoliosis deformations (mean degree Cobb angles > 50° Duramaz et al., 2018; Lonner et al., 2019; Mariconda et al., 2016)) if compared with those treated with less invasive interventions. Post-surgery body aesthetic changes are more easily detectable and thus more capable to influence the self-reported measure of body image perceptions. However, it was found that severity of deformity does not necessarily correlate with patients' decision to undergo surgery, which suggests again possible discrepancies between psychological beliefs/attitudes on body image and objective measures of physical appearance (Borges et al., 2017). This is even more evident when considering the effects of brace and physiotherapy on body image, with brace-wearing found not to affect body image per se, regardless of its esthetic impact (Cantele et al., 2020; Schwieger et al., 2016).

Finally, the third research question on the eventual associations between the perceived body image and the clinical parameters of scoliosis severity (i.e., Cobb angle, ATR, hump height) reveled not clear association between the two. Radiographic measures were found to be more often related to body image perception indices, though correlations are weak (see Table S2). Surprisingly, the relation between spinal deformity surface measures (e.g., ATR, hump height) and body image is seldom considered, even if these immediately visible aesthetic deformities may have a substantial impact on body image perception. These last two points on the lack of association between objective measures and subjective perception of body image, need to be carefully considered as they entail the necessity to regularly examine body image in the adolescents affected by idiopathic scoliosis, although apparently not needing it.

Body Schema

This review's main finding regrading body schema disorders in adolescents with idiopathic scoliosis is the big gap in literature on this topic. Indeed, only 4 studies assessing body schema alterations were identified for inclusion in this review. These works provide indirect indices of abnormal central integration mechanisms in scoliosis, specifically, altered postural perception, abnormal subjective verticality perception and a lower awareness of the body, which seem to correlate with some clinical variables of scoliosis severity (Cobb angle (Cheung et al., 2006), frontal tilt (Le Berre et al., 2019)).

The paucity of literature on this topic is surprising, given the rising number of theories trying to address the contribution of CNS abnormalities to scoliosis curvature progression (Burwell & Dangerfield, 2002; Chu et al., 2011). The etiology of idiopathic scoliosis includes sensorimotor integration deficits which lead to body schema representation abnormalities (Burwell et al., 2009). Thus, assessing

precociously body schema integrity in at risk subjects, may prevent scoliosis curvature worsening and give new insights for a better understanding of its etiopathogenesis. However, the development of new assessment instruments for body schema is mandatory to accomplish this aim.

Future Directions

In light of this review, future research on adolescents with idiopathic scoliosis should focus on the multifactorial nature of this condition, which is a spine disorder but with highly comorbid psychological consequences, and a complex etiopathogenesis. Specifically addressing body representation disorders which are both a precipitating and maintaining factor of this condition will in turn improve traditional treatments and provide new perspectives on its pathogenetic mechanisms. A theoretical comprehensive model of this is proposed in Fig. 2.

Conclusion

Body representation disorders are serious clinical conditions affecting adolescents with idiopathic scoliosis. However, up to date research on this topic is inadequate. This literature review clearly highlighted the prevalence of body image disorders and the lack of studies on body schema alterations. It also highlighted the lack of clear associations of these disorders with clinical measures of scoliosis severity as well as the absence of standardized assessments and treatment procedures. Body image dissatisfaction, negative attitudes toward perceived body appearance and body schema alterations should be considered as highly comorbid conditions of adolescents affected by scoliosis. Thus, when dealing with adolescents with scoliosis, clinicians should consider the multifactorial nature of this condition and set convenient assessments and treatments in association with the traditional ones (i.e., brace, surgery, and physiotherapy). Adolescent idiopathic scoliosis is a complex, multifactorial condition that would benefit from a multidisciplinary treatment approach, especially one that includes a focus on body representations.

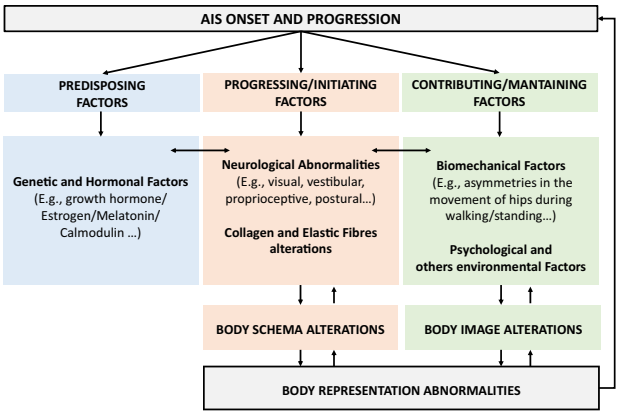

Fig. 2 Multifactorial model of AIS onset and progression. Schematic representation of the contribution of multiple factors concurring to adolescent idiopathic scoliosis onset and progression. Body schema and body image alterations are initiating and maintaining factors concurring to scoliosis progression

Appendix

PubMed search strategy.

