



**Protocol for an HTA report: Does therapeutic writing help people with long term conditions? Systematic review, realist synthesis and economic modelling**

Journal:	<i>BMJ Open</i>
Manuscript ID:	bmjopen-2013-004377
Article Type:	Protocol
Date Submitted by the Author:	31-Oct-2013
Complete List of Authors:	Meads, Catherine; Brunel University, Health Economics Research Group PerezNyssen, Olga; QMUL, Wong, Geoff; QMUL, Steed, Liz; Barts and The London School of Medicine and Dentistry, Centre for Primary Care and Public Health Bourke, Liam; QMUL, Ross, Carol; Cumbria NHS, Hayman, Sheila; Medical Foundation for the Care of Victims of Torture, Field, Victoria; Freelance, Lord, Joanne; HERG, Brunel University, Greenhalgh, Trisha; Queen Mary University of London, Centre for Primary Care and Public Health Taylor, Stephanie; Queen Mary University of London, Primary Care and Public Health
<b>Primary Subject Heading</b>:	Patient-centred medicine
Secondary Subject Heading:	Health economics
Keywords:	systematic review, realist review, PUBLIC HEALTH

SCHOLARONE™  
Manuscripts

# Protocol for an HTA report: Does therapeutic writing help people with long term conditions? Systematic review, realist synthesis and economic modelling

Meads C<sup>1</sup>, Nyssen OP<sup>2#</sup>, Wong G<sup>2</sup>, Steed L<sup>2</sup>, Bourke L<sup>2</sup>, Ross CA<sup>3</sup>, Hayman S<sup>4</sup>, Field V<sup>5</sup>, Lord J<sup>1</sup>, Greenhalgh T<sup>2</sup>, Taylor SJC<sup>2</sup>,

<sup>1</sup> Health Economics Research Group, Brunel University

<sup>2</sup> Centre for Primary care and Public Health, Barts and The London School of Medicine and Dentistry, Queen Mary University of London

<sup>3</sup> Cumbria Partnership NHS Foundation Trust

<sup>4</sup> Medical Foundation for the Care of Victims of Torture

<sup>5</sup> Freelance experienced therapeutic writing practitioner

# - Correspondence to:

Olga Pérez Nyssen, o.pereznyssen@qmul.ac.uk

<sup>2</sup>Centre for Primary Care and Public Health

Blizard Institute

Barts and The London School of Medicine and Dentistry

Yvonne Carter Building

58 Turner Street

London E1 2AB

## ABSTRACT

### Introduction

Long-term medical conditions (LTCs) cause reduced health-related quality of life and considerable health service expenditure. Writing therapy has potential to improve physical and mental health in people with LTCs, but its effectiveness is not established. This project aims to establish the clinical and cost-effectiveness of therapeutic writing in LTCs by systematic review and economic evaluation, and to evaluate context and mechanisms by which it might work, through realist synthesis.

### Methods

Included are any comparative study of therapeutic writing compared to no writing, waiting list, attention control or placebo writing in patients with any diagnosed LTCs that report at least one of the following; relevant clinical outcomes; quality of life; health service use; psychological, behavioural or social functioning; adherence or adverse events. Searches will be conducted in the main medical databases including Medline, Embase, PsychInfo, The Cochrane Library and Science Citation Index. For the realist review further purposive and iterative searches through snowballing techniques will be undertaken. Inclusions, data extraction and quality assessment will be in duplicate with disagreements resolved through discussion. Quality assessment will be using GRADE criteria. Data synthesis will be narrative and tabular with meta-analysis where appropriate. *De novo* economic modelling will be attempted in one clinical area if sufficient evidence available, and performed according to the NICE reference case.

### Funding and Dissemination

This project was funded by the National Institute for Health Research Health Technology Assessment (NIHR HTA) Programme (project number 11/70/01) and will be published in full in Health Technology Assessment. The views and opinions expressed therein are those of the authors and do not necessarily reflect those of the HTA programme, NIHR, NHS or the Department of Health. Findings will be disseminated through peer-reviewed journals, national and international conferences and relevant public health networks. This project is registered with PROSPERO database - CRD42012003343.

## Background

### Therapeutic writing

Writing as a form of therapy to improve physical or mental health has a long history<sup>1</sup> and can take many formats including those from a psychotherapeutic background such as therapeutic letter writing,<sup>2</sup> specific controlled interventions such as emotional disclosure/ expressive writing,<sup>3</sup> to more recent approaches such as developmental creative writing<sup>4</sup> and other epistolary approaches such as blogging.<sup>5</sup> Other forms of potentially therapeutic writing include reflective diaries, free-writing, short stories, song-writing, unsent letters and memoirs. Additionally, therapeutic writing interventions might be delivered in different contexts: as individual self-help therapy at home, in a health-care centre, as part of a programme in a rehabilitation clinic or within a group of people with similar or different health conditions, in person or through the world wide web. People engaging in therapeutic writing can either receive feedback from a peer, from a healthcare professional, from their writing group or receive no feedback.<sup>6</sup>

With the development of UK organisations such as LAPIDUS (Association for Literary Arts in Personal Development) dedicated to the promotion of therapeutic writing based on the premise that it has health benefits, it is important to evaluate the effectiveness of a variety of different approaches

The most evaluated form of therapeutic writing is the expressive writing intervention as described by Pennebaker and colleagues<sup>3, 7, 8, 9</sup> Expressive writing is a technique whereby people are encouraged to write (or talk into a tape recorder) in private about a traumatic, stressful or upsetting event, usually from their recent or distant past. They write for 15-30 minutes typically for 3-4 days within a relatively short time period such as consecutive days or within 2 weeks. Participants are encouraged to write about their deepest thoughts and feelings concerning an event or experience they have not talked about with others. The control group may receive no treatment, or a written exercise with a non-emotional topic, or be on a waiting list. Randomised controlled trials (RCTs) of expressive writing have been conducted in a wide variety of participants including healthy students, people undergoing psychological stressors, such as bereavement, or in people with LTCs, such as rheumatoid arthritis and asthma.

Expressive writing is thought to be beneficial for longer term health effects and as such is now frequently referred to in general psychology textbooks as a potentially beneficial intervention.<sup>10</sup> However, some have argued that this intervention is too brief to have any long lasting effects.<sup>9, 11</sup> There has been remarkably little critique published specifically on expressive writing.

Those within the field of developmental creative writing however consider that Pennebaker's expressive writing paradigm may be more a starting point in learning to release emotion through writing, but that added benefit may occur with more 'free writing' which could allow for development and shaping of the material, which leads to a

1  
2  
3  
4  
5  
6  
7  
8  
9  
10  
11  
12  
13  
14  
15  
16  
17  
18  
19  
20  
21  
22  
23  
24  
25  
26  
27  
28  
29  
30  
31  
32  
33  
34  
35  
36  
37  
38  
39  
40  
41  
42  
43  
44  
45  
46  
47  
48  
49  
50  
51  
52  
53  
54  
55  
56  
57  
58  
59  
60

'a new relationship with aspects of self-experience'.<sup>4</sup> It is this connection with a core sense of self from which creative writing is said to derive benefit.<sup>4</sup> With newer forms of writing, such as blogging, association with increased perceived social support has been demonstrated.<sup>5</sup> