The natural history of crouch gait in bilateral cerebral palsy: a systematic review

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The Natural History of Crouch Gait in Bilateral Cerebral Palsy: A Systematic Review.

Abstract

Aim: To systematically review the natural history of crouch gait in bilateral cerebral palsy (CP) in the absence of surgical intervention and to review any relationship between clinical variables and progression of knee crouch.

Methods: Relevant literature was identified by searching article databases (PubMed, CINAHL, EMBASE, and Web of Science). Included studies reported on participants with bilateral CP who had 3-dimensional gait analysis on at least two occasions with no surgical interventions between analyses.

Results: Five papers (4 retrospective cohort studies; 1 case report) comprised the final selection. Studies varied in follow-up times and participant numbers. Increased knee flexion over time was reported in the four retrospective studies with two distinct patterns of increasing knee flexion evident. Only the case-study reported improved knee extension between assessments. Four studies demonstrated increased hamstring tightness over time with the biggest increases related to longer follow-up time rather than increase in crouch. .

Conclusion and Implications: The existing literature suggests that the natural history of crouch gait is towards increasing knee flexion over time. Future prospective studies of bigger groups are needed to examine the relationship between increasing crouch and clinical variables.

Shortened Title: Crouch gait in cerebral palsy

What this paper adds?

Crouch gait is very prevalent in cerebral palsy (CP) and has been shown to increase the forces acting on the knee and the energy cost of gait. It has been suggested that if left untreated this gait pattern can lead to pain, joint deformity, radiological abnormalities and loss of independent gait. This systematic review suggests that, in the absence of surgical intervention, the natural history of crouch gait is towards increasing knee flexion over time. The rate of progression of crouch gait appears to increase in those with knee flexion greater than 20^0 during gait suggesting that this might be an indicator for more urgent intervention. While studies have shown that not all those who walk in crouch have short hamstrings, this pattern is typically treated with surgical lengthening of this muscle group. This systematic review found that while hamstring tightness increased over time, this appeared to occur secondary to walking in crouch rather than contributing to the progression of this pattern. However, the results should be interpreted with caution due to the limited number of studies meeting the inclusion criteria of this systematic review as well as the methodological limitations of the included studies. This review highlights that longer term follow ups of larger numbers are needed to examine the multi-factorial nature of crouch progression in CP. Any further study should include assessment of function and participation in conjunction with gait analysis in the laboratory.

Keywords: cerebral palsy; crouch gait; knee flexion; kinematics.

1. Introduction

Cerebral Palsy (CP) refers to a group of clinical presentations due to an insult to the developing brain. It is the most common cause of motor deficiency in young children and has recently been reported to occur in 2.1 per 1000 live births (Oskoui, Coutinho, Dykeman, Jette, & Pringsheim, 2013). The associated motor deficiency usually leads to muscle spasticity, weakness and joint contractures which have a significant effect on gait and it is generally accepted that some of these patients lose their walking ability as they get older (Andersson & Mattsson, 2001). Numerous gait presentations are associated with CP but crouch gait is one of the most common pathological patterns, with a reported prevalence of 72%-76% in this population (Wren, Rethlefsen, & Kay, 2005). Crouch gait is defined as excessive flexion of the knee throughout the stance phase of gait (Law, 2014). This flexed knee posture during gait leads to excessive forces and demands on the knees (Steele, Demers, Schwartz, & Delp, 2012; Steele, van der Krogt, Schwartz, & Delp, 2012) and increases the energy cost of gait (J. Rose, Gamble, Medeiros, Burgos, & Haskell, 1989). It is thought that if left untreated crouch often leads to knee and low-back pain (Jahnsen, Villien, Aamodt, Stanghelle, & Holm, 2004), bone deformities (Kerr Graham & Selber, 2003) and ultimately can lead to significant radiological abnormalities (O'Sullivan, Walsh, Kiernan, & O'Brien, 2010) and loss of independent gait (Opheim, Jahnsen, Olsson, & Stanghelle, 2009).

