Dudney II and Sherburn
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REVIEW **Open Access**

Spinal cord injury without radiologic abnormality: an updated systematic review and investigation of concurrent concussion

William P. Dudney II[1*] and Eric W. Sherburn[2]

Abstract

Study design Systematic review.

Objectives The objectives were to systematically review the literature since the most recent systematic reviews for both adult and pediatric spinal cord injuries without radiologic abnormality (SCIWORA) in order to provide an update on the condition's epidemiology and characteristics and investigate the relationship between SCIWORA and concurrent concussion.

Methods A review was conducted according to the 2020 guidelines of the Preferred Reporting Items for Systematic Reviews and Meta-Analyses. The databases PubMed and OvidSP were searched on February 27, 2022. Inclusion criteria were individuals of any age, diagnosis of SCIWORA with or without abnormalities on MRI, and articles published from 2013 to 2014 (adults only) and 2014–present (all ages). Exclusion criteria were any spinal fracture or dislocation, studies that were narrative reviews, letters, book chapters, or editorials. Risk of bias was assessed using tools from the Clinical Advances Through Research and Information Translation Group at McMaster University and the Joanna Briggs Institute. Collected data were synthesized using Microsoft Excel.

Results Since the most recent systematic reviews, a total of 61 studies were identified, resulting in 2788 patients with SCIWORA. 69.55% of patients were pediatric, 30.45% adult. The most prevalent reported mechanism of injury was sports-related (39.56%) followed by fall-related (30.01%) and vehicle-related (27.23%). The vast majority of injuries occurred at the level of the cervical spine (82.59%). Of the 61 included studies, only 5 reported cases of concurrent concussion and/or TBI.

Discussion Since the most recent previous systematic reviews, there has been a 64% increase in reported cases of SCIWORA, likely as the result of advances in imaging technology and better awareness of SCIWORA. Still, SCIWORA remains a diagnosis most prevalent in children and young adults, the most common cause being sports-related injury. With the pathogenesis of SCIWORA sharing a predisposition to concussion, the significant lack of reporting of SCIWORA with concurrent concussion suggests that there have been missed diagnoses of either SCIWORA with concurrent concussion or vice versa, leading to longer recovery times, unrecognized and/or untreated underlying pathology, and possibly additional unnecessary morbidity. When the diagnosis of either SCIWORA or concussion is suspected, the other should additionally be considered in order to minimize the possible extended recovery time and related comorbidities. Limitations included the prevalence of lower quality studies such as case reports/series,

*Correspondence:
William P. Dudney II
willdudney@gmail.com
Full list of author information is available at the end of the article

insufficient reporting of study characteristics, and variability among neurologic impairment scales used and how SCIWORA was defined.

Keywords SCIWORA, Spinal cord injury, Concussion, Sports, Trauma

Introduction

Background

Pang and Wilberger first described SCIWORA in 1982 as a syndrome "reserved for those children with objective signs of myelopathy as a result of trauma, whose plain films of the spine, tomography, and occasionally myelography carried out at the time of admission showed no evidence of skeletal injury or subluxation" (Pang and Wilberger 1982). For years, it was regarded as a diagnosis exclusive to the pediatric population until Hirsh et al. described an adult case in 1993 (Hirsh et al. 1993). Since then, the overall reporting of SCIWORA in both children and adults has grown due to an increasing availability of diagnostic tools and awareness of the syndrome (Pang 2004; Boese and Lechler 2013). In the last 10 years, there have been three systematic reviews published on SCIWORA: one, Boese and Lechler's, in 2013 focusing on only adults (Boese and Lechler 2013) and two, Boese et al.'s and Carroll et al.'s, in 2015 focusing on only children (Carroll et al. 2015; Boese et al. 2015). These articles have been crucial in providing a more accurate incidence of SCIWORA, a deeper understanding of the syndrome, as well as an update on the definition of SCIWORA in response to the evolving imaging technology in recent decades. With advances in magnetic resonance imaging (MRI) especially, spinal cord lesions that were once not evident on X-ray or computed tomography (CT) are more often now being revealed on MRI, for example as various signal intensities suggesting cord edema, hematoma, contusion, and hemorrhage, among others. A number of articles have been published that discuss the implications of these MRI findings in regard to the definition of SCIWORA, some of which suggest a different diagnosis be made (Farrell et al. 2017; Mahajan et al. 2013; Dreizin et al. 2015; Trigylidas et al. 2010). Alternatively, the term 'real SCIWORA' has been employed when findings are negative across all imaging modalities, including MRI (Dreizin et al. 2015; Nagasawa et al. 2017; Asan 2018). For the sake of more precise diagnosis, we agree that the definition of SCIWORA going forward should maintain negative findings on all imaging results. However, for describing the foregoing diagnoses of SCIWORA with findings on MRI, the classification system created by Boese and Lechler (2013) can be useful, as it includes diagnoses of SCIWORA made throughout the evolving understanding of the condition and allows for more consistent categorization.

It is also pertinent to recognize that the most common mechanism of injury (MOI) for SCIWORA in the pediatric population is sports-related (Carroll et al. 2015; Knox 2016). According to Knox's nationwide database search in 2012, sports injuries account for 41% of pediatric SCIWORA cases (Knox 2016). The percentage of sports-related SCIWORA increases with age and by ages 11–17 years, sports-related injury accounts for 57% of cases (Knox 2016). Regardless of the specific cause of SCIWORA, Knox emphasizes the importance for clinicians to recognize that when SCIWORA occurs, it is manifested by high-energy trauma, frequently accompanied by concomitant non-spinal injuries (Knox 2016). With head trauma being the most common concomitant injury with SCIWORA in children, a high prevalence of SCIWORA with concurrent concussion is expected (Knox 2016). While there have been some published reports associating SCIWORA with concussion, we were surprised that there were not significantly more in the literature, suggesting that SCIWORA is likely underreported or underrecognized in the setting of concussion, particularly sports-related concussion (SRC). To our knowledge, there has not been a systematic review addressing the association between SCIWORA and concussion.

Main text

Rationale

There has been an abundance of studies regarding SCIWORA published since the most recent systematic reviews in 2013 and 2015. With the growing awareness and reporting of SCIWORA and the dynamic nature of its definition due to MRI, it would be beneficial to update the medical community on the syndrome's epidemiology, mechanism of injury, diagnosis, and implications. Importantly, one of the most prevalent mechanisms of injury for SCIWORA, especially in the pediatric population, is sports-related injury. SCIWORA, therefore, is a diagnosis that sports medicine physicians should be familiar with. As sports-related SCIWORA regularly presents similarly to sports-related concussion/traumatic brain injury (TBI), the two diagnoses often co-present. There have been some papers discussing the sports-related nature of SCIWORA, but few that emphasize the relationship between SCIWORA and concussion and the need to consider SCIWORA in the differential diagnosis for concussion.

