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Abstract

The aim of this chapter is to present a case study of why and how an umbrella review was conducted and reported within the context of the conservative management of adolescents with idiopathic scoliosis. We present and discuss the findings of an umbrella review of systematic reviews regarding both the screening and the effectiveness of nonsurgical interventions for adolescents with idiopathic scoliosis. The mandate for school-based screening programs for adolescents with scoliosis is a highly contentious and strongly debated issue. As a result of numerous controversial and discordant recommendations presented over a number of years by different societies and organizations that have been based on poor-quality systematic reviews, patients, therapists, insurance providers, policy makers, and researchers remain uncertain and confused. Nonsurgical interventions for adolescents with idiopathic scoliosis, including scoliosis-specific exercise interventions and bracing, are also controversial. We evaluated the validity as well as the methodological quality of all existing systematic reviews on this topic. We summarized, appraised, analyzed, and synthesized all available studies meeting the minimal criteria for a systematic review. The methodological quality of the majority of the included 27 systematic reviews, as assessed with the AMSTAR risk of bias tool, was disappointingly low. More, good-quality primary and secondary studies of

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higher level designs are urgently needed in this topic. We demonstrated the role of an umbrella review, as a research tool, to prevent misleading information and erroneous guidance for stakeholders, resulting from poorly conducted systematic reviews.

18.1 Introduction

18.1.1 General Assumptions

The volume of knowledge currently generated in medical journals worldwide is phenomenal [1, 2]. At the same time the science of evidence synthesis has also developed rapidly [3–5]. Furthermore the importance of high-quality bodies of evidence together with the evolution of the evidence based from primary clinical trials to high-quality systematic reviews has significantly strengthened the quality of evidence currently available within most scientific and medical fields [6]. This overwhelming amount of knowledge and information both in quantitative and in qualitative areas has resulted in the need for a novel method of research – a systematic and umbrella review of systematic reviews.

This handbook elaborates on the methodology, developments, and challenges, as well as the dilemmas regarding the conduct and reporting of methodological issues concerning umbrella reviews. The aim of this chapter is to present a case study of why and how an umbrella review was conducted and reported within the context of a specific clinical research question. In this chapter we present a case study on the conservative management of adolescents with idiopathic scoliosis (AIS). We demonstrate how a careful methodological analysis of current existing systematic reviews can provide health-care professionals as well as service users engaged in the care of patients with AIS with new evidence and information regarding the credibility of published secondary research papers. We present and discuss the findings of an umbrella review of systematic reviews regarding both the screening for AIS and the effectiveness of nonsurgical interventions in adolescents with AIS.

18.1.2 Why Was this Umbrella Review Needed?

Screening programs are legitimate, provided that early treatment interventions for patients diagnosed with AIS as a result of screening are effective [7] – on the other hand, patients with false-positive diagnoses may undergo unnecessary and potentially harmful treatments. In the case of AIS, screening and nonsurgical treatment methods are both the subject of controversial opinions and published papers with discordant results from systematic reviews. These have resulted in contradictory recommendations and statements that have frequently lead to opinion-based practice guidelines being developed (Tables 18.1 and 18.2). The selection of this case was also prompted

by the description of the specific role of the umbrella review in informing evidence-based decision-making, highlighted by Cornell and Laine in their editorial [8]:

(...) practice guidelines often require answers to not only one but several linked questions. A common example concerns screening interventions for which direct clinical trials that randomly assign participants to a screening or control group are lacking. In such cases, guideline developers typically seek answers to a sequence of interrelated questions: How prevalent is the condition? What is its natural history? Is a good screening test available? What are the benefits (and harms) of early detection and treatment? (...)

We performed an umbrella review to find answers to research questions regarding the reliability (for quantitative questions) and credibility (for qualitative questions) of all reviews addressing the screening as well as the nonsurgical management of a single condition – AIS, to inform policy makers and all engaged in the diagnosis, prevention, therapy, and education of people with this condition.

Systematic reviews, including those in the orthopedics [9], surgery [10], and physiotherapy [11] fields, vary in terms of their quality. Thus, another reason for this umbrella review was to assess the quality of the evidence from secondary studies, regarding the conservative management of AIS.

We aimed at evaluating the validity as well as the methodological quality of all existing systematic reviews on this topic. Our goal was also to examine which papers labeled as “systematic reviews” were in fact opinion pieces or narrative reviews.

18.1.3 Terminology and Methodological Issues: The Report as a Case Study within this Methods Handbook

Before we discuss the case study, we would like to address a number of methodological issues as well as issues concerning terminology characteristic for the comparatively new, developing, and evolving type of research synthesis methods: the umbrella review, which we believe are important in the context of the case report that we present.

Currently different terminologies are used to describe systematically developed reviews of systematic reviews (and, in some instances, also of primary studies). These terms include complex systematic reviews [12], overview of reviews [13, 14], systematic umbrella review [15], overview of systematic reviews [16, 17], umbrella systematic review [18, 19], analysis of systematic reviews [20], metaepidemiologic study [21], systematic review of systematic reviews [22], systematic review of systematic reviews and meta-analyses [23] or systematic review of meta-analyses [24], systematic map of systematic reviews [25], and even the term “survey” of reviews [26] that has been used. We will be using the term “umbrella review” throughout this chapter, even when referring to publications whose authors have used different terms for studies of this type.

The term “umbrella review” also has a number of different usages and understandings [13, 27–30]. We conducted an “umbrella systematic review of systematic reviews.” We did not include primary studies for analysis. We conducted an umbrella

review, as elaborated within the Cochrane Handbook [13] and the Joanna Briggs Institute's (JBI) [29] guidelines. We also followed the guidelines described by Ioannidis [27] and Grant and Booth [30].

Umbrella reviews of systematic reviews are needed not only to summarize large bodies of evidence [27, 28]. Different types of research questions are addressed within umbrella reviews. These have included the quality of reporting methods [31], financial conflicts of interest of review authors [20], reporting the quality of search methods in systematic reviews [26], reporting publication bias [21], as well as the handling of missing outcome data [23]. To our best knowledge, we conducted the first comprehensive, explicit, and systematic overview of systematic reviews addressing screening and nonsurgical interventions for adolescents with idiopathic scoliosis.

The rigor for the selection of systematic reviews for inclusion within umbrella reviews in terms of the design and type of systematic reviews is increasing. Umbrella review of meta-analyses [32], umbrella review of systematic reviews and meta-analyses of observational studies and randomized trials [33], and umbrella review of meta-analyses of observational studies [34] have all been published. Although rare and unusual, quantitative data syntheses (meta-analysis) are also possible within umbrella reviews [35, 36]. In our case study, we present the more typical umbrella review, with a narrative describing the evidence from the systematic reviews of any types of primary studies that were included.

18.2 Methodology and Reporting

In contrast to systematic reviews of primary studies, umbrella reviews lack dedicated, published reporting guidelines. We followed both the PROSPERO protocol, which is universal for systematic reviews of primary studies and overviews of systematic reviews, and the PRISMA reporting guidelines [37], which though originally developed for systematic reviews of primary studies is also applicable to umbrella reviews [38].

18.2.1 The Case Study on Adolescent Idiopathic Scoliosis: Report of the Umbrella Review

To address the whole subject matter regarding the two interrelated issues – the screening programs and the treatment methods – we divided our umbrella review process into two parts. The first part discusses the systematic reviews on school screening for AIS. The second part presents the systematic reviews on nonsurgical interventions for AIS. Such approaches have recently been used in both a published combined report of an “umbrella review of systematic reviews” and “a systematic review of primary studies regarding conservative interventions for tendinopathy” [16]. Similar methodologies have also been reported in an umbrella review of systematic reviews and guideline documents addressing diet and physical activity interventions and policies [39].

Protocol registration and resource publications. This chapter is based on our study “Effects of nonsurgical management for patients with idiopathic scoliosis. An overview of systematic reviews” that has been registered at PROSPERO (CRD42013003538) and has previously been presented in two reports, regarding screening [40] and nonsurgical interventions [41].

18.2.2 Background

18.2.2.1 Description of the Health Problem

The prevalence of AIS is estimated at 2–4 % of children aged 10–16. The ratio of girls to boys ranges from 1:1 for spinal lateral deviations (curve angles) below 10° to over 10:1 for curves exceeding 30°. The risk of progression of untreated scoliosis depends both on the skeletal maturity and the size of the curve. The tendency for progression is also more frequent among girls [42–45]. The deformity may have lasting consequences and can be accompanied with pulmonary complications (life endangering in patients with very severe curves), pain symptoms, difficulties in participation, and psychological disorders [42–46].

18.2.2.2 Screening and Nonsurgical Management

As a prevalent condition, with individual variations, affecting both the physical and psychosocial functioning in the long term, AIS is considered by numerous clinicians, researchers, and authors to be an “important healthcare problem meeting the requirements for a screening program” [47, 48].

Nonsurgical interventions for the treatment of adolescents with AIS typically constitute a variety of physical modalities: braces of various types and modes of application, scoliosis-specific exercises, as well as a number of diverse physical therapy modalities such as electrical stimulation, manual therapy, and different types of osteopathic and chiropractic interventions [49–51].

18.2.2.3 National and International Guidelines and Recommendations

School screening The mandate for school-based screening programs for adolescents with scoliosis is a highly controversial, strongly debated issue [52–54]. As a result of controversial and discordant recommendations, in the USA, screening has remained mandatory in some US States (Arkansas, Alabama, California, Florida, Pennsylvania, Texas, Utah) is recommended in Minnesota, is not required in Montana and Oregon, and was repealed in Indiana and Maryland. Table 18.1 summarizes the recommendations and position statements of different institutions. The table also classifies the available documents as either opinion-based statements or systematically developed practice guidelines.

Conservative treatment Nonsurgical interventions for adolescents with AIS are subject to similar controversies. To illustrate the polarity, examples of opinions from experts can be seen in Table 18.2.

Table 18.1 Summary of current recommendations regarding school screening for scoliosis

Developer/initiative (year) ^a	Recommendation	Type
Canadian Task Force on Preventive Health Care (1994)	“insufficient evidence (...) to indicate that screening for idiopathic scoliosis in adolescents is either effective or ineffective in improving the outcome”	EB
National Health and Medical Research Council, Australia (2002)	“Good evidence to recommend against screening”	EB
National Self-Detection Program for Scoliosis ^b (Spine Society of Australia, current as of April 2014)	Two-tier self-detection program for girls, replacing school screening programs	N
Italian guidelines ^c (2005)	“School screening programs (...) should be conducted”; “scientific evidence”: E2 – fair scientific consensus	CB/EB
SRS-AAOS-POSNA-AAAP ^d position statement (2008)	“Societies recognize the benefits that can be provided by effective clinical screening programs” and “do not support any formal recommendations against scoliosis screening, given the available literature”	CB
Society for Spinal Orthopaedic and Rehabilitation Treatment, SOSORT (2012)	School screening useful for clinical purposes; several improvements to the programs postulated	CB/EB
Ministry of Health Malaysia, HTA Section (2009)	“Screening for scoliosis among school children is recommended only for high risk group such as girls at 12 years or age”; fair level of evidence to suggest that school scoliosis screening program was able to detect scoliosis at a younger age and with smaller Cobb angle; [and] reduce the frequency of surgical treatment; evidence to suggest its cost-effectiveness	EB
UK National Screening Committee ^e (2012)	“Screening should not be offered,” “systematic population screening programme not recommended,” “(...) there is little evidence that screening would be necessary to pick up children needing surgery. (...) many children would be subjected to unnecessary X-rays and treatment, which may themselves be harmful. This could cause unnecessary stress to children and their families”	EB
Institute for Clinical Systems Improvement (2013)	Recommendations from USPSTF (2004) and SRS-AAOS-POSNA-AAAP (2008) reported; level III service: “(...) the evidence is currently incomplete (...); providing these services is left to the judgment of individual medical groups, clinicians, and their patients”; the SRS-AAOS-POSNA-AAAP position statement evaluated as “low quality evidence”	EB