For these reasons improving knee extension in stance phase is an integral aim of multi-level orthopaedic surgery in CP. Crouch has typically been treated with surgical lengthening of the hamstrings. Studies have reported improved knee function following hamstring lengthening though increased anterior pelvic tilt is a common finding after such surgery (DeLuca, Ounpuu, Davis, & Walsh, 1998; Dreher et al., 2012; Kay, Rethlefsen, Skaggs, & Leet, 2002; Park et al., 2009; Sung et al., 2013).
However, studies based on computer musculo-skeletal modelling have suggested that not all those who walk in crouch have short hamstrings (Arnold, Liu, Schwartz, Ounpuu, & Delp, 2006). For this reason more recent treatment has focused on augmenting the power of the knee extensors (Joseph, Reddy, Varghese, Shah, & Doddabasappa, 2010) and the need for hamstring lengthening has been questioned (Healy, Schwartz, Stout, Gage, & Novacheck, 2011). Extension osteotomy of the femur with or without patellar tendon shortening provides an alternative means of surgically correcting flexed knee gait. This technique has shown comparable effects compared to hamstring lengthening but also leads to increased anterior pelvic tilt (Klotz et al., 2016; Sossai et al., 2015).
A recent systematic review examined the effectiveness of management of crouch gait in CP. Of the 30 papers included which addressed orthopaedic surgical interventions, 27 addressed the effects of hamstring lengthening and only three investigated extension osteotomy and patellar tendon advancement/shortening (Galey, Lerner, Bulea, Zimbler, & Damiano, 2017). This review highlighted that hamstring lengthening remains the only well supported intervention even though surgical practices have evolved beyond this simplistic approach. One of the difficulties in assessing the effectiveness of surgical intervention in CP is that studies tend to be small, retrospective and

uncontrolled (Galey et al., 2017; McGinley et al., 2012). Understanding the natural progression of pathological gait in CP in the absence of surgical intervention is therefore important to provide a baseline against which any intervention can be assessed. However, to date, few gait studies have focused on the natural history of walking in children and young adults with CP and the need for larger clinically based prospective studies of walking in CP using objective gait analysis data has been highlighted (Opheim, McGinley, Olsson, Stanghelle, & Jahnsen, 2013).

The primary aim of this systematic review was to determine the natural progression of knee flexion during gait in adults and children with bilateral spastic ambulant cerebral palsy, GMFCS I-III. A secondary aim was to review measures of hamstring tightness to examine any relationship between this clinical variable and the progression of crouch gait.

2. Methods

The protocol for this review was according to the preferred reporting items for systematic reviews and meta-analyses (PRISMA) statement (Moher et al., 2015) and was registered with the PROSPERO register of systematic reviews (http://www.crd.york.ac.uk/PROSPERO, registration number CRD42016035766). The PRISMA checklist is attached as Appendix B.

2.1 Search Strategy

A literature search was conducted using the following databases from inception to October 2017- Pubmed, Cumulative Index to Nursing and Allied Health Literature (CINAHL), Excerpta Medica Database (EMBASE), and Web of Science.

The search term strategy, allowing for syntactical differences between data-bases, was as follows: “Cerebral Palsy” OR “spastic diplegia” OR “diplegia” AND “knee” OR “crouch” OR “crouch knee” OR “flexed knee gait” AND “kinematics” OR “biomechanics” OR “walking” OR “gait analysis” OR “gait disorders” OR “gait.” All terms were searched in both titles and abstracts.

2.2 Inclusion Criteria

Published studies in the English language on the progression of crouch gait in CP were included if they met the following criteria:

1. Retrospective and prospective cohort and observational studies of adults or children;
2. They reported on ambulant participants with bilateral cerebral palsy;
3. Assessment included 3-dimensional gait analysis on at least two occasions to include knee flexion angle;
4. No surgical interventions between gait analyses.

2.3 Study Selection and Quality assessment

Two of the review authors (RO’S and HF) independently reviewed titles and abstracts and full text studies were then obtained where it was necessary to determine if the article met inclusion criteria. Disagreements were resolved by a third author (FH).