Objectives

The objectives of this study were to systematically review the literature since the most recent systematic reviews for both adult and pediatric spinal cord injuries without radiologic abnormality (SCIWORA) in order to (1) provide an update on the condition's epidemiology and characteristics, and (2) investigate the relationship between SCIWORA and concurrent concussion.

Methods

Literature search

We conducted this review according to the 2020 guidelines of the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) (Page et al. 2020). On February 27, 2022, one reviewer (WD) performed a database search via PubMed and OvidSP. All available resources were selected in OvidSP and included: JBI EBP Database, AMED (Allied and Complementary Medicine) (1985 to February 2022), EBM Reviews—Database of Abstracts of Reviews of Effects (1st Quarter 2016), EBM Reviews—Cochrane Methodology register (3rd Quarter 2012), EBM Reviews—Health Technology Assessment (4th Quarter 2016), EBM Reviews—NHS Economic Evaluation Database (1st Quarter 2016), ERIC (1965 to January 2022), Health and Psychosocial Instruments (1985 to January 2022), International Pharmaceutical Abstracts (1970 to February 2022), APA PsycInfo (1806 to February Week 4 2022), Journals@Ovid Full Text, Your Journals@Ovid, Books@Ovid, Northern Light Life Sciences Conference Abstracts (2010 to 2022 Week 06), Embase Classic + Embase (1947 to 2022 February 27), Ovid Emcare (1995 to 2022 Week 7), Ovid MEDLINE(R) and Epub Ahead of Prine, In-Process, In-Data-Review and Other Non-Indexed Citations, Daily and Versions(R) (1946 to February 27, 2022). Because we intended this review to be an update since the most recent systematic reviews to present, and because the most recent 3 systematic reviews were published at different times within a 3-year span depending on their age specifications, we had separate search criteria associated with that timing. The first search criteria was "((sciwora OR sciworet OR sciwoctet OR spinal cord injur* without radio* abnormalit* OR spinal concussion) AND (adult))" and was limited to 2013–2014 [Boese and Lechler's (2013) search was performed in December 2012] in order to account for only the adult patients recorded between the publication of Boese and Lechler's (2013) systematic review of adult SCIWORA and the following two pediatric studies (Carroll et al. 2015; Boese et al. 2015). The second search criteria was "(sciwora OR sciworet OR sciwoctet OR spinal cord injur* without radio* abnormalit* OR spinal concussion)" and was limited to 2014–present, accounting for all cases, adult and pediatric, since the most recent systematic reviews. The same search criteria were used for both PubMed and OvidSP. Inclusion criteria was (1) individuals of any age, (2) diagnosis of SCIWORA with or without abnormalities on MRI, and (3) articles published from 2013–2014 (adults only) and 2014-present (all ages), as previously mentioned. Exclusion criteria was (1) any spinal fracture or dislocation, (2) studies that were narrative reviews, letters, book chapters, or editorials, and (3) articles not otherwise meeting inclusion criteria. One reviewer (WD) carried out the selection process, as outlined in the flow diagram (Fig. 1, see below in Results). Citations from both databases were downloaded and then, uploaded and managed in EndNote (EndNote 20.2 for macOS, Clarivate, Philadelphia, PA, USA). Duplicates were removed from the initial search results using the EndNote 'Find Duplicates' function. The remaining studies' abstracts and/or titles were screened for relevance to our review. The full texts of the relevant studies were then accessed and assessed according to our inclusion and exclusion criteria. The studies that met our criteria were finally assessed for their inclusion of concussion/TBI data.

Data collection and synthesis

One reviewer (WD) collected data from the included studies and managed them in Microsoft Excel for Mac version 16.16.5 (Microsoft Corporation, Redmond, WA, USA). Outcome data were manually transferred from the studies into the Excel spreadsheet. The collected data from the studies included author, publication year, PubMed reference number (PMID) or accession number, study design, number of SCIWORA cases, sex, mean age, mechanism of injury, spinal level, American Spinal Injury Association Impairment Scale (AIS) grade (initial/only and final), time until first and last MRI, initial and final MRI results, SCIWORA classification, and presence or absence of reported concurrent concussion/TBI. SCIWORA classification was based on the MRI imaging patterns established by Boese and Lechler, where Type I stands for no detectable abnormalities on MRI, IIa stands for extraneural abnormalities only, IIb stands for intraneural abnormalities only, and IIc stands for both extraneural and intraneural abnormalities (Boese and Lechler 2013). According to the same classification scheme, intraneural abnormalities include "edema, hemorrhage, contusion, and partial or complete transection" of the spinal cord, while extraneural abnormalities include "disc protrusion or herniation, flavum bulging, spondylosis, ossification of posterior longitudinal ligament, prevertebral soft tissue swelling, or ligamentous abnormalities" (Boese and Lechler 2013). The majority of the outcomes were directly transferable from their original studies to our spreadsheet of collective data. For mean age, the

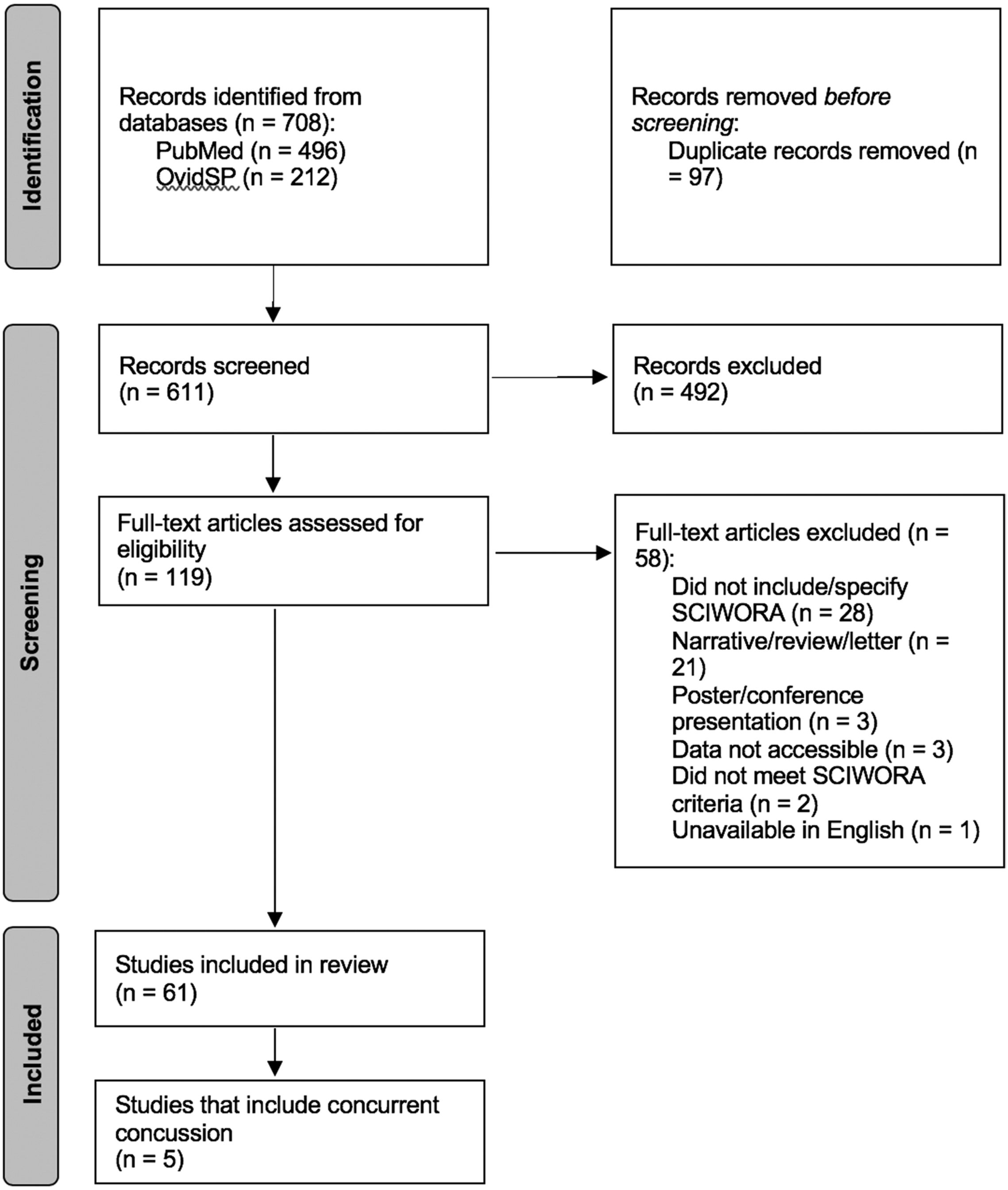

Fig. 1 PRISMA 2020 flow diagram (SCIWORA: spinal cord injury without radiologicabnormality)

values were either already calculated or were unavailable. We did not attempt to extrapolate mean age across different studies due to the duplicate data in some instances and difference in study designs. Spinal level and AIS grade were only collected as they were presented in the studies; we did not categorize those values ourselves. For SCIWORA classification, we either collected values that were already categorized in their studies, or categorized them ourselves only if the MRI results were sufficiently descriptive and included all findings from each patient in the corresponding studies. For further data synthesis, the studies, organized in a collective Excel spreadsheet, were selected for their inclusion of certain outcomes, such as spinal level or AIS grades, after which we used the Excel charting function to present the data visually. As this review is not a meta-analysis, we did not perform statistical analysis on our data.

Assessment of risk of bias, reporting bias, certainty, and strength of recommendation

One reviewer (WD) assessed each studies' risk of bias. All 61 included studies were first categorized according to their study design. 29 of those studies did not explicitly state their study design, so the reviewer categorized them using a tool from the Centre for Evidence-Based Medicine (CEBM) at Oxford University (https://www.cebm.ox.ac.uk/resources/ebm-tools/study-designs). For cohort studies, we used the Tool to Assess Risk of Bias in Cohort Studies from the Clinical Advances Through Research and Information Translation (CLARITY) Group at McMaster University (https://www.evidencepartners.com/resources/methodological-resources/tool-to-assess-risk-of-bias-in-cohort-studies-distillersr). For case reports, we used the Joanna Briggs Institute (JBI) Critical Appraisal tools for use in JBI Systematic Reviews Checklist for Case Reports (https://jbi.global/critical-appraisal-tools). For case series, we used the JBI Checklist for Case Series (https://jbi.global/critical-appraisal-tools). For case–control studies, we used the Tool to Assess Risk of Bias in Case–Control Studies from the CLARITY Group (https://www.evidencepartners.com/resources/methodological-resources/tool-to-assess-risk-of-bias-in-case-control-studies-distillersr). For cross-sectional studies, we used the JBI Checklist for Analytical Cross Sectional Studies (https://jbi.global/critical-appraisal-tools). Lastly, for the 3 systematic reviews, we used the JBI Checklist for Systematic Reviews (https://jbi.global/critical-appraisal-tools). Because the majority of the included studies were non-interventional, many assessment tools, such as those developed by Cochrane, were non-applicable. Therefore, we chose to use the risk of bias assessment tool developed by the CLARITY Group at McMaster University, which drew from the Downs and Black instrument and the Newcastle–Ottawa Scale. However, tools from the CLARITY Group for case reports, case series, cross-sectional studies, and systematic reviews either did not exist or were not applicable to the non-interventional nature of our reviews' studies, so the JBI tools were used instead.

We did not use a particular tool to assess reporting bias, but rather referred to Cochrane's page on reporting biases to elucidate possible reporting biases from our review (https://methods.cochrane.org/bias/reporting-biases).

We did not assess certainty as it pertains to our study, since the tools (e.g., GRADE) used to do so require data from interventional studies or meta-analyses, such as power, confidence intervals, and heterogeneity (I^2).

We assessed the strength of recommendation for our study. We utilized the American Academy of Family Physicians Strength of Recommendation Taxonomy (AAFP SORT) tool and its algorithm to determine the "Strength of Recommendation Based on a Body of Evidence" (Ebell et al. 2004).

Results

Literature search

The results of our literature search, screening, and article assessment can be seen in the flow diagram (Fig. 1). Searches from the databases PubMed and OvidSP yielded 708 total initial results. 97 duplicate records were identified via EndNote, leaving 611 unique records. Those records were screened for relevance, and 492 were found to be not relevant to our review and were excluded. The remaining 119 articles were accessed in their full-text format and assessed for eligibility and inclusion/exclusion criteria. 58 full-text articles were excluded for reasons outlined in the flow diagram (Fig. 1) and Table S1 (see Additional file 1). Many studies that were excluded pertained to spinal cord injuries in general, for example, but did not specify SCIWORA as a distinct injury, and so were excluded. In total, 61 studies met our inclusion criteria for review (Boese and Lechler 2013; Carroll et al. 2015; Boese et al. 2013, 2015, 2016; Nagasawa et al. 2017; Asan 2018; Knox 2016; Alas et al. 1976; Ellis and McDonald 2015; Ellis et al. 2019; Acer et al. 2018; Araki et al. 2015; Babcock et al. 2018; Bansal and Chandanwale 2016; Bansal et al. 2020; Bazán et al. 2013; Bonfanti et al. 2019; Brauge et al. 2020; Butts et al. 2021; Canosa-Hermida et al. 2019; Cheng et al. 2013; Compagnon et al. 2020; Dubey et al. 2018; Fiaschi et al. 2016; Freigang 2021; Huang et al. 2013; Iaconis Campbell et al. 2018; Inoue et al. 2017; Jung et al. 2014; Kanezaki et al. 2014; Khan et al. 2017; Khatri et al. 2014; Kim et al. 2016, 2021; Liang et al. 2019; Liu et al. 2015; Machino et al. 2019; Makino et al. 2014; Martinez-Perez et al. 2017; Mohanty et al. 2013; Na and Seo 1106; Nakamoto et al. 2013; Ouchida et al. 1976; Park et al. 2015; Piatt 2015; Qi et al. 2020; Ren et al. 2017; Ribeiro da Silva et al. 2016; Sakti et al. 2018; Sanghvi et al. 2013; Sun et al. 2014; Ullah et al. 2020; Urdaneta et al. 2013; Verzelli et al. 2013; Wang et al. 2016, 2019; Yaqoob Hakim et al. 2021; Zhang and Xia 2015; Zhu et al. 2019; Zou et al. 1976). Out of those 61 studies, only 5 included data regarding concurrent concussion/TBI with SCIWORA (Knox 2016; Alas et al. 1976; Ellis and McDonald 2015; Ellis et al. 2019; Boese et al. 2013).