Table 18.1 (continued)

Developer/initiative (year) ^a	Recommendation	Type
Scoliosis Research Society International Task Force on Scoliosis Screening (2013)	“(…) an expert panel supports scoliosis screening in 4 of the 5 domains (…) of the World Health Organisation criteria for a valid screening procedure”	CB/EB
US Preventive Services Task Force/Agency for Healthcare Research and Quality (2004, 2012)	“Do not screen for idiopathic scoliosis”; recommendation against; grade D	EB (2004)
US Preventive Services Task Force/Agency for Healthcare Research and Quality (2014)	Idiopathic scoliosis not among recommendations	n/a
“Bright Futures” initiative ^f (current as of April 2015)	The Bright Futures/AAP periodicity schedule does not include screening for scoliosis	N
American Academy of Family Physicians (2015)	“The AAFP recommends against the routine screening of asymptomatic adolescents for idiopathic scoliosis (2004)” ^g ; grade D (USPSTF classification, prior to 2007) ^h	EB (USPSTF, 2004)

EB evidence based, CB consensus based, N narrative describing a program or recommendations, n/a not applicable

^aFor references see resource publication [40]

^bEndorsed by the Paediatrics and Child Health Division of the Royal Australasian College of Physicians

^cEndorsed and approved by many Italian professional bodies, mandated by the Italian Ministry of Health

^dScoliosis Research Society, American Academy of Orthopedic Surgeons, Pediatric Orthopedic Society of North America, American Academy of Pediatrics; AAP endorsed the position statement; however, AAP also leads the “Bright Futures” initiative

^eApproved by the British Orthopaedic Association, British Scoliosis Society, Institute of Child Health, Royal College of General Practitioners, Royal College of Surgeons, Scoliosis Association

^fLaunched under the leadership of the Maternal and Child Health Bureau of the Health Resources and Services Administration, led by AAP and partnered by numerous agencies, groups, and organizations [<http://brightfutures.aap.org>]

18.2.3 Methods

18.2.3.1 Inclusion and Exclusion Criteria

To deliver the most comprehensive evidence synthesis possible, we summarized, appraised, analyzed, and synthesized all available studies meeting the minimal criteria for a systematic review. Papers were considered as systematically developed reviews if they reported on methods to search, identify and select studies, and critically appraised relevant evidence [55]. We considered all systematic reviews with

Table 18.2 Opinions regarding nonsurgical interventions for adolescents with idiopathic scoliosis

<i>Negative comments:</i>
“Time and common sense prevent me from discussing any other treatment modality than bracing”
“Treatment options for patients with scoliosis range from the unproven or harmful to the beneficial”
“Physical therapy, chiropractic care, biofeedback and electric stimulation have not been shown to alter the natural history of scoliosis”
“Patients should be aware of the absence of evidence for these [physiotherapy] treatments”
<i>Positive statements:</i>
“Bracing and spinal surgery have been proven to alter the natural history of curve progression”
“Exercise-based therapies, alone or in combination with orthopaedic approaches, are a logical approach to improve and maintain flexibility and function in patients at risk for pain, pulmonary dysfunction, and progression”
“The triad of out-patient physiotherapy, intensive in-patient rehabilitation and bracing has proven effective in conservative scoliosis treatment in central Europe”

For references see resource publication [41]

meta-analyses as well as qualitative systematic reviews with descriptive syntheses of findings from individual studies. To address research questions other than effectiveness – risk of adverse effects, prevalence, and test accuracy – and to allow for the analysis of the methodological rigor of the systematic reviews that were included, we did not limit the inclusion criteria to systematic reviews of randomized controlled trials, but considered systematic reviews of any types of primary studies, including those of different types of non-randomized studies.

18.2.3.2 Search Strategy

We prioritized databases and other resources and began searching the potentially more productive databases (this approach termed or known as “purposive searching” [5] is described as being more efficient and less time-consuming than the typical comprehensive search strategies applied within systematic reviews of primary studies) [56]. These databases included the following types: databases of systematic reviews, databases with separate indexing of systematic reviews, guideline registries, general bibliographic databases, and then websites of relevant institutions. Search strategies, keywords, and limits are detailed in the resource publications [40, 41], and their detailed reporting exceeds the volume of this chapter. To document the process of searching, Table 18.3 has been included and shows databases and other resources that were searched in the “screening” umbrella review [40] together with the order of the search. In the “interventions” review [41], we performed a similar search, but with details specific to the different research question.

18.2.3.3 Study Selection

We independently conducted the searches as well as the initial selection of studies by their title and/or abstract. Full papers were then examined for eligibility. Disagreements were resolved by discussion. The combined PRISMA search flow for the selection of included studies for the two parts of the umbrella review is shown in Fig. 18.1.

Table 18.3 Databases searched in the screening umbrella review together with the “purposeful” [5] order of searching beginning with the potentially more productive databases [40]

Databases of systematic reviews, guideline registries and databases with separate indexing of systematic reviews and guidelines:

The Centre for Reviews and Dissemination databases – DARE, HTA, NHSEED, Cochrane Database of Systematic Reviews (CDSR), Joanna Briggs Institute, Campbell Library, Cochrane Effective Practice and Organisation of Care (EPOC) Group, the AHRQ databases and resource lists from USPSTF, AHRQ Evidence-based Practice Centers (EPC Reports) and National Guideline Clearinghouse, PEDro, INAHTA, and TRIP

Websites of institutions:

USPSTF, CTFPHC, NHMRC, UK Screening Portal/UK NSC Policy Database, Scottish Intercollegiate Guidelines Network (SIGN), National Institute for Health and Care Excellence, UK (NICE)