Quality assessment was carried out independently by two authors (RO'S and HF). However, as the lead author (RO'S) was also an author on one of the included papers (O'Sullivan et al., 2010); this paper was reviewed by another two of the review authors (HF and FH). The quality assessment of the majority of the included articles was performed using a modified version of the Downs and Black checklist (Downs & Black, 1998) proposed by Schmid et al (Schmid, Schweizer, Romkes, Lorenzetti, & Brunner, 2013). This checklist discarded items on interventions and reduced the item on statistical power of the measurements from the original six to three options. Instead, items on the reproducibility of measurement procedures were added. The final checklist consists of 17 items with a maximum score of 20 points, including the five different categories ''quality of reporting'' (eight items, maximum 10 points), ''external validity'' (three items, maximum three points), ''internal validity – bias'' (three items, maximum three points), ''internal validity – confounding'' (two items, maximum two points) and ''statistical power'' (one item, maximum two points). One of the included studies was a case report, so in this instance, the Joanna Briggs Institute critical appraisal checklist for case reports was used (Briggs, 2016).

2.4 Data extraction and synthesis

For the included studies, relevant data was extracted by one author (RO'S) using a custom data extraction form. Extracted data included study details, study design, characteristics of the study sample, number lost to follow up, time between assessment, knee flexion angle during stance phase of gait and clinical measure of hamstring tightness. If included, summary gait scores (Gillette Gait Score (GGI) or Gait Deviation Index (GDI)) were also extracted (Schutte et al., 2000; Schwartz & Rozumalski, 2008). The original authors were contacted in two cases to request additional information before data extraction. In one case the authors supplied data on bilateral CP patients only, which was not available in the original manuscript (Bell, Õunpuu, DeLuca, & Romness, 2002). In another data were supplied for each of the individual time-points (Butler, Steele, Torburn, Gamble, & Rose, 2016).

A meta-analysis was not conducted due to the heterogeneity of the data retrieved and the data were summarised narratively.

3. Results

3.1 Search Results

The search strategy yielded 1368 unique citations from four databases. After title and abstract review, 18 remained for full text review and five articles comprised the final review. Included articles and reasons for exclusion of 13 of the 18 articles for which full text was obtained are outlined in Figure 1.

Insert Figure 1 about here

Figure 1. Flow diagram of the literature search

3.2 Study Design

Of the five studies, four were retrospective cohort studies and one was a case study. The study type was not clearly stated in one case though it is assumed to be a retrospective cohort. The mean sample size of the five studies was 15.2 participants (range 1 to 30). The single participant case study reported on annual gait analysis over 8 consecutive years. The four retrospective studies reported on two gait analyses with a mean of 3.28 years between assessments (range 1.05 to 8 years).
Two studies divided their over-all group into sub-groups. Gough (Gough & Shortland, 2008) presented data on two sub-groups. The first, Gough SND (surgery not done) group, were those for who surgery was recommended based on a gait analysis but was not carried out due to a decision by the family or local medical team. The second, Gough SNR (surgery not recommended) group, comprised those who were not recommended surgical intervention following a gait analysis. Rose (G. E. Rose, Lightbody, Ferguson, Walsh, & Robb, 2010) divided the group based on the length of follow-up into a shorter interval (mean 5 years) and longer interval (mean 7.5 years). The study design is summarised in Table 1 along with participant characteristics and extracted data.

3.3 Study participants

Of the 76 total participants in the included studies, 40 were male and 36 female, all had a diagnosis of bilateral CP and all were under 18 years (mean age at first assessment 8.41±1.76 years). The GMFCS levels were reported for 49 participants (I=10; II=33; III=5; IV=1) but not specified for the remaining 27 participants from two studies (Bell et al., 2002; O'Sullivan et al., 2010). None of the reported groups had any surgical intervention between assessments. One study reported that all participants suffered a disruption of the knee extensor mechanism between the two assessments (O'Sullivan et al., 2010). The participant data for each study is summarised in Table 1.

