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Screening and diagnostic tools for complex regional pain syndrome: a systematic review

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Abstract

Complex regional pain syndrome (CRPS) is a severely painful condition that presents with a constellation of symptoms. The understanding of the pathophysiology of CRPS has evolved over time, as have the diagnostic criteria. Our primary objective was to identify screening and diagnostic tools for CRPS and summarize their feasibility, measurement properties, and study quality. A secondary objective was to identify screening and diagnostic tools used for CRPS in pediatric populations (0-21 years of age). A systematic review of English articles in electronic databases (PsycINFO, MEDLINE, Embase, CINAHL, CENTRAL, and Web of Science) was conducted with the aid of a librarian in November 2018 and updated in July 2020. Studies were included if the tool was a screening or diagnostic tool, the tool included self-report or physical examination, and the primary objective of the study was to evaluate the measurement properties or feasibility of use. For each study, data were extracted for quality indicators using the QUADAS-2 tool. No screening tools were identified. Four diagnostic tools were identified: the Veldman criteria, International Association for the Study of Pain criteria, Budapest Criteria, and Budapest Research Criteria. There are no diagnostic tools validated for use in pediatric CRPS. Because there are no extant screening tools for CRPS, all people with suspected disease should undergo rapid diagnostic assessment by a clinician. For adults, the Budapest Criteria are the preferred diagnostic tool. Future research is recommended to develop a diagnostic tool for pediatric populations and screening tools for both pediatric and adults.

Keywords: Systematic review, Complex regional pain syndrome, Diagnostic criteria, Screening tools, Chronic pain

1. Introduction

Complex regional pain syndrome (CRPS) is a severely painful condition typically in the distal region of a limb.[27] It most commonly occurs after a trauma, for which the pain is disproportionate to the extent of trauma and tissue damage.[27] Further to pain, an array of symptoms are usually present including abnormalities in sensation, trophic changes, vasomotor, motor, and autonomic dysfunction.[20] There are 2 types of CRPS: CRPS-1, which refers to CRPS in the absence of nerve damage, and CRPS-2 with related nerve damage.[27] The pathophysiology is not fully understood, although a constellation of factors have been proposed including neurogenic inflammation, maladaptive plasticity, and sensitization of nociceptors.[20]

Terminology and diagnostic criteria for CRPS have evolved. During the American Civil War, *causalgia* was used to describe burning pain after nerve injury in wounded soldiers, associated with allodynia, color, and trophic changes.[23,24] It was later described as *reflex sympathetic dystrophy* in 1943.[8] Other terms included *shoulder-hand syndrome*, *algodystrophy*, and *Sudeck atrophy*, to describe similar physiological phenomena. In 1993, the International Association for the Study of Pain (IASP) revised their taxonomy and introduced the term CRPS.[21]

The incidence of CRPS has been reportedly 5.5–25.2 cases per 100,000 person years in the United States[39] and the Netherlands,[26] respectively. Females are 3 times more likely to be diagnosed with CRPS, with cases most common in women of age 61 to 70 years.[26] The upper extremity is more frequently affected than the lower extremity,[26] and nearly half report a fracture as the inciting trauma.[26] In children and adolescents, CRPS is rare, although the exact incidence remains unknown. Pediatric CRPS affects predominately females (85%) and most often in the lower extremity (71%).[1]

A systematic review in 2014[2] revealed mixed results to explain the prognosis of CRPS. The authors concluded that some symptoms (pain, swelling, discoloration, and temperature changes) resolve between 6 and 13 months after symptom onset, whereas other symptoms (function and motor changes) tend to be chronic in nature (>1 year). Authors speculate that perhaps early interventions may be correlated with earlier symptom resolution. Some studies suggest CRPS is milder in

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children with a more favorable prognosis; however, this is not well understood.[8,18,22]

Complex regional pain syndrome is highly complex and given the large number of potential signs and symptoms, it can be challenging to diagnose. There is no gold standard radiological, laboratory, genetic, or electrical diagnostic test for CRPS. Over time, several clinical tools specifying signs and symptoms have been developed; however, they vary in their description of the disease. Having clear diagnostic criteria would allow clinicians to identify the disease accurately and initiate appropriate treatments. A screening tool would further allow clinicians to expedite access to treatments and referrals to specialists. From a research lens, a consensus on diagnostic criteria would aid in defining study populations, allowing comparisons between studies.

The primary objective was to identify and summarize the measurement properties and feasibility of screening and diagnostic tools for CRPS in all ages. A secondary objective was to identify and summarize screening and diagnostic tools used for CRPS in children and adolescents up to 21 years of age.

2. Methods

This systematic review was conducted in accordance with Preferred Reporting Items for Systematic Review and Meta-analysis guidelines (PRISMA).[4] Review methods and criteria were outlined in advance and are registered with PROSPERO systematic review protocol (#CRD42020129103).

2.1. Eligibility criteria

To address our primary objective, eligibility criteria were (1) studies that included a screening or diagnostic tool for CRPS, (2) tool based on self-report and/or physical examination, (3) studies that evaluated the measurement properties or feasibility of the tool, and (4) the tool's measurement properties were evaluated in a minimum of 2 peer-reviewed articles by different investigators. Studies were excluded if (1) the tool included quantitative sensory testing, radiological, genetic, laboratory, or electrical testing or (2) the tool was designed to further characterize previously diagnosed CRPS.