General bibliographic databases:

MEDLINE through PubMed, Web of Science, and SportDiscus through EBSCO and Google Scholar

Gray literature:

Registered protocols, reviews in progress, guidelines in development, and registered titles:

PROSPERO, CDSR, the USPSTF registry of the topics in progress, the CTFPHC protocols, HSR Project Database, NICE, AHRQ EPC Reports database (for the EPC Reports in Progress), HSRProj Database, the NHMRC website and the Systematic Review Data Repository (SRDR) database, the Conference Proceedings Citation Index – Science from the Web of Knowledge

For abbreviations not explained here, see Tables 18.1 and 18.5

18.2.3.4 Scope of the Systematic Reviews

Inclusion and exclusion criteria, formulated to the two umbrella reviews according to the PEO (problem/population–exposure/issue–outcome) and PICO (problem/population–intervention–comparator/control–outcome) principles, respectively, are summarized in Table 18.4.

18.2.3.5 Types of Outcomes

We analyzed and summarized both patient relevant and surrogate (or intermediate) outcomes of both the screening of patients with AIS as well as the effectiveness of nonsurgical interventions, taking into account qualitative and other not “numeric” issues. For us this is especially relevant as regards AIS – surrogate outcomes (e.g., curve angle, angle of trunk rotation, curve progression) are frequent in the available systematic reviews whereas patient-oriented ones (e.g., quality of life, body image) are less popular and frequent.

18.2.3.6 Data Extraction, Methodological Quality, and Level of Evidence Assessment

Data Extraction

We independently extracted the data, using predefined data extraction forms. Discrepancies were resolved through discussion.

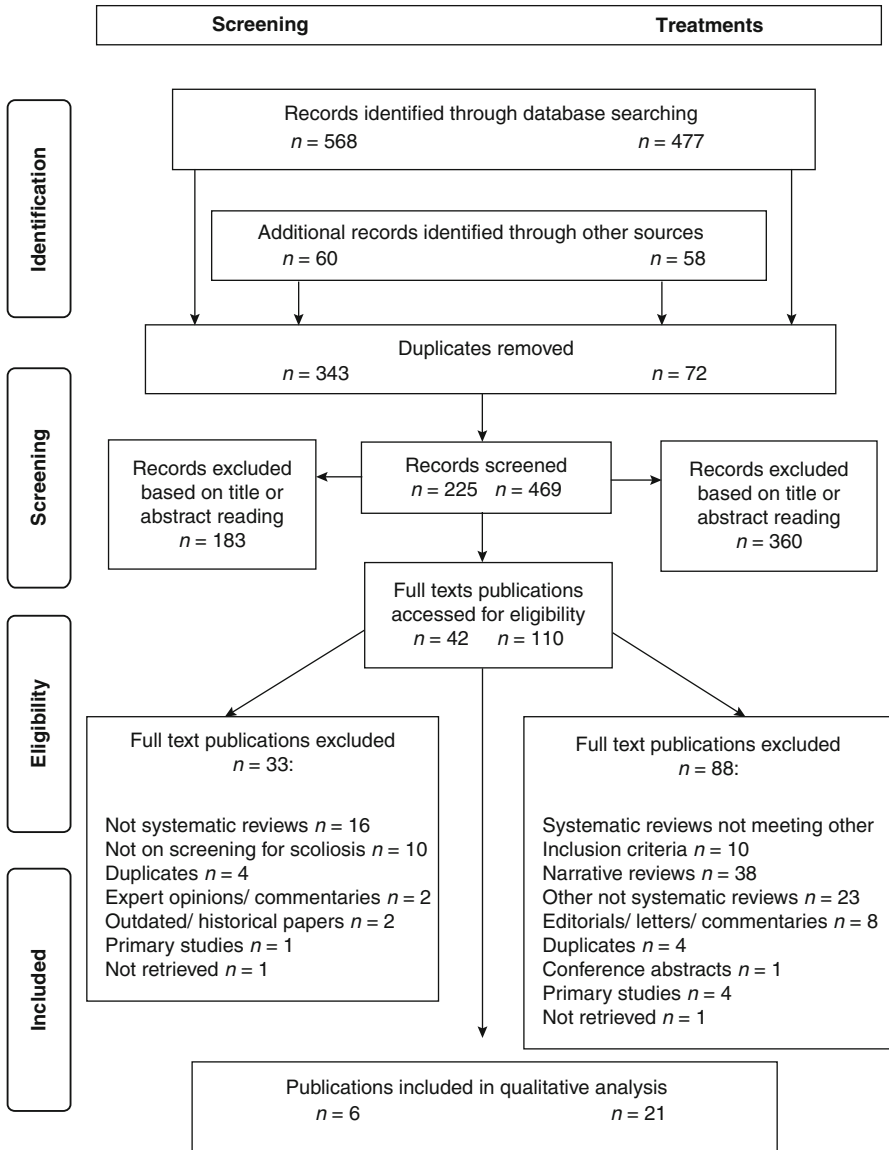


Fig. 18.1 Compiled PRISMA flow diagrams for the selection of included systematic reviews for screening (*left*) and treatment methods (*right*)

Methodological Quality

We used the “Assessment of Multiple Systematic Reviews” (AMSTAR) risk of bias tool [57] to assess the methodological quality of included reviews. The AMSTAR comprises 11 items addressing criteria relating to the assessment of methodological rigor (Table 18.1). The items are scored “yes,” “no,” “cannot answer,” or “not

Table 18.4 Criteria for inclusion and exclusion in the screening and intervention reviews

