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Citation for final published version:

Quinn, Lori, Busse-Morris, Monica, Carrier, Judith, Fritz, N., Harden, Jane, Hartel, L., Kegelmeyer, D., Kloos, A. and Rao, A. 2017. Physical therapy and exercise interventions in Huntington's disease: a mixed methods systematic review protocol. JBI Database of Systematic Reviews and Implementation Reports 15 (7) , pp. 1783-4433. 10.11124/JBISRIR-2016-003274 file

Publishers page: <http://dx.doi.org/10.11124/JBISRIR-2016-003274>
<<http://dx.doi.org/10.11124/JBISRIR-2016-003274>>

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Physical therapy and exercise interventions in Huntington's disease: a mixed methods systematic review protocol

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Review question/objective

The review question is: What is the effectiveness of physiotherapy and therapeutic exercise interventions in people with Huntington's disease and what are patients, families and caregivers' perceptions of these interventions?

The specific objectives are:

1. To determine the effectiveness of physiotherapy and therapeutic exercise interventions in people with Huntington's disease on physical and cognitive function.
2. To explore the perception of people with Huntington's disease and their families and caregivers with regard to their experience of physiotherapy and exercise interventions
3. This mixed methods review seeks to develop an aggregated synthesis of quantitative, qualitative and narrative systematic reviews on physiotherapy and exercise interventions in Huntington's disease, in an attempt to derive conclusions and recommendations useful for clinical practice and policy decision-making.

Background

Huntington's disease (HD) is a rare, dominantly inherited, neurodegenerative condition caused by a cytosine, adenine, and guanine (CAG) segment repeat expansion in the Huntingtin (HTT) gene. Huntington's disease has a prevalence of 6-13/100,000 in the general population;¹a 2012 meta-

analysis reported the worldwide prevalence of HD was 2.71 per 100,000 (95% CI: 1.55-4.72).² It predominantly affects the brain, causing dysfunction and death of medium spiny striatal projection neurons and thus disruption of corticostriatal pathways with resultant impairment of cognition, motor function, and behaviour.¹ These impairments result in decreasing independence in activities of daily living and quality of life^{3,4} even from relatively early in the disease.

Therapeutic exercise interventions are a promising area of research in neurodegenerative diseases.^{5,6} Addressing motor and cognitive impairments in neurodegeneration may provide a long-term beneficial effect to delay disease progression, maximize functional abilities, and maintain independence over a longer period. Loss of independent mobility and care dependency have been shown to be important predictors of nursing home admissions.^{7,8} The potential to develop interventions that facilitate independent living and strategies to manage symptoms are urgently required.

Despite the potential for physiotherapy and exercise interventions, which may be provided by physiotherapists, exercise trainers or other healthcare personnel, to assist people with HD, there is little evidence to suggest that people with HD are routinely referred for physical therapy.⁹ This may be attributable to the limited scientific support for the effectiveness of physiotherapy and exercise interventions. The Physiotherapy Working Group of the European Huntington Disease Network (EHDN) developed a Physiotherapy Guidance Document in 2009.¹⁰ A systematic literature search was conducted to summarize the available evidence prior to this date, with the view to providing therapists with information and recommendations for physiotherapy standards of care for people with Huntington's disease. This Guidance Document, however, was based largely on expert opinion and the limited available literature at the time, which involved a small number of mostly descriptive studies. Subsequent to the publication of the Guidance Document, the EHDN group developed treatment-based classifications to guide clinical decision-making across the life course of the disease.¹¹

In the past 7 years, there has been a significant increase in the number and quality of physiotherapy and exercise studies in HD.¹²⁻¹⁸ These studies have ranged from evaluation of short-term exercise programs,^{13,14} video-game home interventions,¹⁵ as well as inpatient multidisciplinary rehabilitation programs.^{12,16,17} Studies have utilized both quantitative and qualitative assessments, and in addition there is a growing body of textual references supporting exercise and physiotherapy in this population. While the EHDN Guidance Document¹⁹ and subsequent treatment-based classifications^{11,20} were an important first step to providing information about patient management in this relatively rare disease, we are now at a point where more rigorous clinical guidelines can be developed. Importantly, the treatment-based classifications, and the evidence in support of associated interventions, require detailed validation.