To address our second objective and identify and summarize tools that are used to diagnose and screen for CRPS in children and adolescents, eligibility criteria mirrored the criteria above for the primary objective, with the exception of removing criteria (3) and (4) related to studies that evaluated measurement properties, and restricted the search to studies that included patients of age 0 to 21 years. This age range was chosen because pediatric hospitals vary in their age cutoffs, ranging from 18 to 21 years as the upper limit. This secondary objective used the same exclusion criteria as described above. This secondary search, with criteria (3) and (4) removed, was completed in anticipation that the primary search may not reveal any diagnostic tools validated for use in pediatric setting. Understanding which tools are currently used in pediatric CRPS will shed light on what experts believe to be the best diagnostic tool for use in this age range.

2.2. Search strategy

The search strategy was developed in collaboration with a medical librarian.

Potential studies were identified through electronic database searches in PsycINFO, MEDLINE, Embase, Cumulative Index to Nursing and Allied Health Literature (CINAHL), Cochrane Central Register of Controlled Trials (CENTRAL), and Web of Science. Database search was conducted on November 21, 2018, and updated on July 31, 2020. Records were limited to English language studies. Two separate searches were conducted, one to identify CRPS tools evaluated in all ages (primary search) and another to identify tools currently used in pediatric CRPS (secondary search). The first search included key words relevant to CRPS, measurement properties, and the names of tools that were known to these authors. The second search included words relevant to CRPS, pediatric, child, adolescent, youth, and names of tools that were known to these authors. Refer to Supplementary File 1 (available at http://links.lww.com/PAIN/B228), MEDLINE Search Strategy, for an example of the search strategy used.

2.3. Study selection

Studies identified through the database search were uploaded into the web application Rayyan[31] to facilitate reviewing study titles and abstracts. Duplicate studies were removed. Study titles and abstracts were screened for eligibility by a research assistant. A random selection of study titles and abstracts (15%) were screened by a second member of the research team (G.M.). Discrepancies were discussed with a third member of the research team (A.H.) and resolved by consensus.

2.4. Data collection

To meet both objectives, 2 data collection forms were created and piloted with 5 randomly selected articles and refined accordingly. Data collection was performed by authors G.M. and A.H. and research assistants M.M. and F.N.

For the primary search (examining the measurement properties of diagnostic and screening tools for CRPS), information was extracted from each included study on: (1) tool characteristics (screening or diagnostic, language, time to complete, scoring, and cost); (2) tool constructs (number of items, signs, symptoms, equipment, and pain quality); (3) study sample (size, age range, sex, country of study, and comparison group); and (4) measurement properties.

Risk of bias of individual studies was assessed using the Quality Assessment of Diagnostic Accuracy Studies tool (QUADAS-2) by 2 raters (A.H. and K.B.).[45] QUADAS-2 is a recommended tool for use in systematic reviews of diagnostic accuracy studies. The tool evaluates risk of bias and applicability in 4 domains: patient selection, index test, reference standard, and flow and timing. Each of the 4 domains receives a rating of high, low, or unclear for risk of bias, with the first 3 also rated for applicability. Risk of bias in each domain is assessed using signaling questions to identify potential risk of bias concerns. For example, study patient selection should be done without inappropriate exclusion criteria, and the interpretation of the index and reference standard test must be done without knowledge of the result of the other. Applicability is rated based on whether the study matched the review questions. The index test is the novel test that is being evaluated for diagnostic accuracy, and the reference standard is a test that is used as a comparator. Ideally, a reference standard is 100% sensitive and specific and reveals the absolute truth about a diagnosis, positive or negative (sometimes referred to as a gold standard test). For this review, selection of the reference standard test was based on study design where an existing tool or physician diagnosis was typically used as the reference test. Given that no widely accepted and evaluated reference standard exists, no tool used as the reference standard received higher than an "unclear" score

for risk of bias within the QUADAS-2 tool.[45] Discrepancies were resolved by consensus.

With respect to the secondary search specific to the pediatric population, data extracted from each study included tool name, study setting, type of study, and study sample characteristics (including number of participants and age range).

3. Results

3.1. Study and tool selection

The primary search to identify and summarize the measurement properties of diagnostic tools for CRPS identified 20 studies involving 4 diagnostic tools for inclusion in the review. The search of electronic databases provided a total of 6444 citations. After eliminating duplicates, reviewing abstracts, full text of the remaining studies (n = 35) were reviewed in detail. From this review, 15 did not meet the inclusion criteria, resulting in 20 included studies. Refer to **Figure 1A** for the PRISMA flow diagram.[25]

The secondary search to summarize diagnostic tools for CRPS used in pediatric populations identified a total of 64 studies involving 10 diagnostic tools. The search of electronic databases provided a total of 831 citations; after eliminating duplicates and reviewing abstracts, the full text of remaining studies (n = 174) were reviewed in detail. From this review, 107 did not meet inclusion criteria, resulting in 67 included studies. Please refer to **Figure 1B**, PRISMA flow diagram for full details.[25]

3.2. Characteristics of included tools

From our primary search we identified 13 unique diagnostic tools, but no screening tools. Four diagnostic tools were evaluated in more than one article by different investigatory groups: the Veldman criteria, IASP criteria, Budapest Research Criteria, and Budapest Criteria. **Table 1** summarizes 9 diagnostic tools that were excluded as the measurement properties were only evaluated in one peer-reviewed article, organized in order of publication year. Findings related to these 4 tools are reported below in order of date of first publication. **Table 2** summarizes the criteria included in each of these 4 tools, and **Table 3** summarizes the measurement properties of each tool per each of the 20 studies included in this review. Specific details on validity (particularly specificity, sensitivity, and predictive validity) and reliability are included in **Table 3**. Sensitivity refers to the proportion of patients *with* the disease (CRPS) who *test positive*, and specificity refers to the proportion of patients *without* the disease who *test negative*.[9] Predictive validity refers to the probability of the disease, given the test results, expressed in positive (probability of disease in patients who test positive) and negative (probability of absence of disease in patients who test negative) predictive values.[9]

Table 1

Excluded studies validating other diagnostic tools.