	Inclusion criteria	Exclusion criteria
<i>Screening review:</i>		
Population	Schoolchildren, both girls and boys, with no geographical or other (e.g., societal, racial, cultural) restrictions, within the growth spurt associated with the risk of development of AIS, typically 10–12 years of age; however, no strict age criteria were defined	Papers including other populations of children, e.g., with comorbidities, such as Duchenne muscular dystrophy
Exposure or issue	Any reviews addressing “school screening for AIS”	Papers addressing other screening programs that did not exclusively address screening tests for adolescent idiopathic scoliosis, e.g., general health examinations; screening programs for other types of scoliosis were also excluded (e.g., adult scoliosis)
Outcomes	Primary outcome: any recommendation that stated “to recommend screening or not to recommend screening, i.e., a “yes” or “no” with regard to the authors’ recommendations; also any secondary outcomes	Not specified
<i>Intervention review:</i>		
Population	Systematic reviews addressing adolescents of both genders with AIS, diagnosed and managed between the ages of 10–18 years, with no restriction as to bone age (Risser sign), with mild, moderate, and/or severe AIS (11–24°, 25–44°, and 45° Cobb and greater, respectively)	Reviews on early-onset (infantile or juvenile) scoliosis, reporting on scoliosis secondary to other conditions, e.g., Duchenne dystrophy, cerebral palsy, spinal cord injury, neurofibromatosis
Interventions	Nonsurgical interventions applied as a sole treatment or as combinations of different nonsurgical interventions: braces of any type (both rigid and soft) and mode of application (any number of hours a day or nighttime); any approach (s) or “school” of scoliosis-specific exercise treatment, regardless of the severity of the deformity, both as a single intervention and as part of a group of different complex interventions, e.g., supplementing brace treatment (add-on treatment); chiropractic; manual therapy; electrical stimulation; general conditioning (usual) exercises; any other nonsurgical interventions	Reviews on generalized and non-curve-specific exercises or other physiotherapeutic interventions administered to patients with AIS for other reasons, e.g., respiratory physiotherapy, spinal stabilization exercises, or electrical stimulation due to low back pain or leg pain; pre- or postoperative physiotherapeutic management of AIS patients; natural history or observation (“watchful waiting”) as a form of therapy; reviews on screening, diagnostics, prognosis, economic analysis, or other research questions other than nonsurgical interventions

(continued)

Table 18.4 (continued)

	Inclusion criteria	Exclusion criteria
Comparative interventions	Bracing, or scoliosis-specific exercises versus scoliosis-specific exercises plus other interventions, or different forms of these interventions (e.g., different modes of exercises or different types of braces); natural history or observation; other forms of nonsurgical interventions applied for scoliosis curve correction, e.g., chiropractic, manual therapy, electrical stimulation	Not specified
Outcomes	All short- and long-term outcomes that addressed the effectiveness and adverse effects of nonsurgical interventions; both patient-centered (e.g., pain, quality of life, depression, sense of stigmatization) and surrogate, secondary, or intermediate outcomes (e.g., curve progression, angle of trunk rotation, jaw deformity); the number of surgeries or the number needed to treat to avoid one surgery (need for surgery) as a criterion of failure of the nonsurgical interventions	Not specified

applicable.” The maximum score is 11. Scores 0–4, 5–8, and 9–11 indicate low-, moderate-, and high-quality reviews, respectively [58]. We conducted the appraisal independently. Exceptions were the Cochrane reviews [59, 60] that were included and coauthored by JB-S, when MP and a collaborator performed the independent appraisals. Assessments were conducted using guidelines for scoring AMSTAR questions [57, 58]. Disagreements were resolved by discussion.

Levels of Evidence

We assessed the level of evidence from each included review, considering the types of studies included, using the new Oxford Centre for Evidence Based Medicine (OCEBM) [61, 62], the JBI [63, 64] classifications, and, for the screening reviews, additionally, the improved National Health and Medical Research Council (NHMRC) hierarchy [65]. We decided to apply more than one classification because different classifications vary as regards their content and characteristics, and this allowed us to assess the included papers more comprehensively. The NHMRC document proposed the only hierarchy corresponding to the characteristics of screening reviews, while the OCEBM and JBI classifications are more suitable for intervention reviews.

18.2.4 Results

18.2.4.1 Description of Included Reviews/Quantity of Research Available

For the screening review, six articles met the criteria for inclusion within the analysis from a total of 224 papers (see Fig. 18.1 and Table 18.1): two quantitative systematic reviews, one of which included a meta-analysis and four systematic analyses of evidence which were part of or supplementing recommendation documents. For the non-surgical interventions, from a total of 469 titles or titles and abstracts of papers, 21 papers met the criteria for inclusion: 18 systematic reviews addressed the effectiveness of different interventions, 1 review evaluated usual physical activity, and 2 systematic reviews addressed the side effects in braced patients. Overall the reviews addressed numerous, patient-centered, and surrogate short- and long-term outcomes.

18.2.4.2 Methodological Quality of Included Reviews/Quality Assessment

Overall the quality of systematic reviews regarding screening ranged from the comparatively recent (2009) moderate-quality (AMSTAR score 6) (Fong et al. [66] and Sabirin et al. [67]) through to the outdated (2002) moderate-quality NHMRC review [68] to the poor-quality recent (2011) UK NSC [46] and the outdated 2004 USPSTF review [69], that is nonetheless still used for current and recent recommendations [52, 70]. The quality of the intervention reviews ranged from low methodological quality to high quality. Only two of the included reviews were of high quality [61, 62], while three were of moderate quality [72–74], and the remaining 16 reviews were found to be of low methodological quality [75–90] (Table 18.5).

18.2.4.3 Levels of Evidence of the Included Reviews

The six reviews relating to screening that we analyzed matched neither the improved NHMRC nor the new OCEBM levels of evidence hierarchy, with the exception of Fong et al. [66], which can be classified as a level 3 evidence in the OCEBM classification (Table 18.2). The levels of evidence from the reviews on interventions ranged from 1+ to 4, with some reviews not matching the OCEBM and the JBI hierarchies. The classification of the levels of evidence depended mainly on the type of included individual studies and also on the specific level of evidence hierarchy applied (Table 18.6).