Search number	Query	Sort by	Filters	Search details	Results
8	#3 AND #7		Humans, English, Child: birth-18 years	(("scoliosis"[MeSH Terms] AND ("humans"[MeSH Terms] AND "english"[Language] AND ("infant"[MeSH Terms] OR "child"[MeSH Terms] OR "adolescent"[MeSH Terms])) OR (("adolescences"[All Fields] OR "adolescency"[All Fields] OR "adolescent"[MeSH Terms] OR "adolescent"[All Fields] OR "adolescence"[All Fields] OR "adolescents"[All Fields] OR "adolescent s"[All Fields]) AND ("idiopathic"[All Fields] OR "idiopathically"[All Fields] OR "idiopathics"[All Fields]) AND ("scoliosis"[MeSH Terms] OR "scoliosis"[All Fields] OR "scolioses"[All Fields]) AND ("humans"[MeSH Terms] AND "english"[Language] AND ("infant"[MeSH Terms] OR "child"[MeSH Terms] OR "adolescent"[MeSH Terms])))) AND ("humans"[MeSH Terms] AND "english"[Language] AND ("infant"[MeSH Terms] OR "child"[MeSH Terms] OR "adolescent"[MeSH Terms])) AND ((("body image"[MeSH Terms] AND ("humans"[MeSH Terms] AND "english"[Language] AND ("infant"[MeSH Terms] OR "child"[MeSH Terms] OR "adolescent"[MeSH Terms]))) OR (("body image"[MeSH Terms] OR ("body"[All Fields] AND "image"[All Fields]) OR "body image"[All Fields] OR ("body"[All Fields] AND "schema"[All Fields]) OR "body schema"[All Fields]) AND ("humans"[MeSH Terms] AND "english"[Language] AND ("infant"[MeSH Terms] OR "child"[MeSH Terms] OR "adolescent"[MeSH Terms]))) OR (("body image"[MeSH Terms] OR ("body"[All Fields] AND "image"[All Fields]) OR "body image"[All Fields]) AND ("humans"[MeSH Terms] AND "english"[Language] AND ("infant"[MeSH Terms] OR "child"[MeSH Terms] OR "adolescent"[MeSH Terms])))) AND ("humans"[MeSH Terms] AND "english"[Language] AND ("infant"[MeSH Terms] OR "child"[MeSH Terms] OR "adolescent"[MeSH Terms]))	172
7	#4 OR #5 OR #6		Humans, English, Child: birth-18 years	("body image"[MeSH Terms] AND ("humans"[MeSH Terms] AND "english"[Language] AND ("infant"[MeSH Terms] OR "child"[MeSH Terms] OR "adolescent"[MeSH Terms])) OR (("body image"[MeSH Terms] OR ("body"[All Fields] AND "image"[All Fields]) OR "body image"[All Fields] OR ("body"[All Fields] AND "schema"[All Fields]) OR "body schema"[All Fields]) AND ("humans"[MeSH Terms] AND "english"[Language] AND ("infant"[MeSH Terms] OR "child"[MeSH Terms] OR "adolescent"[MeSH Terms]))) OR (("body image"[MeSH Terms] OR ("body"[All Fields] AND "image"[All Fields]) OR "body image"[All Fields]) AND ("humans"[MeSH Terms] AND "english"[Language] AND ("infant"[MeSH Terms] OR "child"[MeSH Terms] OR "adolescent"[MeSH Terms])))	10,508
6	Body image		Humans, English, Child: birth-18 years	"body image"[MeSH Terms] OR ("body"[All Fields] AND "image"[All Fields]) OR "body image"[All Fields]	10,431
5	Body schema		Humans, English, Child: birth-18 years	"body image"[MeSH Terms] OR ("body"[All Fields] AND "image"[All Fields]) OR "body image"[All Fields] OR ("body"[All Fields] AND "schema"[All Fields]) OR "body schema"[All Fields]	10,508
4	Body representation [MeSH Terms]		Humans, English, Child: birth-18 years	"body image"[MeSH Terms]	7,123