Tool name	First author and publication year
Tahmoush 1981	Tahmoush 1981
Pediatric RSD diagnostic criteria	Stanton 1993
CRPS symptom probability scoring Scale	Sandroni 1998
Skin temperature	Wasner 2002
Atkins criteria	McBride 2008
Japanese CRPS diagnostic criteria	Sumitani 2010
Finger stiffness	Garg 2010
4 Novel bedside tests	Kuttikat 2017
CRPS prediction score	Ott 2018

Reason for exclusion: Tools were excluded because the measurement properties were only evaluated in one peer-reviewed article. CPRS, complex regional pain syndrome; RSD, reflex sympathetic dystrophy.

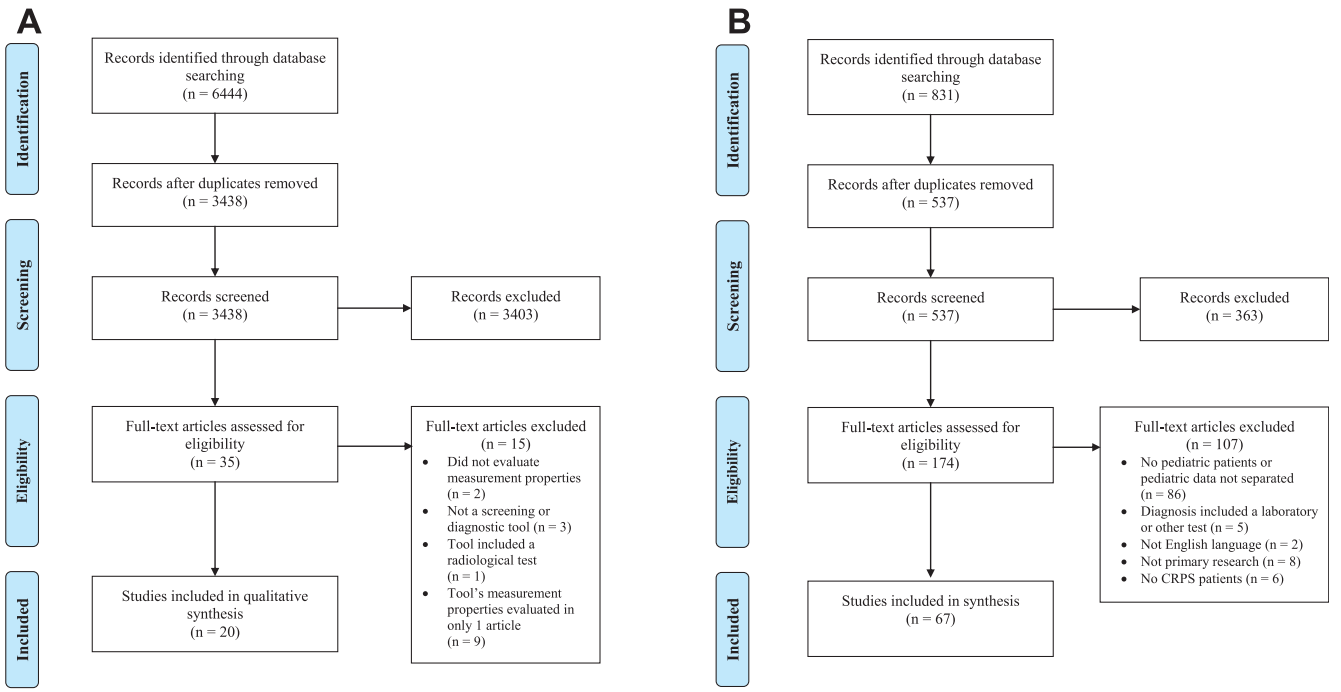

Figure 1. (A) Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) flow diagram for primary search. (B) Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) flow diagram for secondary (pediatric only) search.

Table 2

Description of criteria in included diagnostic tools for complex regional pain syndrome.

Tool criteria	Tool			
	Budapest Criteria	Budapest Research Criteria	IASP	Veldman
Signs and/or symptoms	Both	Both	Signs	Unclear
Number of criteria	4	4	4	3
Signs and symptoms				
Sensory changes	✓	✓	✓	
Edema	✓	✓	✓	✓
Increased sudomotor activity	✓	✓	✓	
Temperature differences or asymmetry	✓	✓		✓
Skin color changes or asymmetry	✓	✓	✓	✓
Motor dysfunction	✓	✓		✓
Trophic changes (hair, nail, or skin changes)	✓	✓		
Other criteria				
Trauma preceded pain			✓	
Pain is disproportionate to tissue trauma			✓	
Continuous pain	✓	✓	✓	✓
No other diagnosis can explain the signs/ symptoms	✓	✓	✓	
Pain aggravated by movement				✓

Sign = observed by clinician; Symptom = reported by patient. IASP, International Association for the Study of Pain Criteria.

There were no studies identified that evaluated the measurement properties of the 4 diagnostic tools in a well-defined cohort of children or youth (up to 21 years of age). Mean age of study participants for each included study is reported in **Table 3**. Most studies did not report the age range; therefore, it remains unclear whether any children or youth were included in these samples.[4,5,11,12,14,28,29,34–36,39,40] Three studies reported the age range with the lower limit > 21 years.[17,44,47] Two studies reported the age range including lower limits < 21 years (Ott[30]: range 10-85, mean 50.9; and Yim[46]: range 16-72, mean 40.5). However, these 2 studies did not describe how many participants were younger than 21 years of age, and given the mean age reported,it is likely a small proportion. Furthermore, the data analysis was performed on the entire set of participants and therefore it is difficult to make an inference on the diagnostic validity in participants younger than 21 years of age.