Data from most recent systematic reviews (Boese and Lechler 2013; Carroll et al. 2015; Boese et al. 2015)

The data presented in Table 1 represent the summative data from the most recent systematic reviews (Boese

Table 1 Study characteristics of previous systematic reviews

Reference #	Author	Publication year	Study design/population	Cases of SCIWORA	Sex			Mean age (years)
				N	Male	Female	NA	
Boese and Lechler (2013)	Boese and Lechler	2013	Systematic review/adult	w/o MRI: 261; w/ MRI: 1132	w/o MRI: 125; w/ MRI: 845	w/o MRI: 13; w/ MRI: 187	w/o MRI: 123; w/ MRI: 100	NA
Carroll et al. (2015)	Carroll et al	2015	Systematic review/pediatric	433	252	116	65	10.03
Boese et al. (2015)	Boese et al	2015	Systematic review/pediatric	114	76	37	1	7.77

SCIWORA spinal cord injury without radiologic abnormality, *MRI* magnetic resonance imaging

and Lechler 2013; Carroll et al. 2015; Boese et al. 2015), adult and pediatric. By including these studies, we intended to keep track of historical SCIWORA reporting in an effort to be as comprehensive as possible in amalgamating SCIWORA characteristics, as well as to provide a reference for scope in relation to the subsequent studies that were published. It is important to note that there are overlapping of studies between the two 2015 pediatric systematic reviews (Carroll et al. 2015; Boese et al. 2015). Those articles, which create some duplicate data, include the following:

1. Bondurant and Oro 1993, $N=1$, male (1) (Bondurant and Oró 1993)
2. Bosch et al. 2002, $N=189$, male (125), female (64) (Bosch et al. 1976)
3. Buldini et al. 2006, $N=2$, male (1), female (1) (Buldini et al. 2006)
4. Dickman et al. 1991, $N=26$, male (19), female (7) (Dickman et al. 1991)
5. Duprez et al. 1998, $N=1$, male (1) (Duprez et al. 1998)
6. Ergun 2003, $N=1$, female (1) (Ergun and Oder 2003)
7. Feldman 2008, $N=3$, female (2), NA (1) (Feldman et al. 2008)
8. Grubenhoff and Brent 2008, $N=1$, male (1) (Grubenhoff and Brent 2008)
9. Kim et al. 2008, $N=1$, female (1) (Kim et al. 1976)
10. Lee et al. 2006, $N=1$, male (1) (Lee et al. 2006)
11. Liao et al. 2005, $N=9$, male (6), female (3) (Liao et al. 2005)
12. Matsumara et al. 1990, $N=1$, female (1) (Matsumura et al. 1990)
13. Mortazavi et al. 2011, $N=1$, male (1) (Mortazavi et al. 2011)
14. Pollina and Li 1999, $N=1$, male (1) (Pollina and Li 1999)
15. Shen et al. 2007, $N=1$, female (1) (Shen et al. 2007)
16. Trigylidas et al. 2010, $N=3$, male (2), female (1) (Trigylidas et al. 2010)
17. Trumble and Myslinski 2000, $N=1$, female (1) (Trumble and Myslinski 2000)
18. Yamaguchi et al. 2002, $N=1$, female (1) (Yamaguchi et al. 2002)

Table 2 MOI from previous systematic reviews

Pediatric MOI	*N*
Sports	180
MVC/RTA	129
Fall	106
Other	40
Abuse	6
Birth trauma	2
Adult MOI	***N***
"Trauma-mechanism"	1393

MOI mechanism of injury, *MVC* motor-vehicle collision, *RTA* road traffic accident

Thus, the total overlap between the two systematic reviews yielded duplicates of $N=244$, male ($N=159$), female ($N=84$), and NA ($N=1$). After adjusting for these duplicates, the recorded cases of SCIWORA up to these systematic reviews was a total of $N=1696$, male ($N=1139$), female ($N=269$), and NA ($N=288$). Besides the number of cases, we were unable to separate the duplicate data for other outcomes that the studies calculated. Consequently, some of the following data (Tables 1, 2, Figs. 2, 3, 4, 5) might be partially inflated. Data for mean age were not available for the adult systematic review.

The mechanism of injury (MOI, Table 2) for the adult systematic review was described as all cases being related to a "trauma-mechanism" (Boese and Lechler 2013). For the pediatric cases, the most common MOI was sports-related ($N=180$), followed by motor-vehicle collision (MVC)/road traffic accident (RTA) ($N=129$) and falls ($n=106$).

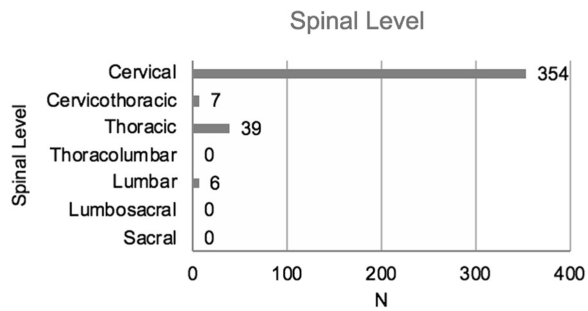

Fig. 2 Spinal levels from previous systematic reviews

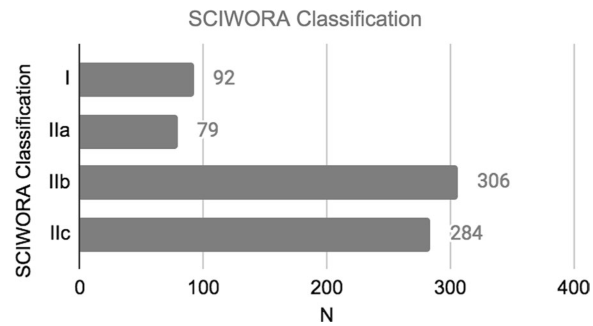

Fig. 5 SCIWORA classification from previous systematic reviews

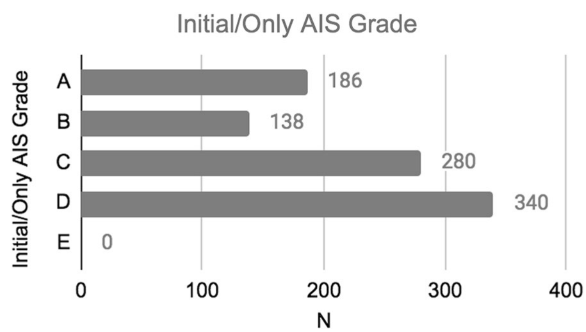

Fig. 3 Initial/only AIS grades from previous systematic reviews (*AIS* American Spinal InjuryAssociation Impairment Scale)

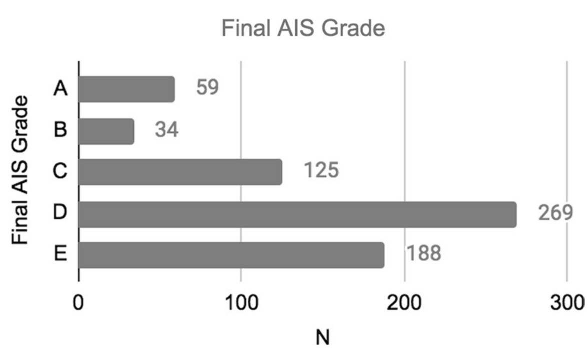

Fig. 4 Final AIS grades from previous systematic reviews (*AIS* American Spinal InjuryAssociation Impairment Scale)

Only Carroll et al. (2015) reported spinal level (Fig. 2), with cervical being the most predominant, followed by thoracic, then cervicothoracic, and lastly lumbar.

Carroll et al. (2015) only reported one set of values for AIS grades. With that said, the most predominant initial AIS grade was D, followed by C (Fig. 3), and the most predominant final AIS grade was D, followed by E (Fig. 4).

SCIWORA classification (Fig. 5) was limited by the number of cases with MRI results. From the cases with MRI results, though, the most predominant findings were IIb (intraneural only) followed by IIc (intraneural and extraneural).

None of these systematic reviews discussed concurrent concussion/TBI.

Data since previous systematic reviews

The overall number of cases of SCIWORA since the previous systematic reviews is 2788, 922 of those being male and 519 being female; 1347 did not specify sex (Additional file 1: Table S2). All studies except for Urdaneta et al. (2013) reported mean age.

With mechanism of injury (Table 3) collated into 4 categories (sports-related, fall-related, vehicle-related, and other), the most predominant MOI overall is sports-related ($N=555$), followed by fall-related ($N=421$), then vehicle-related ($N=382$), and finally, other ($N=45$). The majority of the sports-related injuries were not specified by each sport, and some were specified only as distinctly as "team" or "individual" sports, for example (Alas et al. 1976; Babcock et al. 2018). The leading specified sports-related MOI, though, was martial arts/judo ($N=62$), followed by American football ($N=50$). A common fall ($N=315$) was the most predominant fall-related injury, and all but one vehicle-related injury was an MVC or RTA. The other MOIs were mainly assault/violence ($N=26$) or rough play ($N=14$).

The most predominant spinal level (Fig. 6) by far was cervical ($N=1290$), with the next level, thoracic, having over a thousand fewer cases ($N=235$). At least one injury to each level was reported.

The highest number of cases of initial/only AIS grade reported (Fig. 7) was D ($N=235$), followed by C ($N=174$). A number of studies only reported one AIS grade, leading to fewer overall reported final AIS grades.

Table 3 MOI from studies since previous systematic reviews

MOI	*N*	Total
Sports-related		555
Sports, not otherwise specified	304	
"Team sports," not otherwise specified	62	
Martial arts/judo	62	
American football	50	
Gymnastics/cheer/dance/"back bend"	19	
"Individual sports," not otherwise specified	12	
"Winter sports," not otherwise specified	10	
Hockey	8	
Rugby	7	
"Water sports," not otherwise specified	5	
Biking	4	
Wrestling	4	
Diving	3	
Climbing	1	
Horseback riding/equestrian	1	
Soccer	1	
Standing high jump	1	
Surfing	1	
Fall-related		421
Fall	315	
Fall from height (FFH)	79	
"Falling objects", "stuck by falling objects", "heavy object smashing injury", "fall of heavy object", "Heavy damage to the neck"	22	
Fall down stairs (FDS)	5	
Vehicle-related		382
RTA/MVC	381	
Motorcycle accident	1	
Other		45
Assault/violence	26	
"Rough play", "playing",	14	
"Traction injury"	1	
"Active neck stretching"	1	
Passive rotation	1	
Passive flexion–extension	1	
"Work accident"	1	

MOI mechanism of injury, *RTA* road traffic accident, *MVC* motor-vehicle collision

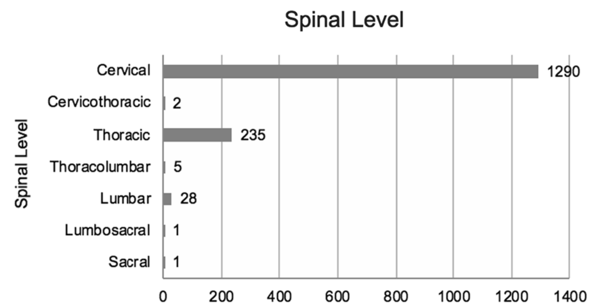

Fig. 6 Spinal levels from studies since previous systematic reviews

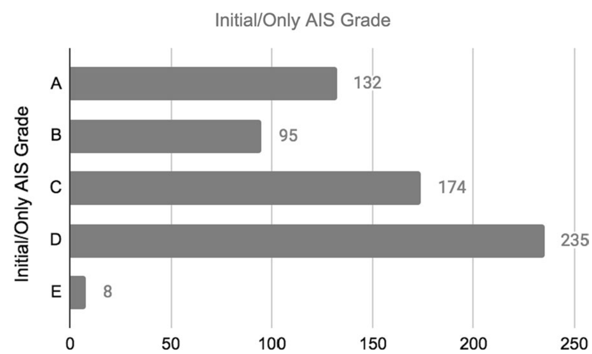

Fig. 7 Initial/only AIS grades from studies since previous systematic reviews (*AIS* AmericanSpinal Injury Association Impairment Scale)

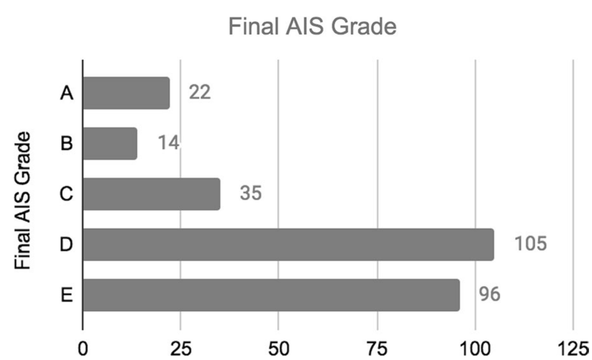

Fig. 8 Final AIS grades from studies since previous systematic reviews (*AIS* AmericanSpinal Injury Association Impairment Scale)

Of those reported final grades (Fig. 8), though, the highest amount remained D ($N=105$), followed by E ($N=96$).

Some studies opted to employ the Frankel grade system instead of the AIS (Brauge et al. 2020; Freigang 2021; Huang et al. 2013; Ribeiro da Silva et al. 2016). Of this data, from the initial/only reported grades (Fig. 9), the leading score was C ($N=21$) followed by D ($N=13$), while the leading score for the final reported grades (Fig. 10) was majoritively E ($N=45$).

A few studies opted to employ the Japanese Orthopaedic Association (JOA) score instead of the AIS (Inoue et al. 2017; Wang et al. 2019; Zhang and Xia 2015). Of this data, the mean initial JOA score was 8.24, and the mean final JOA score was 12.52.

The most predominant SCIWORA classification (Fig. 11) was IIc (extraneural and intraneural, $N=83$). There was not a great difference between the other types, with 66 cases of I (no detectable abnormalities), 59 cases of IIa (extraneural only), and 61 cases of IIb (intraneural only).

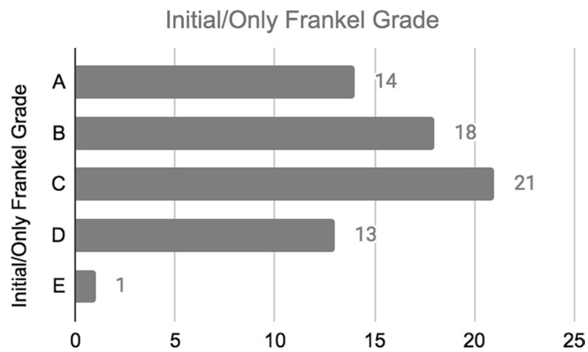

Fig. 9 Initial/only Frankel grades from studies since previous systematic reviews

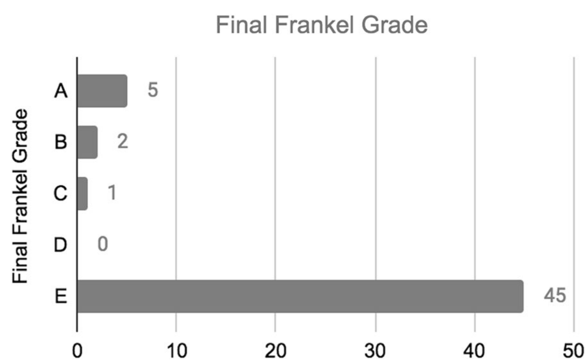

Fig. 10 Final Frankel grades from studies since previous systematic reviews

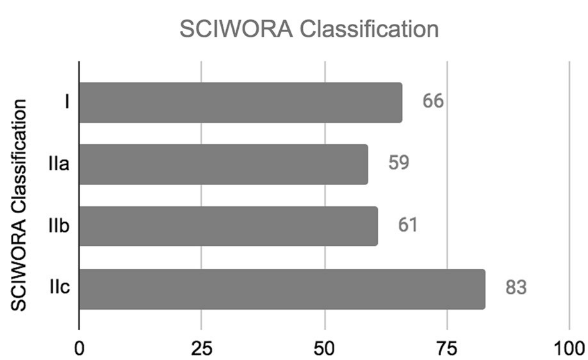

Fig. 11 SCIWORA classification from studies since previous systematic reviews

Studies reporting concurrent concussion

Of the 61 studies included in our review, only 5 studies (Knox 2016; Alas et al. 1976; Ellis and McDonald 2015; Ellis et al. 2019; Boese et al. 2013) reported cases of concurrent concussion and/or TBI. Two of those articles, Boese et al. (2013) and Knox (2016), categorize their findings simply as "brain injury" or "brain/head trauma," respectively. These findings are recorded as concomitant with the patients' SCIWORA. However, the authors do not further investigate or offer elaborate interpretation of these findings. Knox does state that "head trauma was the most common associated injury in all age groups, [and] the highest rate was found in the youngest age groups" (Knox 2016). On their paper exploring epidemiological trends in sports-related cervical spine and spinal cord injury, Alas et al. found, after running a linear regression, that "concurrent TBI was found to co-present with cervical SCIWORA at a significantly higher rate than other mechanisms of sports injury [and that] SCIWORA was a significant predictor for concurrent TBI across all sports" (Alas et al. 1976). In Ellis et al.'s "Cervical Spine Dysfunction Following Pediatric Sports-Related Head Trauma," the authors detail how patients with cervical spine dysfunction (CSD), which includes SCIWORA, with concurrent SRC were "significantly more likely to be female, which may be attributable to gender-related differences in cervical spine musculature [and that] patients with SRC with CSD were also found to exhibit higher PCSS (Post-Concussion Symptom Scale) scores at initial assessment and were more likely to demonstrate subjective and objective evidence of coexisting vestibulo-ocular dysfunction compared with those without CSD" (Ellis et al. 2019). Finally, Ellis and McDonald (2015) authored the first case report that we have found directly discussing SCIWORA with concurrent concussion. In their case, the patient experienced cervical SCIWORA with concurrent sports-related concussion, specifically hockey-related. The authors note that there is "wide variability, subtlety, and delayed onset of the clinical manifestations associated with both SRC and SCIWORA among children and adolescents [that] present significant obstacles to recognizing and managing these conditions, especially when they occur in the same patient" (Ellis and McDonald 2015). They also discuss diagnosis, management, and treatment strategies for both injuries when they coexist.

Risk of bias assessment

(See Additional file 1: Table S3: Assessment of risk of bias.) About half of the included studies were cohort studies, some of which showed lower risk of bias, including Knox (2016) and Sun et al. (2014) Some cohort studies that showed higher risk of bias include Makino et al. (2014), Urdaneta et al. (2013), and Machino et al. (2019). Among the case reports, a few studies (Acer et al. 2018; Butts et al. 2021; Liang et al. 2019) showed minimal risk of bias, while one study (Araki et al. 2015) showed particularly higher risk of bias. For the case series, while no studies appeared to have higher risk of bias, two studies showed particularly lower risk of bias (Kim et al. 2016; Yaqoob Hakim et al. 2021). Babcock et al. (2018) and

Compagnon et al. (2020) were the only studies in their categories—case–control studies and cross-sectional studies, respectively—and both showed lower risk of bias. Lastly, the 3 systematic reviews all showed lower risk of bias in their assessment (Boese and Lechler 2013; Carroll et al. 2015; Boese et al. 2015). Overall, although there were some studies that showed higher risk of bias than others, we did not deem any of the studies to have risk of bias significant enough to exclude them from our review.