Table 1. Study characteristics and extracted data

Study	Study Type	Sub-Groups	N	GMFCS Levels I/II/III/IV	Sex M/F	Gait Analysis Session	Mean Age(yrs)	Interval (yrs)	KFMS (^{0})	MKES (^{0})	Mean Popliteal Angle º(SD)	Summary Gait Score (GDI† or GGI‡)
Bell	RC	N/A	19		6/13	1	7.3(1.8)	4.3 (2.5)		11.8(14)		
						2	11.6(3.43)			14.3 (18.67)		
Butler	CS	N/A	1	1/0/0/0	1/0	1	6	1		36.1	67.5	59.5†
						2	7	1		30.3	72.5	60.5
						3	8	1		17.4	67.5	64
						4	9	1		21.55	67.5	60.5
						5	10	1		30.6	72.5	63.5
						6	11	1		17.45	75	69
						7	12	1		14.55	67.5	67
						8	13			21.4	70	68.5
Gough	RC	SNR	15	4/10/1/0	6/9	1	8.7	1.05		5	56	286‡
						2	9.75			7	60	207
		SND	15	4/9/2/0	7/8	1	10.1	1.16		24	65	1079
						2	11.17			34	68	1144
O'Sullivan	RC	N/A	8		6/2	1	10.63(2.92)	3.6	21.6 (10.35)		58.5(8.18)	
						2	14.13(3.09)		44.4 (18)		71.8(4.37)	
Rose	unclear	Full Group	18	1/14/2/1	14/4	1	7.7	6.3	10.5	7.5	53	72.5†
						2	14		18.5	16	66.1	71.83
		Shorter Interval	9	0/6/2/1	6/3	1	8.2	5	12	8	52.9	69.2
						2	13.2		16.5	14	60.5	67
		Longer Interval	9	1/8/0/0	8/1	1	7.3	7.5	9	7	58.1	75.8
						2	14.8		21	18	71.25	76.7

KFMS=Knee flexion at mid-stance; MKES=Maximal knee extension in stance;
RC=Retrospective cohort; CS= case study SNR=surgery not recommended; SND= surgery not done; N/A= Not Applicable
†Gait Deviation Index (GDI); ‡Gillette Gait Index (GGI)

3.4 Qualitative assessment

The methodological quality of the four included retrospective studies is presented in Table 2. The modified Downs and Black scores ranged from 9 to 13 (maximum score: 20). All studies scored below 50% on the Reporting sub-scale and zero on the Power sub-scale.

The included case study scored 'yes' on 6 of the 8 items on the Joanna Briggs Institute critical appraisal checklist for case reports. This report scored 'unclear' on the remaining two items- 'Was the post-intervention clinical condition clearly described?' and, 'Were adverse events (harms) or unanticipated events identified and described?'

Table 2. Methodological quality of included studies.

	Modified Downs and Black Scale					
Study	Reporting/10	External Validity/3	Bias/3	Confounders/2	Power/2	Total/20
Bell et al.	9	1	1	2	0	13
Gough et al.	7	2	1	1	0	11
O'Sullivan et al.	6	1	1	1	0	9
Rose et al.	8	1	2	0	0	11

3.5 Progression of crouch gait

Four studies reported on maximal knee extension in stance phase while one study reported on knee flexion at mid-stance (O'Sullivan et al., 2010). Irrespective of the variable reported, increased knee flexion during stance was reported over time in four of the included studies and only the single participant case-study (Butler et al., 2016) reported on a net improvement in knee extension between first and last assessment. Table 1 and Figure 2 show the measure of knee flexion in stance versus age at each assessment for all of the five studies including sub-groups where relevant.

Insert Figure 2 about here

Figure 2. Progression of knee flexion in stance versus age

An over-all gait summary score was reported by three studies (see Table 1). The GDI was used by two studies. Rose(G. E. Rose et al., 2010) found no significant change in the GDI for the group as a whole (from 72.5 to 71.83), while Butler(Butler et al., 2016) reports an improvement from 59.5 to 68.5 in a single participant over eight years. Gough (Gough & Shortland, 2008) used the GGI and found no significant changes in either the SNR group (from 286 to 207) or the SND group (from 1079 to 1144). However, there was a significant difference between the SNR and SND groups at baseline.

3.6 Clinical measure of hamstring tightness

All but one study (Bell et al., 2002) reported on popliteal angle measure of hamstring tightness and all four of these studies showed some degree of increased hamstring tightness over the course of the study. The progression in hamstring tightness is summarised in Table 1 and Figure 3. The mean increase in popliteal angle reported was 8.09^{o} (range 2.5^{o} to 13.3^{o}). Knee flexion contracture was reported in one study that demonstrated an increase in contracture from 1.5^0 to 18.2^o although this group had a disruption of the knee extensor mechanism between assessments (O'Sullivan et al., 2010).