Search number	Query	Sort by	Filters	Search details	Results
3	#1 OR #2		Humans, English, Child: birth-18 years	("scoliosis"[MeSH Terms] AND ("humans"[MeSH Terms] AND "english"[Language] AND ("infant"[MeSH Terms] OR "child"[MeSH Terms] OR "adolescent"[MeSH Terms])) OR (("adolescences"[All Fields] OR "adolescency"[All Fields] OR "adolescent"[MeSH Terms] OR "adolescent"[All Fields] OR "adolescence"[All Fields] OR "adolescents"[All Fields] OR "adolescent s"[All Fields]) AND ("idiopathic"[All Fields] OR "idiopathically"[All Fields] OR "idiopathics"[All Fields]) AND ("scoliosis"[MeSH Terms] OR "scoliosis"[All Fields] OR "scolioses"[All Fields]) AND ("humans"[MeSH Terms] AND "english"[Language] AND ("infant"[MeSH Terms] OR "child"[MeSH Terms] OR "adolescent"[MeSH Terms]))	9,968
2	Adolescent idiopathic scoliosis		Humans, English, Child: birth-18 years	("adolescences"[All Fields] OR "adolescency"[All Fields] OR "adolescent"[MeSH Terms] OR "adolescent"[All Fields] OR "adolescence"[All Fields] OR "adolescents"[All Fields] OR "adolescent s"[All Fields]) AND ("idiopathic"[All Fields] OR "idiopathically"[All Fields] OR "idiopathics"[All Fields]) AND ("scoliosis"[MeSH Terms] OR "scoliosis"[All Fields] OR "scolioses"[All Fields])	4,609
1	Scoliosis[MeSH Terms]		Humans, English, Child: birth-18 years	"scoliosis"[MeSH Terms]	9,826

See Table 5.

Table 5 Articles quality assessment

Articles	Aim of the work	Selection bias (inclusion/exclusion criteria)	Performance Bias (data collection/processing)	Detection bias (Outcomes)	Results presentation	Statistical approach	Quality score (Max 12)
	The research question is clearly stated	Participant's inclusion and exclusion criteria are clearly defined	1. Data collection is clearly described and reliable 2. Presence of a control group	Outcomes are relevant to answer research question	Test/questionnaires/clinical scores are clearly reported	1.Statistical procedures performed are clearly described 2. Power analysis	
Asher et al. (2004)	2	1 (exclusion criteria not defined)	1 (lack of a control group)	2	2	1	9
Babaee et al. (2014)	2	2	1 (lack of a control group)	2	2	1	10
Brewer et al. (2013)	2	2	1 (lack of a control group)	2	2	1	10
Cheshire et al. (2017)	2	2	1 (lack of a control group)	2	2	1	10
Cheung et al. (2002)	2	1 (exclusion criteria not defined)	2	2	2	1	10
Çolak et al. (2017)	2	2	1 (lack of a control group)	2	2	1	10
Danielsson et al. (2012)	2	1 (exclusion criteria not defined)	1 (lack of a control group)	0	2	1	7 EXCLUDED
Duramaz et al. (2018)	2	2	2	2	2	2	12
Duri et al. (2019)	2	1 (exclusion criteria not defined)	1 (lack of a control group)	0	1	1	6 EXCLUDED
Le Berre et al. (2019)	2	2	2	2	2	1	11
Lee et al. (2016)	2	2	1 (lack of a control group)	2	2	1	10
Lendzion et al. (2018)	2	2	1 (lack of a control group)	2	2	1	10
Lonner et al. (2019)	2	2	1 (lack of a control group)	1	2	1	9
Mariconda et al. (2016)	2	2	1 (lack of a control group)	2	2	1	10
Misterska et al. (2011)	2	2	1 (lack of a control group)	1	1	1	8
Misterska et al. (2013)	2	2	1 (lack of a control group)	1	2	1	9
Misterska et al. (2014)	2	2	2	1	2	1	10
Paolucci et al. (2017)	2	2	2	2	2	1	11
Pérez-Prieto et al. (2014)	2	2	2	2	2	1	11
Picelli et al. (2016)	2	1	1 (lack of a control group)	2	2	1	10
Schwieger et al. (2016)	2	2	1 (lack of a control group)	2	2	1	10

Table 5 (continued)

Articles	Aim of the work	Selection bias (inclusion/exclusion criteria)	Performance Bias (data collection/processing)	Detection bias (Outcomes)	Results presentation	Statistical approach	Quality score (Max 12)
	The research question is clearly stated	Participant's inclusion and exclusion criteria are clearly defined	1. Data collection is clearly described and reliable 2. Presence of a control group	Outcomes are relevant to answer research question	Test/questionnaires/clinical scores are clearly reported	1.Statistical procedures performed are clearly described 2. Power analysis	
Schwieger et al. (2017)	2	2	1 (lack of a control group)	2	2	1	10
Soliman (2018)	2	2	2	2	2	1	11
Wang et al. (2014a, 2014b)	2	2	1 (lack of a control group)	2	2	1	10
Yagci et al. (2018)	2	2	2	1	2	1	10
Yagci et al. (2020)	2	1 (exclusion criteria not defined)	2	2	2	1	10

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Declarations

Conflict of interest The authors report no conflict of interests.

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