We searched for previous or currently registered systematic reviews on the topic of physiotherapy and exercise in HD in the Cochrane and Joanna Briggs Institute databases, PROSPERO, the World Health Organization (WHO) International Clinical Trials Registry (ICRTP), Medline and CINAHL. In 2003, a systematic review on the effectiveness of physiotherapy, occupational therapy and speech therapy in Huntington's disease was published; however, at the time there was minimal available literature to include.²¹ Studies examining the outcomes of physiotherapy, occupational therapy, and speech pathology interventions for people with Huntington's disease up to May 2002 were included in this review, and it was concluded that there was a low level of evidence to support the use of physiotherapy for addressing impairments of balance, muscle strength, and flexibility in Huntington's disease. No other systematic reviews or protocols were found following a search of the Joanna Briggs Institute, Cochrane and Prospero databases.

In this proposed mixed methods review, the quantitative component will seek to incorporate a wider range of study designs, including but not limited to, cohort studies (with control), case-controlled studies, descriptive and case series designs. A qualitative and textual component will also be incorporated to help understand why initiatives do or do not work from the perspective of people with HD, their families and caregivers. Combining quantitative, qualitative and textual syntheses in the same review will make this the first mixed methods systematic review that considers the effectiveness of a range of physiotherapy and therapeutic exercise interventions in people with Huntington's disease and the experiences and perceptions of patients, their families and caregivers with regard to these interventions. In addition, each study will be categorized according to the established treatment-based classifications,^{11,20} and the evidence for each classification will be discussed in the narrative synthesis. If a study cannot be adequately categorized, this will be discussed separately and recommendations for additional categories will be made as appropriate

Keywords

Huntington's disease; exercise; physical therapy; physiotherapy; physical activity; treatment-based classifications

Inclusion criteria

Types of participants

Inclusion criteria for included paper have been developed based upon the authors' awareness of the characteristics of studies and papers published in this area. For the first objective the quantitative component of this review will include studies that include those of any age or gender who have been clinically or genetically diagnosed with Huntington's disease, including those that have tested positive for the Huntington's disease gene in premanifest, prodromal, early, mid or late stage disease.

For the second objective the qualitative and textual component of this review will consider studies that include those of any age or gender who have been clinically or genetically diagnosed with Huntington's disease, including those that have tested positive for the Huntington's disease gene in premanifest, prodromal, early, mid or late stage disease will be included, in addition to studies which include or focus on family members and caregivers that relate to this patient group.

Types of intervention(s)/phenomena of interest

The quantitative component of this review will consider studies that evaluate physiotherapy and exercise interventions such as aerobic exercise, strengthening exercises, gait or walking training, treadmill training, balance training, yoga, Pilates, Tai-chi (and variants), relaxation, technology-delivered exercise, dance, aquatics, daily living strategies, sensory stimulation, cueing (i.e., visual, verbal or physical prompts including attentional strategies with internal cues); chaining (i.e., breaking down the task); dual-task training (i.e., motor-cognitive training), task-specific training, education, flexibility range of motion (ROM) exercises, breathing exercises, wheelchair evaluation, seating, wheelchair mobility training, positioning, splinting, posture (alignment and exercises), manually assisted cough, and non-invasive ventilation. Other physiotherapy and exercise interventions as determined by the literature in the area will also be incorporated, as necessary. Comparators of interest for those included interventions, where applicable, will be usual care. Usual care at present involves no specific intervention for individuals with Huntington disease. Studies whose primary aim is to evaluate a pharmacological or psychological intervention will be excluded.

The qualitative component of this review will consider all studies that identify and explore the perceptions and experiences of people with Huntington's disease, their families and caregivers with regard to physiotherapy and exercise interventions.

The textual component of this review will consider all studies that describe the perceptions and experiences of people with Huntington's disease and their families and caregivers with regard to physiotherapy and exercise interventions.