Reporting bias

There are several areas where reporting biases might have factored into the collection and synthesis of our study's results. First, there may have been publication bias, as data on SCIWORA may not be a priority for publication for some journals due to its relative obscurity or unfamiliarity. Underdiagnosis and underreporting of SCIWORA in practice probably plays a role in its lack of publication as well. There may have been language bias in our review as some articles were not accessible in the USA or were unavailable in English. Time lag bias may have played a role as studies conducted but not published before we performed our literature search were not included. There may have been some location bias, as studies only found in databases that were not queried would not have been included in our search results. To that end, we attempted to minimize this risk by using databases relevant to and widely inclusive of the topic of SCIWORA while also remaining consistent with the databases used in the previous systematic reviews (Boese and Lechler 2013; Carroll et al. 2015; Boese et al. 2015). Lastly, outcome reporting bias may have occurred as some studies, especially those concerning general spinal cord injuries, might have reported SCIWORA unintentionally or unspecifically, thus not appearing in our literature search results.

Strength of recommendation

Using the algorithm for determining the Strength of Recommendation Based on a Body of Evidence, we concluded that the strength of recommendation for our study is A, which the AAFP SORT tool describes as a "recommendation based on consistent and good-quality patient-oriented evidence" (Ebell et al. 2004). This applies to the recommendation regarding considering the diagnosis of concurrent concussion when diagnosing SCIWORA, and vice versa, which we comment on in the Discussion section below.

Discussion

Since the previous systematic reviews, one regarding adult SCIWORA in 2013 (Boese and Lechler 2013) and two regarding pediatric SCIWORA in 2015 (Carroll et al. 2015; Boese et al. 2015), there has been what seems to be an increase in recognition and reporting of the condition. The previous 3 systematic reviews include data since the inception of the diagnosis of SCIWORA in 1982 (Pang and Wilberger 1982), so with our review picking up where they left off, this compilation of SCIWORA epidemiology should be more or less comprehensive. With that being said, there were 1696 total reported cases of SCIWORA among the previous 3 systematic reviews, spanning from 1982 to 2013/2015. Since then, there have been 2788 additional reported cases of SCIWORA from 2013/2015 to present, a 64% increase. Expanding on this sizable increase in reported cases, it is important to note that this occurred in the span of 9 years compared to the previous span of over 30 years. This difference is most likely due to a number of factors. Advances in imaging technology, particularly MRI, have provided physicians with much better imaging quality and techniques for analyzing soft tissue abnormalities that were previously undetected or unknown using earlier radiologic technology like CT or plain films (Boese and Lechler 2013; Boese et al., 2013, 2015, 2016; Farrell et al. 2017; Liu et al. 2015; Machino et al. 2019; Ouchida et al. 1976; Sun et al. 2014). Not only have there been advances in the technology of radiologic studies, there has also been an increase in the imaging's availability and widespread, routine use. When patients suspected to have spinal cord injuries arrive at trauma or emergency centers, whereas it used to be common practice to only obtain plain films or CT, it is becoming increasingly common to expeditiously obtain at least CT and subsequently MRI, especially when plain films and CT are negative, but the patient is still exhibiting signs of neurologic dysfunction (Boese and Lechler 2013; Liu et al. 2015). Of course, the integration of this imaging protocol would not be possible without consummate collaboration between the initial providers (emergency medicine, clinicians, emergency medical services/paramedics, athletic trainers, etc.) and specialists (neurosurgeons, trauma surgeons, radiologists, neurologists, etc.). The better awareness of SCIWORA, even as a part of the differential diagnosis for spinal cord injury and neurologic dysfunction, that is emerging in the medical community will go hand-in-hand with the improvements in collaboration and protocol development we are seeing.

Although we could not calculate mean age across all studies in our review, it is important to note that there were dramatically more pediatric cases reported than adult cases. In fact, there were 1907 pediatric cases compared to 835 adult cases [46 cases being without reported age (Urdaneta et al. 2013)]. While it is possible that SCIWORA among the adult population is disproportionately underreported relative to the pediatric population, the greater number of pediatric cases suggests that SCIWORA remains a diagnosis more common in patients of relatively younger age. There are several proposed reasons why this is the case, which numerous studies have described in more detail (Pang and Wilberger 1982; Pang 2004; Carroll et al. 2015; Ellis and McDonald 2015). In summary, though, children have high elasticity of the spinal ligaments, causing greater deformation of the spinal cord relative to the vertebral column, without damaging the vertebrae. Children also have less developed neck musculature and a greater ratio of head size-to-body size, manifesting in unsupported head movements, namely flexion and extension. The uncinate processes, facets, and vertebral bodies are also relatively underdeveloped, leading to hypermobility. Fragile vertebral bodies and higher water content in the annulus and intervertebral disk also predispose the discs to greater expandability forces and risk for injury. Since the spine does not reach unified stability comparable to the adult skeleton until at least 8 years of age, those younger than 8 years are at much higher risk of injury.

The higher ratio of cases in younger individuals is likely related to the fact that the most predominant mechanism of injury was sports-related. Sports-related injury is the most common MOI among the pediatric population, findings consistent with our study as well as in Carroll et al.'s (2015). Therefore, with a higher proportion of pediatric cases in our study there was bound to be a significant portion of sports-related injury. This could also be due to the fact that several studies were focused on sports-related injury specifically (Alas et al. 1976; Ellis and McDonald 2015; Ellis et al. 2019; Araki et al. 2015; Babcock et al. 2018; Ren et al. 2017). Furthermore, there were not any studies, more than case reports at least, focusing specifically on fall-related or vehicle-related injuries, leading to possibly fewer reported cases than would be evident if there were such studies.

The cervical spine was the most predominant affected spine level by a considerable margin. There are a few probable reasons why. First, similar to there being more studies focused on sports-related injury, there were more studies focused on cervical spine injury than any other spinal level. In the study titles alone, there were 17 studies (Alas et al. 1976; Ellis and McDonald 2015; Ellis et al. 2019; Babcock et al. 2018; Cheng et al. 2013; Fiaschi et al. 2016; Huang et al. 2013; Kim et al. 2021; Machino et al. 2019; Martinez-Perez et al. 2017; Mohanty et al. 2013; Ouchida et al. 1976; Ribeiro da Silva et al. 2016; Sun et al. 2014; Urdaneta et al. 2013; Wang et al. 2019; Zhang and Xia 2015) focused on the cervical spine and 5 studies (Dubey et al. 2018; Iaconis Campbell et al. 2018; Khatri et al. 2014; Ren et al. 2017; Sanghvi et al. 2013) focused on the thoracic spine, the next most prominent spine level. Additionally, the fact remains that the cervical spine is the most susceptible to the mechanical forces that cause SCIWORA, such as hyperextension/flexion, axial loading, and distraction. In addition to the mechanisms detailed above, the cervical spine is disproportionately at risk for injury as the combination of the underdeveloped vertebrae, ligaments, and musculature and the greater head to body size ratio "introduces a high fulcrum of motion at the cranium and increased risk for cervical injury at the atlanto-occipital and atlanto-axial joints" (Alas et al. 1976). Lastly, there exists the congruence of sports-related and cervical spine injury being the most predominant of their respective categories, as the vulnerabilities that lead to both—i.e., younger age with less mature anatomy and the mechanical forces at work (hyperextension/flexion, axial loading, distraction, etc.)—coexist and exacerbate each other.