3.2.1. Veldman criteria

Veldman criteria were first published in 1993 to diagnose reflex sympathetic dystrophy.[43] According to these criteria, a diagnosis can be made if: (1) 4 of 5 are present (unexplained diffuse pain, difference in skin color relative to the other limb, diffuse edema, difference in skin temperature relative to the other limb, or limited active range of motion); (2) occurrence or increase of above signs and symptoms after use; and (3) above signs and symptoms present in an area larger than the area of primary injury/operation and including the area distal to the primary injury.[43] These criteria do not require any specific tools or equipment. The original criteria published by Veldman in 1993[43] use the terms "signs and symptoms" in criteria 2 and 3, and subsequent versions of the criteria published in the studies[28,34–36] that evaluate the criteria only use the term "symptoms" in criteria 1 to 3. For example, related to skin color changes, it is unclear if a patient would meet the criteria if they reported skin color changes at any time (symptom), or if the physician must observe this at the time of examination (sign).

Six studies evaluated the measurement properties of the Veldman criteria,[28,30,33–36] 4 of which were from the same investigatory group. Five studies were exclusive to CRPS Type 1 in the Dutch population. Perez[33] evaluated the discriminant validity of the individual criteria. The study by Ott et al.[30] was the only study to report the tool's overall sensitivity (67%) and specificity (87%). Two studies evaluated the construct validity of the individual items by comparing physician examination to objective measures of symptoms (edema, temperature, range of motion, and pain).[28,33] For example, physicians' assessment of edema as compared to volumetric measurement. Both studies reported good agreement (51%-96% agreement) between physician examination and objective measures across symptoms with respect to presence or absence of symptoms,[28,33] but poor correlations (0%-71% association) with respect to symptom severity.[33] Similar results were found with respect to interrater reliability in another study, which reported that physicians agreed upon the presence or absence of symptoms (range 88%-100%) but poor agreement with respect to symptom severity, particularly with temperature and discoloration.[34] With respect to concurrent validity, its relatedness to the Budapest Criteria and Budapest Research Criteria was quite poor (Cohen's kappa coefficient [κ] range 0.29-0.42), resulting in disagreement in diagnosis between the tools in 26% to 39% of study participants.[35]

3.2.2 International Association for the Study of Pain criteria

The IASP criteria for diagnosing CRPS were first created at an IASP meeting in Orlando in 1993, and later published in 1994.[21] According to the IASP criteria, a patient is diagnosed with CRPS if they meet all 4 criteria: (1) presence of an initiating noxious event or a cause of immobilization; (2) continuing pain, allodynia, or hyperalgesia for which the pain is disproportionate to any inciting event; (3) evidence at some time of edema, changes in skin blood flow, or abnormal sudomotor activity in the painful region; and (4) diagnosis is precluded by the existence of conditions that would otherwise account for the degree of pain and dysfunction. Our search identified 10 studies that evaluated the measurement properties of the IASP criteria.[4,11–14,29,30,35,40,44] Many studies examined the individual criteria in the tool, including the sensitivity, specificity,[11,35] and predictive validity[11,13] of each sign and symptom. The overall sensitivity and specificity of the tool ranges from 85% to 100% and 36% to 60%, respectively.[4,13,29,30] Oh et al.[29] examined the diagnostic validity specifically in poststroke patients (sensitivity 100% and specificity

Table 3

Summary of studies evaluating the measurement properties of diagnostic tools.

Study			CRPS sample				Comparison group				Measurement property
First author	Year published	Tool	CRPS type	N	Age (mean)	Sex (% female)	Sample type	N	Age (mean)	Sex (% female)	
Galer[11]	1998	IASP	NR	18	NR	NR	Diabetic neuropathy	30	NR	NR	SE, SP, and predictive validity of individual signs
Harden[12]	1999	IASP	1, 2	123	41.1	64.5%	None	—	—	—	Structural validity including principal component analysis
Oerlemans[28]	1999	Veldman	1	135	53	70%	None	—	—	—	Criterion validity
Bruehl[4]	1999	IASP, BRC	1, 2	117	41	62.4%	Neuropathic pain	42	61.5	50%	IASP: SE (98%), SP (36%); BRC: SE (70%), SP (94%), PPP (80%), NPP (90%)
Perez[34]	2002	Veldman	1	37	41.5	65.8%	None	—	—	—	Interrater reliability
Van de vusse[44]	2003	BRC, IASP	1, 2	25	42.3	92%	None	—	—	—	Interrater reliability
Perez[36]	2005a	Veldman	1	66	48.4	62%	None	—	—	—	SE, SP, and predictive validity of individual signs
Perez[33]	2005b	Veldman	1	66	48.4	62%	None	—	—	—	Criterion validity
Perez[35]	2007	Veldman, BRC, IASP	1	372	49.1	76.9%	None	—	—	—	Concurrent validity, SE, and SP of signs and symptoms
Krumova[17]	2008	BRC	NR	22	53	73%	Healthy; other limb pain	24; 18	33; 41	63%; 50%	Discriminant validity
McBride[41]	2008	BRC	1	66	NR	NR	Colles' fracture	196	NR	NR	Concurrent validity
Van bodegraven[42]	2010	BRC	Warm, cold	95	47	86%	Suspected CRPS	84	48	79.8%	Criterion validity
Harden[14]	2010a	IASP, BRC, BCC	1, 2	114	40.5	63.1%	Neuropathic pain	41	52.6	41.5%	Concurrent validity
Harden[13]	2010b	IASP, BRC, BCC	1	113	39.3	68%	Neuropathic pain	47	53.8	44.7%	Predictive validity, IASP: SE (100%), SP (41%); BCC: SE (99%), SP (68%); BRC: SE (78%), SP (79%)
Sumitani[40]	2010	IASP, BRC, BCC	1, 2	195	47.8	65.1%	Chronic limb pain	146	56.8	51.4%	Concurrent validity, BCC: SE (45%), SP (85%); BRC: SE (20%), SP (96%)
Yim[46]	2011	BRC	1, 2	104	40.5	39%	Suspected CRPS	64	42.2	43%	SE (75%), SP (95%), PPP 96.3%, and NPP 70.1% of an alternate scoring system
Zyluk[47]	2013	BRC	1	15	61	NR	Colles' fracture	105	57	NR	Discriminant validity, concurrent validity
Mailis-Gagnon[19]	2014	BCC	NR	19	47.2	89.5%	Suspected CRPS	39	44.1	79.5%	Discriminant validity
Ott[30]	2018	IASP, BCC, BRC, Veldman	1,2	1043	50.9	71%	Suspected CRPS	421	50.4	68.9%	IASP: SE (0.85), SP (0.60); BCC: SE (0.82), SP (0.68); BRC: SE (0.41) SP 0.94; Veldman: SE (0.68) SP (0.87)
Oh[29]	2019	BCC, IASP	2	6-11*	—	—	Poststroke	72	49	22.2%	BCC: SE (0.99) SP (0.68); IASP SE (1.00) SP (0.41)