Insert Figure 3 about here

Figure 3. Progression of popliteal angle measure of clinical hamstring tightness versus age

4 Discussion

The purpose of this systematic review was to examine the natural history of crouch gait in bilateral CP in the absence of surgical intervention. Overall, the natural history appeared to be towards increasing knee flexion during gait with faster progression evident in those with knee flexion greater than 20^0. Hamstring tightness appears to occur over time secondary to crouch rather than causing this gait pattern. However, the results should be interpreted with caution due to methodological limitations of included studies.

4.1 Progression of crouch

Only one single participant case study (Butler et al., 2016) reported a net improvement in crouch from 36.1^o of knee flexion at age six to 21.4^o at age thirteen. Looking at eight time points allowed distinct periods of improvement and dis-improvement to be appreciated unlike the group studies that only looked at two analyses. However, the authors highlight that the case study is not representative of all children with spastic CP and the crouch improved with aggressive non-operative management, botulinum toxin, dedicated home exercise program and post intervention rehabilitation. Additionally, it is notable that the degree of crouch at first assessment is far in excess of the initial value seen in any of the other groups. These issues highlight the difficulty in extrapolating more generally from this single participant case study.

Outside of this case study, the remaining four studies (Bell et al., 2002; Gough & Shortland, 2008; O'Sullivan et al., 2010; G. E. Rose et al., 2010) all recorded an increase in knee flexion during stance phase between two repeated gait assessments. Although it was not possible to combine the data due to heterogeneity of the groups, Figure 2 highlights that there appears to be two patterns of crouch progression. The first pattern of gradual increase in crouch over time is shown by five of the groups/sub-groups (Gough SNR, Bell, Rose Full Group, Rose Short-Term and Rose Long-Term). The differences in population ages, length of follow-up and degree of crouch at initial assessment make formal analysis difficult but the slopes of the gradual increase in crouch appear to be similar.

In contrast, the Gough SND group and the O'Sullivan group demonstrate a second pattern of more rapid increase in crouch. Again, the heterogeneity of the groups makes analysis difficult but it is notable that in both these groups the degree of crouch at initial assessment was greater than 20^0 compared to a mean of 8^0 (range 5^0 to 11.8^o) in those showing more gradual progression. Gough (Gough & Shortland, 2008) reports that this knee extension in stance value better differentiated those likely to deteriorate from those who are more stable (Gough SNR and Gough SND groups) compared to clinical examination. O'Sullivan highlighted the increased strain on the knee extensors at increased crouch above 20^{o} and also suggested that this degree of crouch contributes to deterioration. Both authors consider this degree of crouch to be an indicator for intervention while the relatively stable gait pattern, as demonstrated by the other 5 sub-groups, does not require surgical intervention. The Gough SND did not have the recommended surgical intervention and this contributed to the deterioration in this group. Gough confirms this by including another group who were similar at baseline but had the recommended orthopaedic surgery which prevented this deterioration and led to an improvement in the crouch pattern. As this reviewed focussed on the natural progression of crouch gait this data is not included in our results. The O'Sullivan group all had x-ray findings consistent with disruption of the knee extensor mechanism, which occurred between the two analyses and the significant deterioration was attributed to this. X-ray findings were not reported in the Gough SND group but it may be that some of the group may have similar knee extensor disruption that contributed to the similar pattern of deterioration.

4.2 Relationship between clinical hamstring tightness and crouch gait

Information on the popliteal angle measure of hamstring tightness was extracted from four studies and all reported increased hamstring tightness between the first and last assessment. The mean increase (8.09±5.03^{0}) in tightness was variable between the studies (range 2.5^{o} to 13.3^{o}) and it appeared that in general, the longer the follow-up time the larger the increase in hamstring tightness irrespective of the increase in crouch during gait. This is highlighted in Figure 3, which in contrast to Figure 2 shows relatively uniform progression in popliteal angle over time which does not appear to be related to the progression of crouch during gait. For example, both the Gough SNR and Gough SND groups had similar minimal increase in popliteal angle measures (4^{o} and 3^{o} respectively) after a follow-up time of one year. This was despite a significant 10^{o} increase in knee flexion during gait in the SND group. The Rose long-term group had a similar increase in crouch (11^{o}) but demonstrated a much larger increase in hamstring tightness (13.15^{o}) on assessments 7.5 years apart. This is consistent with the finding that not all those who walk in crouch have short hamstrings (Arnold et al., 2006) and suggests that the hamstrings tighten over time in response to walking in crouch rather than actually causing crouch gait. A recent systematic review reported that hamstring surgery is the only intervention for crouch for which substantial evidence exists (Galey et al., 2017) and highlighted the need for further research on interventions beyond hamstring lengthening and also the need for prospective study on crouch to examine the multi-factorial contributors to this gait pattern.
The O'Sullivan group had the biggest increase in hamstring tightness (13.3^{0}) over a time period of 3.6 years. However, this group also had significant pathological findings on x-ray, which obviously did influence hamstring tightness, and they were also the only group to report an increase in knee flexion contracture.