Types of outcomes

The quantitative component of this review will consider studies that include outcomes related to physical and cognitive function regardless of how they were measured. For example, outcome measures of physical function include: balance, fitness (cardiovascular function), goal attainment, motor function and performance, muscle strength, number of falls, physical activity, pulmonary function, rate of chest infections, and ulcer staging, spatiotemporal and kinematic parameters of gait and balance, walking ability and endurance

For example, outcome measures of cognitive function include: cognition, psychological measures (depression, anxiety, apathy)

Other physiotherapy outcomes as determined by the literature in the area will also be incorporated, as necessary.

The qualitative components of this review will consider studies that identify patient, family or caregiver perceptions of physiotherapy and exercise interventions including: Patient and family/caregiver experiences, perceived improvement and satisfaction where these have been explored using qualitative methods.

The textual component of this review will consider papers/reports that describe patient, family or caregiver perceptions of physiotherapy and exercise interventions including: Patient and family/caregiver experiences, Perceived improvement, Satisfaction

Context

The qualitative component of this review will consider studies that investigate physiotherapy and exercise interventions that are carried out in the community, hospital settings, clinics, rehabilitation centers, or patient's homes.

Types of studies

The quantitative component of the review will consider both experimental and epidemiological study designs including randomized controlled trials, non-randomized controlled trials, quasi-experimental studies, before and after studies, prospective and retrospective cohort studies, case control studies and analytical cross sectional studies for inclusion. The quantitative component of the review will also consider descriptive epidemiological study designs including case series, individual case reports and descriptive cross sectional studies for inclusion.

The qualitative component of the review will consider studies that focus on qualitative data including, but not limited to, designs such as phenomenology, grounded theory, ethnography, action research and feminist research.

The textual component of the review will consider discussion papers, position papers and other text based on expert clinical opinion.

Search strategy

The search strategy aims to find both published and unpublished studies. A three-step search strategy will be utilized for each component in this review. An initial limited search of PubMed and

CINAHL will be undertaken followed by an analysis of the text words contained in the title and abstract, and of the index terms used to describe article. A second search using all identified keywords and index terms will then be undertaken across all included databases. Thirdly, the reference list of all identified reports and articles will be searched for additional studies. Studies published in English only will be considered for inclusion in this review. Studies published between May 2002 –the present will be considered for inclusion in this review. May 2002 was the completion date for the previous systematic review.²¹

The databases to be searched include:

EBSCO CINAHL, PEDro, MEDLINE via PubMed, Cochrane Library, Scopus, SPORTDiscus, PsycINFO

The search for unpublished studies will include:

Google, Web of Science (Books, proceedings, other), ClinicalTrials.gov, WHO International Clinical Trials Registry Platform (ICTRP), ISRCTN Registry, Prospero, National Guideline Clearinghouse (NGC), Resources from professional organizations such as: Huntington Study Group (HSG), European Huntington's Disease network (EHDN), American Physical Therapy association (APTA). Initial keywords to be used will be: huntington* disease, physical therap*, physical activit*, exercise, physiotherapy*

Assessment of methodological quality

Papers selected for retrieval will be assessed by two independent reviewers for methodological validity prior to inclusion in the review using standardized critical appraisal instruments from the Joanna Briggs Institute Meta-Analysis of Statistics Assessment and Review Instrument (JBI-MAStARI) (Appendix I). Any disagreements that arise between the reviewers will be resolved through discussion, or with a third reviewer.

Qualitative papers selected for retrieval will be assessed by two independent reviewers for methodological validity prior to inclusion in the review using standardized critical appraisal instruments from the Joanna Briggs Institute Qualitative Assessment and Review Instrument (JBI-QARI) (Appendix I). Any disagreements that arise between the reviewers will be resolved through discussion, or with a third reviewer.

Textual papers selected for retrieval will be assessed by two independent reviewers for authenticity prior to inclusion in the review using standardized critical appraisal instruments from the Joanna Briggs Institute Narrative, Opinion and Text Assessment and Review Instrument (JBI-NOTARI) (Appendix I). Any disagreements that arise between the reviewers will be resolved through discussion, or with a third reviewer.

Data extraction

Quantitative data will be extracted from papers included in the review using the standardised data extraction tool from JBI-MAStARI (Appendix II). The data extracted will include specific details about the interventions, populations, study methods and outcomes of significance to the review question and specific objectives.