* Oh 2019 reported demographic data for the total sample size of poststroke patients with and without CRPS.

BCC, Budapest Clinical Criteria; BRC, Budapest Research Criteria; CRPS, complex regional pain syndrome; IASP, International Association for the Study of Pain Criteria; NPP, negative predictive power; NR, not reported; PPP, positive predictive power; SE, sensitivity; Sign, observed by a clinician; SP, specificity; Symptom, reported by patient.

41%). Galer et al.[11] found that the positive predictive value and specificity of individual criteria were poor overall, although the specificity was greater for signs as opposed to symptoms. One-third of patients with diabetic neuropathy met the IASP criteria for diagnosis.[11] With respect to concurrent validity and its relatedness to other tools, the IASP criteria were found to have a large association with the CRPS Severity Score (Eta = 0.69, where Eta represents a nonlinear correlation coefficient with a range from 0 to 1.00).[14] Interrater reliability was examined, and the Cohen's Kappa (κ) value was 0.29 (CI: 0.03-0.55), which was preferable compared with physician diagnosis ($\kappa = 0.20$) but not as strong as the Budapest Research Criteria ($\kappa = 0.38$).[44]

Harden et al.[12] conducted a factor analysis of the individual criteria. They suggested further modification of the diagnostic

criteria, specifically to separate edema, vasomotor, and sudomotor symptoms into distinct categories rather than combined as one. The most significant factors included sensory changes, temperature asymmetry, color changes, edema, sweating, and motor dysfunction. From these results, Harden et al.[12] proposed a new set of criteria, which were later named the Budapest Research Criteria.

3.2.3. Budapest Research Criteria

These criteria were first introduced by Bruehl et al.[4] as a modified version of the IASP criteria, intended for use in research studies to define study populations. The Bruehl et al.[4] criteria have 3 components for diagnosis: (1) continuing pain, which is disproportionate to any inciting event; (2) one symptom in each of 4 categories; and (3) one sign in 2 of 4 categories; categories include sensory, vasomotor, sudomotor/edema, and motor/trophic. These criteria were later revised to include an additional fourth one, named the Budapest Research Criteria.[8] The added criteria stipulate that no other diagnosis could better explain the patient's presentation. The Bruehl et al.[4] criteria were evaluated in 7 research studies.,[4,17,30,35,41,42,44] and the Budapest Research Criteria in 5 studies.[13,14,40,46,47] Harden et al.[13] reported that the Budapest Research Criteria were found to have a more balanced profile of sensitivity (78%) and specificity (79%) compared with the IASP or Budapest Criteria. Similar results were found even when the cutoff scores were modified.[46] By contrast, Ott[30] and Sumitani[40] report the opposite, both noting a more polarized profile with poor sensitivity (20%-41%) and excellent specificity (94%-95%). With respect to concurrent validity, the Budapest Research Criteria had a high degree of relatedness to the Atkins diagnostic criteria ($\kappa = 0.79$) and the CRPS Severity Score (Eta = 0.77). Interrater reliability was found to be moderate ($\kappa = 0.38$). The discriminant validity of individual signs and symptoms was examined,[35] with another notable study[17] that examined the ability of temperature differences to discriminate between CRPS, healthy control, and people with other types of limb pain. Krumova[17] concluded that a temperature side difference of 2°C resulted in a sensitivity of 73% and specificity of 94%.