It seems that the medical community has been slower to adopt the SCIWORA classification system created and proposed by Boese and Lechler. Of the 2788 cases of SCIWORA since the previous systematic reviews, the earliest of which initially proposed the classification system (Boese and Lechler 2013), only 269 total cases were defined with a SCIWORA classification. As imaging (MRI) technology and techniques continue to advance, there will probably be more diagnostic detail that is revealed, adding to the complexities and nuances of spinal cord injury diagnoses. By implementing this classification system, rather than having to modify the term SCIWORA altogether, it allows for straightforward, unambiguous categorization of the type of spinal cord injury within the realm of traditional SCIWORA. It also allows for consistency and comparability between diagnoses from various reporters. Whereas the term 'real SCIWORA' has gained traction when describing SCIWORA with no evidence of intra- or extraneural abnormalities on MRI (Dreizin et al. 2015; Nagasawa et al. 2017; Freigang 2021; Liang et al. 2019), in Boese and Lechler's classification system, Type I would replace that terminology. We advocate for this classification's continued use and integration.

With our review being as comprehensive as possible for the epidemiology of SCIWORA since its earliest recognition, it should also be just as comprehensive for the reporting of SCIWORA with concurrent concussion. We were surprised, then, that only 5 studies actually did report concurrent concussion, head/brain injury, and/or TBI (Knox 2016; Alas et al. 1976; Ellis and McDonald 2015; Ellis et al. 2019; Boese et al. 2013). This should be cause for concern within the medical community, with the implications being that there have been missed diagnoses of either SCIWORA with concurrent concussion or vice versa, leading to longer recovery times, unrecognized and/or untreated underlying pathology, and possibly additional unnecessary morbidity. According to our data, SCIWORA remains a diagnosis most common in children and young adults, and since Knox's study revealed that head trauma was the most common concurrent injury with SCIWORA across all pediatric age groups (Knox 2016), there should be more impetus to consider both diagnoses when either presents itself. This is especially true in the field of sports medicine, in which the diagnosis of concussion/mTBI is so frequent. Also, whereas concussions may be more prevalent in some sports than others, the association between concussion and SCIWORA is constant among all sports (Alas et al. 1976). The diagnosis of concurrent SCIWORA should not be ignored based on the setting or severity of concussion. However, as noted in Ellis et al., a higher severity of initial concussion symptomology does correlate with a higher prevalence of concurrent CSD (including SCIWORA) (Ellis et al. 2019). Consequently, more severe concussions should spark greater suspicion of possible concurrent SCIWORA. Admittedly, there are certain difficulties to diagnosing both SCIWORA and concussion. As Ellis and McDonald indicate, "a significant proportion of adolescents will present with subtle symptoms [of concurrent SCIWORA] including neck pain or stiffness, mild weakness or numbness of the extremities, and gait or postural instability that can be misattributed easily to the patient's SRC … [and] the presence of these symptoms always should alert the treating physician to the possibility of an occult cervical spine injury," particularly SCIWORA (Ellis and McDonald 2015). The symptoms associated with SCIWORA can have a delayed presentation, causing delayed diagnosis and management and ultimately a probably "poorer overall outcome" (Ellis and McDonald 2015). Although concurrent concussion may confound the diagnostic picture, for the best prognosis it is essential to diagnose SCIWORA as early as possible. Therefore, as Ellis et al. affirm, "all patients who present with a suspected or diagnosed SRC [need] to undergo comprehensive assessment of cervical spine and vestibulo-ocular functioning during initial and follow-up medical assessments" (Ellis et al. 2019). With the existing literature on SCIWORA with concurrent concussion somewhat sparse, there are not firmly established guidelines on the diagnosis and management of the conditions together. Future research is needed to develop these clinical tools and hopefully prevent unnecessarily poor patient outcomes as a result of missed diagnoses and treatment.

Limitations

There are several limitations to our systematic review. First, our included studies consisted of mainly lower quality studies, such as cohort studies and case reports/series, rather than higher quality studies, such as systematic reviews. Also, among many of the studies, there was insufficient reporting and detail of patients' MRI findings as well as their neurologic impairment scales (AIS, Frankel, JOA). Even among the studies that did report neurologic impairment scale scores, the variability between the use of AIS, Frankel, and JOA scales hinders comparability between them. There were also varying definitions of SCIWORA employed, such as SCIWORET, SCIWOCTET, and 'real SCIWORA,' which we accounted for in our inclusion/exclusion criteria and data collection, but still it creates more complications and intricacies in comparing and analyzing the whole data and results.

Conclusions

SCIWORA has become increasingly prevalent as the condition has become more well-known in recent years and with advances in imaging technology and techniques. However, it still remains underrecognized and underdiagnosed overall. This is especially true and troublesome with regard to the diagnosis of SCIWORA and concurrent concussion, which has negative implications for patients' long-term recovery versus morbidity. It is our recommendation [strength of recommendation = A, AAFP SORT (Ebell et al. 2004)] that concurrent SCIWORA should be considered when diagnosing concussion, and vice versa, in order to minimize the possible extended recovery time and related comorbidities.

Abbreviations

SCIWORA	Spinal cord injury without radiologic abnormality
PRISMA	Preferred Reporting Items for Systematic Reviews and Meta-Analyses
CLARITY	Clinical Advances Through Research and Information Translation
JBI	Joanna Briggs Institute
MRI	Magnetic resonance imaging
CT	Computed tomography
MOI	Mechanism of injury
SRC	Sports-related concussion
TBI	Traumatic brain injury
mTBI	Mild traumatic brain injury
AIS	American Spinal Injury Association Impairment Scale
CEBM	Centre for evidence-based medicine
AAFP SORT	American Academy of Family Physicians Strength of Recommendation Taxonomy
MVC	Motor-vehicle collision
RTA	Road traffic accident
JOA	Japanese Orthopaedic Association
CSD	Cervical spine dysfunction
PCSS	Post-Concussion Symptom Scale
SCIWORET	Spinal cord injury without the radiographical evidence of trauma
SCIWOCTET	Spinal cord injury without CT evidence of trauma
FFH	Fall from height
FDS	Fall down stairs

Supplementary Information

The online version contains supplementary material available at https://doi.org/10.1186/s42269-023-01077-y.

Additional file 1. Supplementary materials.

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Author contributions

WD conducted the literature review, synthesized the results, assessed risk of bias, and wrote the content of the article. ES identified the need for this review and acted as mentor and editor throughout the project. All authors read and approved the final manuscript.

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Declarations

Ethics approval and consent to participate

Not applicable.

Consent for publication

Not applicable.

Competing interests

The authors declare that they have no competing interests.

Author details

[1]University of Oklahoma-Tulsa School of Community Medicine, Tulsa, OK, USA. [2]Family and Community Medicine and Sports Medicine Departments, University of Oklahoma-Tulsa School of Community Medicine, Tulsa, OK, USA.

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