18.2.5 Discussion

Detailed narrative characteristics of the content of the reviews that were included in this umbrella reviews are out with both the volume and the scope of this chapter. Therefore our report was limited to a short description of the quantity and quality of the evidence from the systematic reviews that were included and the characteristics

Table 18.5 AMSTAR ratings for reviews included in the quality analysis

Paper (year) [reference]	AMSTAR questions ^a											Total Yes	Overall quality ^b
	1	2	3	4	5	6	7	8	9	10	11		
<i>Screening:</i>													
NHMRC (2002) [68]	Y	CA	Y	Y	N	Y	Y	N	NA	N	N	5	Moderate
USPSTF (2004) [69]	Y	CA	N	N	N	Y	N	N	NA	N	N	2	Low
Negrini et al. (2005) [71]	Y	CA	N	Y	N	N	N	N	NA	N	N	2	Low
Sabirin et al., MaHTAS (2010) [67]	Y	Y	Y	Y	Y	Y	CA	N	NA	N	N	6	Moderate
Fong et al. (2010) [66]	Y	Y	Y	N	N	Y	N	N	Y	Y	N	6	Moderate
UK NSC (2011) [47]	Y	CA	Y	Y	N	Y	N	N	NA	N	N	4	Low
<i>Exercise treatments:</i>													
Negrini et al. (2003) [75]	N	CA	Y	N	N	Y	Y	Y	N	N	N	4	Low
Negrini et al. (2008) [74]	Y	CA	Y	N	N	Y	Y	Y	Y	N	N	6	Moderate
Fusco et al. (2011) [76]	Y	CA	N	N	N	Y	N	N	N	N	N	2	Low
Mordecai and Dabke (2012) [77]	N	CA	N	N	N	Y	N	N	N	N	N	1	Low
Romano et al. (2012) [60]	Y	Y	Y	Y	Y	Y	Y	Y	Y	N	N	9	High
<i>Manual therapy:</i>													
Romano and Negrini (2008) [78]	N	CA	Y	N	N	Y	N	N	N	N	N	2	Low
Gleberzon et al. (2012) [79]	N	CA	Y	N	N	Y	Y	N	NA	N	N	3	Low
McKennedy et al. (2013) [80]	N	Y	N	N	N	Y	Y	Y	N	N	N	4	Low
Posadzki et al. (2013) [72]	N	Y	Y	N	N	Y	Y	Y	Y	N	Y	7	Moderate
<i>Bracing:</i>													
Dolan and Weinstein (2007) [81]	N	N	Y	N	N	Y	N	N	N	N	N	2	Low
Negrini et al. (2010) [59]	Y	Y	Y	Y	Y	Y	Y	Y	Y	N	N	9	High

Table 18.5 (continued)

Paper (year) [reference]	AMSTAR questions ^a											Total Yes	Overall quality ^b
	1	2	3	4	5	6	7	8	9	10	11		
Maruyama et al. (2011) [82]	N	CA	N	N	N	Y	Y	Y	N	N	N	3	Low
Davies et al. (2011) [83]	N	CA	CA	N	N	Y	N	N	Y	N	N	2	Low
Sanders et al. (2012) [84]	N	CA	N	N	N	N	N	N	N	N	N	0	Low
<i>Different combinations of nonsurgical interventions:</i>													
Focarile et al. (1991) [85]	N	Y	N	N	N	Y	Y	Y	N	N	N	4	Low
Rowe et al. (1997) [86]	N	CA	N	N	Y	N	N	N	Y	N	N	2	Low
Lensinck et al. (2005) [73]	N	Y	N	N	N	Y	Y	Y	Y	N	N	5	Moderate
Weiss and Goodall (2008) [87]	N	N	Y	N	N	N	N	N	N	N	N	1	Low
<i>Usual physical activity:</i>													
Green et al. (2009) [88]	N	Y	Y	Y	N	Y	N	N	N	N	N	4	Low
<i>Adverse effects:</i>													
Li et al. (2008) [89]	N	CA	Y	N	N	Y	N	N	Y	N	N	3	Low
Saccucci et al. (2011) [90]	N	Y	Y	N	N	N	N	N	N	N	N	2	Low

Y yes, N no, CA cannot answer, NA not applicable, *NHMRC* National Health and Medical Research Council, Australia, *USPSTF* US Preventive Services Task Force, *MaHTAS* Health Technology Assessment Section, Ministry of Health Malaysia, *UK NSC* UK National Screening Committee

^aQuestions [55, 56]: “1. Was an a priori design provided?, 2. Was there duplicate study selection and data extraction?, 3. Was a comprehensive literature search performed?, 4. Was the status of publication (i.e. grey literature) used as an inclusion criterion?, 5. Was a list of studies (included and excluded) provided? 6. Were the characteristics of the included studies provided?, 7. Was the scientific quality of the included studies assessed and documented?, 8. Was the scientific quality of the included studies used appropriately in formulating conclusions?, 9. Were the methods used to combine the findings of studies appropriate?, 10. Was the likelihood of publication bias assessed?, 11. Were potential conflicts of interest included?”

^bReview quality scores [55, 56]: 0–4 low quality, 5–8 moderate quality, and 9–11 high quality

of the evidence available from the most recent and methodologically sound reviews. All relevant detailed information, comprising all the included papers as well as all the excluded papers, can be found in the original publications [40, 41].