Qualitative data will be extracted from papers included in the review using the standardised data extraction tool from JBI-QARI (Appendix II). The data extracted will include specific details about the populations, phenomena of interest, and context as well as methodology, methods, settings and cultural information study methods related to the review question and specific objectives.

Textual data will be extracted from papers included in the review using the standardised data extraction tool from JBI-NOTARI (Appendix II). The data extracted will include specific details about the populations, study methods related to the review question and specific objectives.

All reviewers will have practiced using the extraction tools in order to apply the tools consistently. Reviewers will extract data independently before conferring.

Authors of both quantitative and qualitative primary studies will be contacted for missing information or to clarify unclear data.

Following segregated synthesis of the included quantitative and qualitative studies, the results of each single method synthesis included in the mixed method review will be extracted in numerical, tabular or narrative format. For example, for syntheses of quantitative data, this will consist of appropriate elements of the meta-analysis Forest plot or, where applicable, an evidence table; for qualitative data, it will consist of appropriate elements of the QARI-view table, for textual opinion data it will consist of appropriate elements of the NOTARI-view table..

Data synthesis

Quantitative data will be, where possible, pooled in statistical meta-analysis using JBI-MAStARI. All results will be subject to double data entry. Effect sizes expressed as odds ratio (for categorical data) and weighted mean differences (for continuous data) and their 95% confidence intervals will be calculated for analysis. Heterogeneity will be assessed statistically using the standard Chi-square. Where statistical pooling is not possible the findings will be presented in narrative form including tables and figures to aid in data presentation where appropriate.

Qualitative research findings will be pooled using JBI-QARI. This involves the aggregation or synthesis of findings to generate a set of statements that represent that aggregation, through assembling the findings (Level 1 findings) rating according to their quality, and categorizing these findings on the basis of similarity in meaning (Level 2 findings). These categories will then be subjected to a meta-synthesis in order to produce a single comprehensive set of synthesized findings (Level 3 findings) that can be used as a basis for evidence-based practice. Where textual pooling is not possible the findings will be presented in narrative form.

The findings of each single-method synthesis included in this review will be aggregated using the JBI method of meta-aggregation. This will involve the configuration of the findings to generate a set of statements that represent that aggregation through coding any quantitative to attribute a thematic description to all quantitative data; assembling all of the resulting themes from quantitative and qualitative syntheses; and the configuration of these themes to produce a set of synthesised findings in the form of a, set of recommendations or conclusions.

The findings will be presented in narrative form including tables and figures to aid in data presentation where appropriate.

Conflicts of interest

Authors of included papers who were also part of the review team will not be included in the review or data extraction.

Acknowledgements

Funding for this review is provided by Huntington Study Group, European Huntington Disease Network, and Griffin Foundation.

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Appendix I: Appraisal instruments

MAStARI appraisal instrument

JBI Critical Appraisal Checklist for Randomised Control / Pseudo-randomised Trial

Reviewer Date

Author Year Record Number

	Yes	No	Unclear	Not Applicable
1. Was the assignment to treatment groups truly random?	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
2. Were participants blinded to treatment allocation?	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
3. Was allocation to treatment groups concealed from the allocator?	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
4. Were the outcomes of people who withdrew described and included in the analysis?	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
5. Were those assessing outcomes blind to the treatment allocation?	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
6. Were the control and treatment groups comparable at entry?	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
7. Were groups treated identically other than for the named interventions?	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
8. Were outcomes measured in the same way for all groups?	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
9. Were outcomes measured in a reliable way?	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
10. Was appropriate statistical analysis used?	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

Overall appraisal: Include Exclude Seek further info.

Comments (Including reason for exclusion)

JBI Critical Appraisal Checklist for Descriptive / Case Series

Reviewer Date

Author Year Record Number

	Yes	No	Unclear	Not Applicable
1. Was study based on a random or pseudo-random sample?	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
2. Were the criteria for inclusion in the sample clearly defined?	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
3. Were confounding factors identified and strategies to deal with them stated?	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
4. Were outcomes assessed using objective criteria?	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
5. If comparisons are being made, was there sufficient descriptions of the groups?	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
6. Was follow up carried out over a sufficient time period?	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
7. Were the outcomes of people who withdrew described and included in the analysis?	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
8. Were outcomes measured in a reliable way?	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
9. Was appropriate statistical analysis used?	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