4.3 Confounding variables and potential limitations.

The majority of the studies were retrospective reviews of two time-points, with one single participant case study included. All participants were diagnosed with bilateral CP and were able to complete a gait analysis and were therefore able to ambulate. However, GMFCS levels I-IV were included highlighting significant variability in presentations. The effect of GMFCS level on progression of crouch could not be analysed as this was not reported for 27 of the participants included in this review. There was also variation in follow-up times and in general participant numbers tended to be relatively small. The largest group studied was 30 participants (divided into two sub-groups) but follow-up time in this case was just over one year (Gough & Shortland, 2008). In contrast, the longest follow-up time was eight years in the case of a single participant case-study (Butler et al., 2016) and the next longest was 7.5 years but this was of a sub-group of only nine participants (G. E. Rose et al., 2010). This again, highlights the need for larger scale prospective studies in this population (Opheim et al., 2013).

In addition the methodological quality of the included studies was low with an average modified Downs and Black assessment score of 10.75 from a possible score of 20. All studies scored less than 50% on the reporting sub-scale and all scored zero on the power sub-scale as none of the included papers carried out a power analysis. This, again, highlights that caution should also be used when interpreting the results of these studies.

4.5 Considerations for future research

This systematic review highlights the need for a larger scale prospective study on the progression of crouch gait over a longer time frame. As with the systematic review on crouch treatment (Galey et al., 2017), there was a lack of functional outcomes reported in any of the studies. While gait analysis can accurately assess the increasing knee flexion over time this may not reflect changes in function and participation. Any further study should include a validated outcome measure of mobility and physical functioning to assess the impact of increasing crouch on these domains.

None of the studies reported on gait in adults. This may be because orthopaedic surgery is the preferred method of treating musculoskeletal deformities associated with CP (Lamberts, Burger, du Toit, & Langerak, 2016) and the included studies did not encounter adult participants who had not had surgery. Studies have highlighted falls and decline in mobility and quality of life in the adult CP population (P. E. Morgan, Soh, & McGinley, 2014; P. Morgan & McGinley, 2014) but this review highlights the significant scarcity of objective gait data in this population.

5 Conclusion

Outside of one single participant case study the other four studies included in this systematic review demonstrate that the natural history of crouch gait in bilateral CP appears to be towards increased knee flexion over time. The heterogeneity of the included study populations and variation in participant numbers and follow up times make formal analysis difficult however, there appears to be two patterns of increasing crouch. The first is a gradual increase in knee flexion over time, which may not require surgical intervention. The second pattern is of more significant, rapid deterioration, which might be prevented with appropriately timed orthopaedic intervention. Knee flexion in stance appears to best differentiate these two groups with values of 20^{o} or more of particular concern and a potential disruption of the knee extensor mechanism should be investigated. Increasing clinical measure of hamstring tightness appears to occur over time secondary to knee flexion rather than causing it. This review highlights the lack of objective gait data in the adult population and the need for further prospective studies to examine the multi-factorial nature of crouch progression in CP. Any further study should include assessment of function and participation in conjunction with gait analysis in the laboratory.