18.2.5.1 Brief Summary of Evidence from Included Reviews

The evidence from included higher quality reviews is summarized in Table 18.2 according to the type of management (screening and nonsurgical interventions) and

Table 18.6 Evidence from higher quality, more recent systematic reviews on screening and non-surgical interventions in AIS

Title (year) [reference]	Findings/conclusions	Level of evidence [OCEBM/JBI]	AMSTAR score ^b /overall quality
Screening tests and programs:			
A meta-analysis of the clinical effectiveness of school scoliosis screening (2010) [66]	Only 17 % of the primary studies included within the meta-analysis of retrospective cohort studies found screening to be ineffective; the authors advocate for school screening, but recommended that the forward bend test should not be used alone within screening programs and that large, retrospective cohort studies are needed	3 ^a	6/moderate
Exercise treatments:			
Exercises for adolescent idiopathic scoliosis (2012) [60], Cochrane review	“Due to a lack of high quality RCTs in this area, there is no evidence for or against exercises, so hardly any recommendations can be given”; “no major risks of the intervention have been reported (...), and no side effects were cited in the considered studies”	1/1a	9/high
Exercises reduce the progression rate of adolescent idiopathic scoliosis: results of a comprehensive systematic review of the literature (2008) [74]	“Exercises can be recommended according to level-1b evidence with the aim of reducing scoliosis progression”; “it is impossible to state anything regarding the kind of exercises .. [or] ..kind of auto-correction to be performed”	3/1b	5/moderate
Manual therapy:			
Osteopathic manipulative treatment for pediatric conditions: a systematic review (2013) [72]	Findings from the AIS RCT: no evidence to support OMT as an effective treatment of mild AIS; the study assessed as high-quality RCT; “more robust RCTs are needed (...). Until such data are available, OMT cannot be regarded as effective therapy for paediatric conditions, and osteopaths should not claim otherwise”	1/1a	7/moderate
Bracing:			
Braces for idiopathic scoliosis in adolescents (2010) [59], Cochrane review	Very low quality of evidence in favor of bracing in terms of curve progression; low evidence in favor of hard bracing vs elastic bracing; serious side effects not documented in the included studies	1/1a	9/high3

Table 18.6 (continued)

Title (year) [reference]	Findings/conclusions	Level of evidence [OCEBM/JBI]	AMSTAR score ^b /overall quality
Different combinations of nonsurgical interventions:			
Effect of bracing and other conservative interventions in the treatment of idiopathic scoliosis in adolescents: a systematic review of clinical trials (2005) [73]	“Effectiveness of bracing and exercises is promising but not yet established”; limited evidence for the effectiveness of braces vs no treatment and vs electrical stimulation (ES); bracing, exercises, or ES as add-on treatment – additional effect cannot be justified; no difference for ES vs no treatment, bracing vs exercises, different types of bracing	1/1b	5/moderate

^aMatched the OCEBM classification only

^bDetails are in Table 18.5

in the order of descending levels of evidence. The table does not include any studies on “usual physical activity” and adverse events [88–90] nor some highly cited older systematic reviews [81, 86] as all those reviews were of low or very low quality.

18.2.5.2 Screening

The screening reviews were heterogeneous, both with regard to the research questions asked and the methodology used for their development. The reviews which supported the recommendation for school screening as well as those recommending against screening are based on different papers selected for inclusion and thus on different evidences or assumptions of the absence of evidence (Fig. 18.2). Conclusions were based on different criteria as follows: the set of criteria for appraising – feasibility, effectiveness, and appropriateness of screening programs, accuracy of screening tests, treatment effectiveness as a criterion justifying the need for screening, and cost-effectiveness. Three of the systematic reviews were found to be of moderate quality; Fong et al. ’s systematic review of retrospective cohort studies with a meta-analysis [66] as well as MaHTAS systematic review by Sabirin et al. (2010) [67] supported screening under certain conditions. The NHMRC document [68], which included a recommendation against screening, was also found to be of moderate quality.

18.2.5.3 Scoliosis-Specific Exercises (SSE)

The most recent of the available reviews was the rigorous Cochrane review [60]. This provided no convincing evidence from RCTs for or against these interventions in terms of curve progression as a primary outcome and no evidence of risks or side effects from performing scoliosis-specific exercises. A moderate-quality review by Negrini et al. [74] recommended the use of SSE exercises based on primary studies classified by the authors as level 1b evidence.

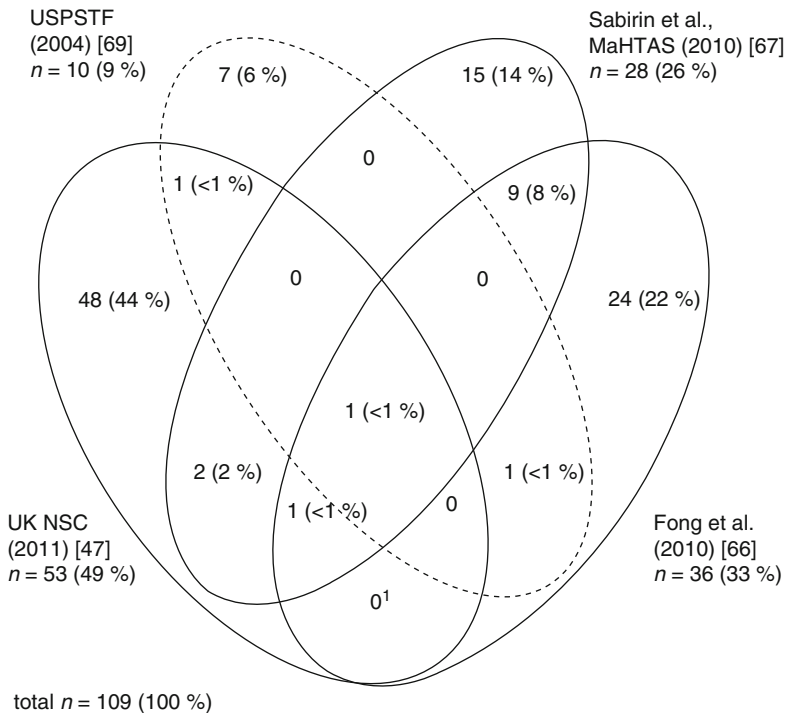


Fig. 18.2 Example Venn diagram showing overlaps across included systematic reviews: overlaps of papers included in four screening systematic reviews. *Numbers and percentages inside the ellipses* show the overlap of studies included in the four reviews. *Percentages outside the ellipses* illustrate the percentage of all 109 studies included in each of the four reviews. *n* number of papers included in the reviews, *USPSTF* the US Preventive Services Task Force, *MaHTAS* Health Technology Assessment Section, Ministry of Health Malaysia, *UK NSC* the UK National Screening Committee; this review also included the systematic review by Fong et al.