3.2.4. Budapest Criteria

In 2003, the IASP held a consensus conference in Budapest with a view to improve the IASP diagnostic criteria for CRPS. This meeting resulted in the creation of the new criteria, named the Budapest Criteria.[15] These criteria largely reflected criteria proposed earlier by Harden and Bruehl in 1999.[4,12] The Budapest Criteria includes motor and trophic features of the disease and more emphasis on signs (ie, observable by the clinician). The Budapest Criteria, intended for clinical purposes, mirror the Budapest Research Criteria with the exception of a difference in scoring. Budapest Criteria are (1) continuing pain that is disproportionate to any inciting event, (2) one symptom in 3 of 4 categories, and (3) one sign in 2 of 4 categories; categories include sensory, vasomotor, sudomotor/edema, and motor/trophic, and (4) no other diagnosis could better explain the patient's presentation. These criteria underwent initial validation in 2010 by Harden et al.,[13] where the Budapest Criteria were compared to the IASP criteria in discriminating between CRPS-1 and other types of neuropathic pain. This study concluded that both criteria had excellent diagnostic sensitivity (IASP criteria 100% and Budapest Criteria 99%), but the Budapest Criteria had superior specificity (68%) compared with the IASP criteria (41%).[13] Analysis of the discriminant validity of individual criteria was also performed in this study, with sensitivity (93%-94%) and specificity (57%-71%). Overall, the tool's diagnostic validity has been evaluated in 4 studies with highly variable results, particularly with respect to the sensitivity (sensitivity ranging from 45% to 99%) and, to a lesser extent, specificity (68%-85%).[13,29,30,40] Three additional studies evaluated additional measurement properties of the Budapest Criteria.[14,19,40] Mailis-Gagnon compared the Budapest Criteria with clinical diagnosis and found that only 27% of patients diagnosed by a community-based provider met the Budapest Criteria.[19] This study reported that more than 80% of patients who did not meet the criteria had another diagnosis to better explain their signs and symptoms, which is the fourth Budapest Criteria.[19] Concurrent validity with the CRPS Severity Score was excellent (Eta = 0.88).

3.3. Quality of studies examining the 4 diagnostic tools

QUADAS-2 risk-of-bias ratings for each study are presented in **Table 4**. **Figure 2** summarizes overall risk of bias and applicability concerns. Overall, the majority of studies showed low concern regarding applicability for the reference standard, index test, and patient selection. Scores for risk of bias indicated greater concern with more than half of studies demonstrating high or unclear risk of bias concerns for the flow and timing, reference standard, index test, and patient selection.

3.4. Diagnostic tools used in pediatric studies

Our secondary search identified 67 studies examining pediatric CRPS and extracted data on the study type as well as the tools used to diagnose CRPS in their study population. **Table 5** summarizes findings from this review and lists 10 diagnostic tools that were reportedly used in pediatric studies of CRPS. More than half of the studies identified in this review used no specific criteria to diagnose CRPS in the study population. Ten diagnostic tools were used in total, 4 of which were previously established tools (the Budapest Criteria, IASP criteria, Veldman criteria, and Japanese Diagnostic Criteria). Six studies used a unique set of diagnostic criteria defined by the study authors to diagnose CRPS. No studies used the Budapest Research Criteria to define their study population. Of note, a large proportion (37%) of studies identified were case studies or series, and only 21% of studies were interventional.

4. Discussion

This systematic review identified 4 diagnostic tools validated for use in adults, none validated in pediatric populations, and no screening tools for any age group. The 4 diagnostic tools identified include the Veldman criteria, IASP criteria, Budapest Criteria, and Budapest Research Criteria. Several studies suggest that early diagnosis intervention may lead to a more favorable outcome and potentially prevent disability and poor quality of life.[2,22] The importance of early diagnosis is recognized by the IASP that recommends rapid assessment of acute CRPS.[6] Furthermore, an accurate diagnosis is critical, given that CRPS has specific treatments that differ from other types of chronic pain; for example, common interventions for CRPS include specific physiotherapies (graded motor imagery), pharmacotherapy (intravenous ketamine), and interventions (spinal cord stimulation).[7]

The results of this review represent how our understanding of this rare disease has evolved. In 1993, expert consensus agreed upon the term CRPS, and defined 2 subtypes (1 and 2).[21] In the same year, 2 sets of diagnostic criteria (the IASP criteria and Veldman criteria) were published. Another critical time point was the expert consensus meeting held by the IASP in Budapest in 2003 whereby the former IASP criteria were replaced with the

Table 4

Study risk of bias and applicability concerns, QUADAS-2.

Study		Index test		Reference standard		Risk of bias				Applicability concerns		
First author	Year published	Tool	Applied	Tool	Applied	Patient selection	Index test	Reference standard	Flow and timing	Patient selection	Index test	Reference standard
Galer[11]	1998	Two-part questionnaire	Physician, self-report	IASP	Physician and self-report	Low	High	High	High	Low	High	High
Harden[12]	1999	Checklist	Physician	IASP	Physician	Unclear	High	Unclear	Unclear	Low	Low	Low
Oerlemans[28]	1999	Veldman (objective)	Researcher	Veldman	Physician	Low	Low	Unclear	Low	Low	Low	Low
Bruehl[4]	1999	Checklist	Physician	IASP	Physician	Low	Unclear	Unclear	Unclear	Low	High	Low
Perez[32]	2002	Veldman	Physician	Veldman	Physician	High	High	High	High	High	Low	Low
Van de vusse[44]	2003	Physician diagnosis	Physician	Physician diagnosis	Physician	High	High	Unclear	High	Low	Unclear	Low
Van de vusse[44]	2003	Physician diagnosis, IASP, BRC	Physician	Physician diagnosis	Physician	High	High	Unclear	High	Low	Low	Low
Perez[36]	2005a	Measured symptoms	Physiotherapist	Veldman	Physician	Unclear	High	Unclear	Unclear	Low	Low	Low
Perez[33]	2005b	Measured symptoms	Physician	Measured symptoms (objective)	Physical therapist/ researcher	Unclear	Unclear	Unclear	Low	Low	Low	Low
Perez[35]	2007	BRC, Veldman	Physician	IASP	Physician	Unclear	Low	Unclear	Low	Low	Low	Low
Krumova[17]	2008	Skin temperature	Not reported	BRC	Not reported	High	Low	Unclear	High	Low	High	Low
Mcbride[41]	2008	Atkins criteria	Clinician (specialty not reported)	BRC	Clinician (specialty not reported)	Low	Low	Unclear	Low	Low	Low	Low
Van bodegraven[42]	2010	Referral	Clinician or self-referral	BRC	Physician	Unclear	High	Unclear	Unclear	High	High	Low
Harden[13]	2010b	BCC	Physician	IASP	Physician	High	Low	Unclear	Unclear	Low	Low	Low
Harden[14]	2010a	CRPS severity score	Physician	IASP, BRC, BCC	Physician	Unclear	Unclear	Unclear	Low	Low	Low	Low
Sumitani[40]	2010	Japan CRPS	Physician	IASP	Physician	High	Low	Unclear	Unclear	Low	Low	Low
Yim[46]	2011	BRC - modified scoring	Physician	BRC	Physician	Low	High	High	Low	Low	Low	Low
Zyluk[47]	2013	CRPS severity score	Physiotherapist	IASP	Physiotherapist	Low	High	High	Low	Low	Low	Low
Mailis-gagnon[19]	2014	Expert diagnosis	Physician	BCC	Physician	Low	High	Unclear	Low	Low	Low	Low
Ott[30]	2018	IASP, BRC, BCC, Veldman, CRPS prediction score	Physician	Physician diagnosis	Physician	Unclear	Low	Unclear	Low	Low	Low	Low
Oh[29]	2019	BRC, BCC, IASP	Physician	BCC	Physician	Low	Low	High	Low	Unclear	Low	Low