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Appendix A: Pubmed Search Strategy

1. Cerebral Palsy (MeSH)
2. Spastic Diplegia (MeSH)
3. cerebral palsy OR spastic diplegia OR diplegia
4. Or/ 1- 3
5. Knee (MeSH)
6. Knee OR crouch OR crouch knee OR flexed knee gait
7. Or/ 5 - 6
8. Gait (MeSH)
9. Gait Disorders, Neurologic (MeSH)
10. Walking (MeSH)
11. gait OR walk* OR kinematics OR biomechanics
12. Or/ 8 - 11
13. Combine 4 and 7 and 12
14. Limit Humans
15. Limit to English

Appendix B: PRISMA Checklist

Section/topic	#	Checklist item	Reported on page #
TITLE			
Title	1	Identify the report as a systematic review, meta-analysis, or both.	1
ABSTRACT			
Structured summary	2	Provide a structured summary including, as applicable: background; objectives; data sources; study eligibility criteria, participants, and interventions; study appraisal and synthesis methods; results; limitations; conclusions and implications of key findings; systematic review registration number.	2
INTRODUCTION			
Rationale	3	Describe the rationale for the review in the context of what is already known.	3
Objectives	4	Provide an explicit statement of questions being addressed with reference to participants, interventions, comparisons, outcomes, and study design (PICOS).	4
METHODS			
Protocol and registration	5	Indicate if a review protocol exists, if and where it can be accessed (e.g., Web address), and, if available, provide registration information including registration number.	4
Eligibility criteria	6	Specify study characteristics (e.g., PICOS, length of follow-up) and report characteristics (e.g., years considered, language, publication status) used as criteria for eligibility, giving rationale.	4
Information sources	7	Describe all information sources (e.g., databases with dates of coverage, contact with study authors to identify additional studies) in the search and date last searched.	4
Search	8	Present full electronic search strategy for at least one database, including any limits used, such that it could be repeated.	Appendix A
Study selection	9	State the process for selecting studies (i.e., screening, eligibility, included in systematic review, and, if applicable, included in the meta-analysis).	4
Data collection process	10	Describe method of data extraction from reports (e.g., piloted forms, independently, in duplicate) and any processes for obtaining and confirming data from investigators.	5
Data items	11	List and define all variables for which data were sought (e.g., PICOS, funding sources) and any assumptions and simplifications made.	5
Risk of bias in individual studies	12	Describe methods used for assessing risk of bias of individual studies (including specification of whether this was done at the study or outcome level), and how this information is to be used in any data synthesis.	4-5
Summary measures	13	State the principal summary measures (e.g., risk ratio, difference in means).	5

Synthesis of results	14	Describe the methods of handling data and combining results of studies, if done, including measures of consistency (e.g., I^2) for each meta-analysis.	5
Section/topic	**#**	**Checklist item**	**Reported on page #**
Risk of bias across studies	15	Specify any assessment of risk of bias that may affect the cumulative evidence (e.g., publication bias, selective reporting within studies).	4-5
Additional analyses	16	Describe methods of additional analyses (e.g., sensitivity or subgroup analyses, meta-regression), if done, indicating which were pre-specified.	N/A
RESULTS			
Study selection	17	Give numbers of studies screened, assessed for eligibility, and included in the review, with reasons for exclusions at each stage, ideally with a flow diagram.	5 and Figure 1
Study characteristics	18	For each study, present characteristics for which data were extracted (e.g., study size, PICOS, follow-up period) and provide the citations.	6 and Table 1
Risk of bias within studies	19	Present data on risk of bias of each study and, if available, any outcome level assessment (see item 12).	8 and Table 2
Results of individual studies	20	For all outcomes considered (benefits or harms), present, for each study: (a) simple summary data for each intervention group (b) effect estimates and confidence intervals, ideally with a forest plot.	8-9 and Table 1 and Figures 2 and 3
Synthesis of results	21	Present results of each meta-analysis done, including confidence intervals and measures of consistency.	N/A
Risk of bias across studies	22	Present results of any assessment of risk of bias across studies (see Item 15).	8 and Table 2
Additional analysis	23	Give results of additional analyses, if done (e.g., sensitivity or subgroup analyses, meta-regression [see Item 16]).	N/A
DISCUSSION			
Summary of evidence	24	Summarize the main findings including the strength of evidence for each main outcome; consider their relevance to key groups (e.g., healthcare providers, users, and policy makers).	9-11
Limitations	25	Discuss limitations at study and outcome level (e.g., risk of bias), and at review-level (e.g., incomplete retrieval of identified research, reporting bias).	10-11
Conclusions	26	Provide a general interpretation of the results in the context of other evidence, and implications for future research.	11
FUNDING			

Funding	27	Describe sources of funding for the systematic review and other support (e.g., supply of data); role of funders for the systematic review.	12