18.2.5.4 Manual Therapies

A recent good-quality systematic review of controlled studies [72] found one high-quality RCT showing no evidence to support osteopathic manual therapy as an effective treatment for mild AIS.

18.2.5.5 Bracing

A rigorous Cochrane review [59] found very low-quality evidence supporting the effectiveness of bracing in reducing curve progression and low-quality evidence favoring hard braces as compared to soft braces. In an earlier, moderate-quality systematic review of prospective controlled trials, Lennox et al. [73] concluded that due to the low-power, weak methodological quality, and clinical heterogeneity of the included studies, drawing firm conclusions was impossible. However the effectiveness of bracing and SSE treatments in reducing curve progression appeared to the authors to be promising.

18.2.5.6 Quality Analyses

The methodological quality of the majority (19 out of 27; see Table 18.5) of the systematic reviews that were retrieved was disappointingly low (Table 18.5), regardless of the limitations of the primary studies included in the reviews. Common errors included the following: no second independent reviewer and blind study selection and/or data extraction, no lists of included and excluded studies, no comprehensive search for evidence, and, perhaps most importantly, no quality assessment of included studies conducted. This is crucial as if the studies in question were of poor quality, then we should not be basing clinical practice or recommendations on the results of these reviews.

The screening reviews also differed significantly with regard to the databases selected and other resources searched. Moreover, significant heterogeneity was found within the reviews as follows: different research designs were considered (prospective trials and retrospective observational studies, systematic reviews, editorials), which were, except in the Fong et al. meta-analysis [66], analyzed separately.

In some of the reviews, the level of evidence hierarchy classification (categories of studies) was reported as a quality assessment. Further, a number of excluded reviews (listed in detail, with reasons for exclusion, as supplementary files to the resource publications) were called “systematic” but actually comprised only a structured and systematic literature search and then presented as a narrative discussion of a few papers of diverse designs. The only intervention systematic review with a meta-analysis by Rowe et al. [86] was seriously flawed methodologically (Table 18.6). Further and crucially the patient group was not homogenous and did not differentiate between juvenile and adolescent IS.

18.2.5.7 Limitations of the Study

Firm conclusions cannot be drawn from this umbrella review as it cannot clearly be established from the individual systematic reviews included that the interventions tested may differ significantly from each other within similar papers even if they have the same label (e.g., bracing – there are considerable differences in the construction, biomechanical principles of action, as well as the length of time worn [49]). The same applies to scoliosis-specific exercise treatment with at least six different schools of thought and approaches available [50].

Secondly, we were not able to firmly distinguish the methodological quality of the process for conducting systematic reviews from the quality of reporting of the reviews that we included and analyzed. The AMSTAR tool does not clearly distinguish between the two, and we did not utilize any measure of the reporting quality of the included reviews (such as PRISMA). It is also important to consider that this overview will need an update every few years to reflect recently published work. This could be undertaken by using another (or modified) appraisal tool rather than the AMSTAR tool (as the appraisal tool itself may influence the findings from an overview of systematic reviews [91]). The reliability and validity of the findings of umbrella reviews conducted with the use of different versions of this tool requires further studies [92].

18.2.6 Conclusions: What this Study Adds

18.2.6.1 Methodological Considerations

When many systematic reviews exist about a given topic, it is critical that the analysis of the methodological quality (i.e., the rigor for the development) of the systematic reviews and – consequently – their credibility are fully assessed as well. Systematic reviews should not only be considered as the base for an umbrella review that summarizes and synthesizes the findings from the currently available systematic reviews [29]. In this scenario an umbrella review's role is not only to summarize or synthesize but also to critically appraise, analyze, and assess the limitations of the systematic reviews that are currently available [13]. The role of an umbrella review is very important as poorly conducted systematic reviews may mislead and provide erroneous guidance for stakeholders – patients, therapists, insurance providers, policy makers, and researchers.

18.2.6.2 Conclusions of Case Study Issue

In this chapter we have explained why an umbrella review undertaking a best evidence synthesis approach was urgently needed for the school screening and nonsurgical treatment of adolescents with idiopathic scoliosis. We have described how we performed the two reviews and illustrated how umbrella reviews may be useful in informing end users in avoiding the misinterpretation of the available evidence from reviews of various quality and credibility.

18.2.6.3 Implications for Practice

A recent Cochrane Collaboration's analysis of the process of guideline development has revealed the underutilization of systematic reviews and meta-analyses in developing practice guidelines [93]. The results of our study can aid policy makers and guideline developers in producing better evidence-informed, up-to-date guidelines, both for the screening of schoolchildren in the risk groups of the development of AIS and for the nonsurgical treatment of adolescents with idiopathic scoliosis.

18.2.6.4 Conclusions and Implications for Research

In conclusion, good-quality primary studies of higher level designs are urgently needed in the areas of screening and conservative methods for the treatment of adolescents with idiopathic scoliosis. Further developments in the conduct of systematic reviews, especially – in the case of the subject of this chapter – using multiple types of studies in systematic reviews [94], will hopefully facilitate finding the right answers to the complex and diverse research questions currently found in research, practice, and policy, through conducting more valid and reliable umbrella reviews based on the greater trustworthiness currently found within systematic reviews.

18.3 Additional Details

We would like to thank Dr. Igor Cieřliński for his contribution to the AMSTAR assessment of the included Cochrane reviews. MP registered the protocol, conceived, and designed the experiments and prepared data extraction tables. MP and JB-S performed the experiments, analyzed the data, and contributed to the writing of the manuscript. The review followed the protocol with the exception of some details of database searching (additional specialty websites and guideline registries, instead of AMED, CINAHL, and EMBASE databases searches were conducted), as the “productivity scheme” of searching was preferred to the comprehensive search strategy [5, 57].

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