BCC, Budapest Clinical Criteria; BRC, Budapest Research Criteria; CRPS, complex regional pain syndrome; IASP, International Association for the Study of Pain Criteria; Physician diagnosis, diagnosis made based on clinical examination and/or expert opinion.

Budapest Criteria. These new criteria were more robust, including more diverse symptoms, particularly with the addition of motor and trophic features.

4.1. Recommendations for clinicians in the adult setting

There is no gold standard laboratory, radiological, or genetic test to diagnose CRPS. This is true for many primary pain disorders, where the etiology is ill defined and can be highly variable with many contributing biological, psychological, and social factors.[27,38] In addition, CRPS is highly complex with the large number of symptoms that coexist with pain. As is the case with many pain disorders, in the absence of a gold standard test, patients are often diagnosed based on a clinical diagnosis. To aid in making a clinical diagnosis for CRPS, clinicians can use one of the 4 diagnostic tools. There are no existing screening tools for

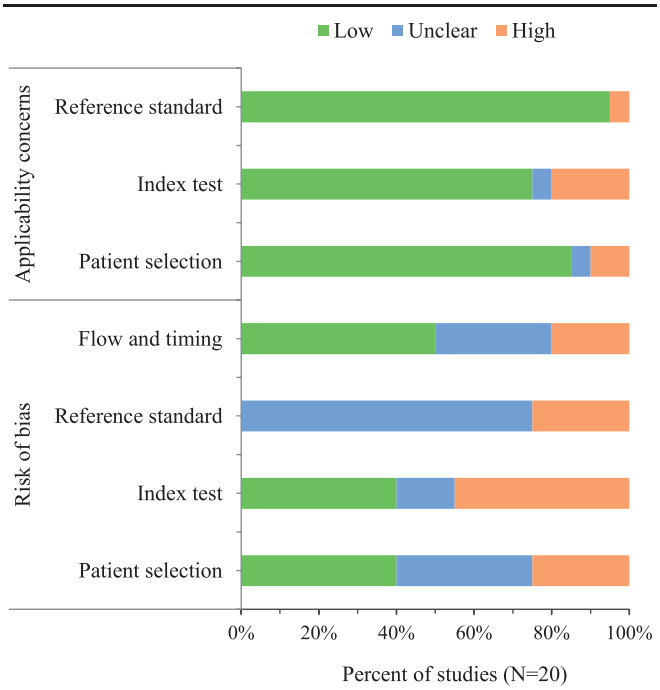

Figure 2. Summary of study risk of bias and applicability concerns, QUADAS-2.

CRPS, and as such, people with suspected CRPS should undergo rapid diagnostic assessment. Reducing wait-times for patients with suspected CRPS is recommended in community-based settings and specialist pain clinics.

Of the 4 diagnostic tools reviewed in this article, there are no significant differences in the feasibility of applying the criteria. All tools require a combination of physical examination and self-report, and none require costly equipment. The Budapest Criteria are explicitly endorsed by the IASP, the international society that makes recommendations on assessment, prevention, and treatment of pain diseases. Clinicians should follow the recommendations of the IASP and use the Budapest Criteria for diagnosing CRPS in adults. This review cannot make recommendations based on the sufficiency of measurement properties of the diagnostic tools because this review did not comprehensively examine them (eg, with a tool such as the COSMIN guideline for systematic reviews of outcome measures).[37] However, based on the data that were gathered, no tool was comprehensively evaluated across all measurement property domains, which would be required to approach a strong recommendation according to COSMIN guidelines.[37] Furthermore, there is a notably high degree of variability across studies in the sensitivity of the Budapest Criteria (45%-99%) and the Budapest Research Criteria (20%-78%). This highlights the importance of using clinical expertise, and not using any diagnostic tool as a standalone assessment.

Table 5

Summary of diagnostic tools used in studies examining pediatric CRPS (N = 67).

Characteristic	n (%)
Publication year (range)	1988-2020
Total sample size	2712
Study design	
Case study/series	25 (37.3)
Observational	28 (41.8)
Interventional	14* (20.9)
Diagnostic tools	
Veldman criteria	1 (1.5)
IASP criteria	8 (11.9)
Budapest Criteria	11 (16.4)
Japanese Diagnostic criteria	3 (4.5)
Custom†	6 (9.0)
Unclear	38 (57.2)

* Interventional: Includes nonrandomized interventional studies (11) and randomized controlled trials (3).
† Authors reported a customized list of diagnostic criteria specific to their study/centre.
CRPS, complex regional pain syndrome; IASP, International Association for the Study of Pain Criteria.