Overall appraisal: Include Exclude Seek further info

Comments (Including reason for exclusion)

JBI Critical Appraisal Checklist for Comparable Cohort/ Case Control

Reviewer Date

Author Year Record Number

	Yes	No	Unclear	Not Applicable
1. Is sample representative of patients in the population as a whole?	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
2. Are the patients at a similar point in the course of their condition/illness?	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
3. Has bias been minimised in relation to selection of cases and of controls?	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
4. Are confounding factors identified and strategies to deal with them stated?	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
5. Are outcomes assessed using objective criteria?	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
6. Was follow up carried out over a sufficient time period?	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
7. Were the outcomes of people who withdrew described and included in the analysis?	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
8. Were outcomes measured in a reliable way?	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
9. Was appropriate statistical analysis used?	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

Overall appraisal: Include Exclude Seek further info.

Comments (Including reason for exclusion)

QARI appraisal instrument

JBI QARI Critical Appraisal Checklist for Interpretive & Critical Research

Reviewer Date

Author Year Record Number

	Yes	No	Unclear	Not Applicable
1. Is there congruity between the stated philosophical perspective and the research methodology?	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
2. Is there congruity between the research methodology and the research question or objectives?	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
3. Is there congruity between the research methodology and the methods used to collect data?	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
4. Is there congruity between the research methodology and the representation and analysis of data?	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
5. Is there congruity between the research methodology and the interpretation of results?	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
6. Is there a statement locating the researcher culturally or theoretically?	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
7. Is the influence of the researcher on the research, and vice-versa, addressed?	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
8. Are participants, and their voices, adequately represented?	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
9. Is the research ethical according to current criteria or, for recent studies, and is there evidence of ethical approval by an appropriate body?	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
10. Do the conclusions drawn in the research report flow from the analysis, or interpretation, of the data?	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

Overall appraisal: Include Exclude Seek further info.

Comments (Including reason for exclusion)

NOTARI appraisal instrument

**JBI Critical Appraisal Checklist for Narrative,
Expert opinion & text**

Reviewer Date

Author Year Record Number

	Yes	No	Unclear	Not Applicable
1. Is the source of the opinion clearly identified?	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
2. Does the source of the opinion have standing in the field of expertise?	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
3. Are the interests of patients/clients the central focus of the opinion?	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
4. Is the opinion's basis in logic/experience clearly argued?	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
5. Is the argument developed analytical?	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
6. Is there reference to the extant literature/evidence and any incongruency with it logically defended?	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
7. Is the opinion supported by peers?	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

Overall appraisal: Include Exclude Seek further info

Comments (Including reason for exclusion)

Appendix II: Data extraction instruments

MAStARI data extraction instrument

**JBI Data Extraction Form for
Experimental / Observational Studies**

Reviewer Date

Author Year

Journal Record Number

Study Method

RCT Quasi-RCT Longitudinal

Retrospective Observational Other

Participants

Setting _____

Population _____

Sample size

Group A _____ Group B _____

Interventions

Intervention A _____

Intervention B _____

Authors Conclusions: _____

Reviewers Conclusions: _____

Study results

Dichotomous data

Outcome	Intervention () number / total number	Intervention () number / total number

Continuous data

Outcome	Intervention () number / total number	Intervention () number / total number

QARI data extraction instrument

**JBI QARI Data Extraction Form for Interpretive
& Critical Research**

Reviewer Date

Author Year

Journal Record Number

Study Description

Methodology
.....
.....

Method
.....
.....

Phenomena of Interest
.....
.....

Setting
.....
.....

Geographical
.....
.....

Cultural
.....
.....

Participants
.....
.....

Data analysis
.....
.....

Authors Conclusions
.....
.....

Comments
.....
.....

Complete

Yes

No

NOTARI data extraction instrument

JBI Data Extraction for Narrative, Expert opinion & text

Reviewer Date

Author Year Record Number

Study Description

Type of Text:

Those Represented:

Stated Allegiance/ Position:

Setting

Geographical

Cultural

Logic of Argument

Data analysis

Authors Conclusions

Reviewers Comments

Data Extraction Complete

Yes

No