4.1.1. Recommendations for clinicians in the pediatric setting

No studies have evaluated the measurement properties of diagnostic tools in pediatric CRPS. Recent efforts are underway by Friedrich et al.,[10] who evaluated 174 youth with CRPS using the Budapest Criteria (unpublished data). Their study found that only 63% of youth who were diagnosed clinically with CRPS met the Budapest Criteria. Several studies suggest that the clinical features of CRPS in children differ from those in adults. In particular, pediatric CRPS may be milder with a more favorable prognosis.[3,10,16] One study suggests that children present most often with sensory and motor symptoms, with trophic changes being more rare.[22]

At present, clinicians should take caution when applying any diagnostic tool to children and adolescents with suspected CRPS. For clinicians in pediatric pain clinics, a clinical diagnosis based on expertise is most appropriate. For clinicians who do not have expertise in pediatric CRPS, at this time, the Budapest Criteria may be helpful to guide diagnosis. Furthermore, it is recommended that community-based providers rapidly refer patients with suspected CRPS to pediatric pain clinics. A list of international pediatric pain clinics can be found on the IASP website, http://childpain.org/wp-content/uploads/2020/02/PedPainClinicList_2020-final.pdf.[32] These specialized centers can assist in diagnosis and treatment. As described above, pediatric pain clinics should make efforts to reduce wait-times for patients with CRPS, with a target time of one week.[6]

4.2. Recommendations for future research

Future research is needed to comprehensively evaluate the spectrum of measurement properties of existing diagnostic tools. This is especially true for the 9 diagnostic tools that were excluded from this review (**Table 2**) because there was only one study evaluating the tool's measurement properties. Two tools in particular (4 Novel Bedside Tests and CRPS Prediction Score) were only recently published, and future evaluation may reveal whether there is merit in their respective use. Studies should indicate clearly the diagnostic criteria that are used and how they are applied, including who applied them and tools that were used (eg, how temperature is measured). Future studies should use a consistent reference standard, ideally the Budapest Research Criteria. A consistent reference standard would facilitate comparing results across studies, with the potential for pooling results in meta-analyses. In future study designs, clinicians must evaluate the patient using the diagnostic criteria without knowledge of the reference standard, and therefore blinded. As symptoms of CRPS are known to fluctuate over time, study participants should be evaluated with the diagnostic criteria and the reference standard in close proximity (less than one week).

Because CRPS is a rare disease, multisite research studies are crucial to minimize the limitations of drawing conclusions from small sample sizes while ensuring standardization in procedures across sites. Studies should avoid unnecessary exclusions or case-control groups that may inflate identified differences between groups.

For research studies examining adults with CRPS, for example, for interventional studies, the IASP recommends the Budapest Research Criteria for diagnosing patients with the intent of defining study populations because there is some evidence to suggest higher specificity with this tool.[13]

Future research is needed to develop and/or validate a diagnostic tool for pediatric CRPS, and a screening tool for CRPS for both children and adults. A self-report screening tool for CRPS would help clinicians who may not have the requisite knowledge, skill, or judgement to use diagnostic criteria. Furthermore, a screening tool would aid in identifying patients on waitlists who need rapid assessment to confirm diagnosis. Ideally, a screening tool would have excellent sensitivity as opposed to a diagnostic tool where a more balanced profile of sensitivity and specificity is best suited.

4.3. Limitations

This review identified and summarized screening and diagnostic tools for CRPS. This review only included studies that evaluated the measurement properties of the tools when looking across the lifespan, and as such, we may have missed newly developed tools that have not yet been validated. Furthermore, we did not evaluate the sufficiency of the measurement properties and therefore cannot provide strong recommendations on this aspect. Another study limitation is with respect to our quality assessment using QUADAS-2, which is a tool for evaluating the risk of bias and applicability of diagnostic accuracy studies. QUADAS-2 was intended to evaluate studies that use a reference standard test that is 100% sensitive and specific. Because there is no such test for CRPS, most studies included in this review used another set of diagnostic criteria in place of a true reference standard. For example, Perez[35] evaluated the diagnostic accuracy of the Budapest Criteria and Veldman criteria compared with the IASP criteria as a reference standard. As a result of these limitations, no study evaluated could receive the highest possible score on the QUADAS-2 diagnostic accuracy assessment.

5. Conclusions

There are 4 diagnostic tools for CRPS in adult populations, and none in children or adolescents. These include the Veldman criteria, IASP criteria, Budapest Criteria, and Budapest Research Criteria. A quality assessment revealed a high risk of bias in the studies that evaluated the measurement properties of these 4 diagnostic tools. Further research is needed to validate these existing tools. The authors recommend that for adults with CRPS, clinicians use the Budapest Criteria for diagnosis in combination with clinical judgement, and researchers to use the Budapest Research Criteria. For pediatric CRPS, there are no valid diagnostic criteria, and caution should be taken if applying any of the above criteria. A clinical diagnosis by a pediatric pain specialist is preferred. Unfortunately, to date, there are no screening tools for CRPS. All people with suspected CRPS should be assessed rapidly by a clinician to undergo diagnostic assessment and appropriate treatment. Future research is recommended to develop a diagnostic tool for pediatric populations and screening tools for both children and adults.

Conflict of interest statement

The authors have no conflicts of interest to declare.

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Appendix A. Supplemental digital content

Supplemental digital content associated with this article can be found online at http://links.lww.com/PAIN/B228.

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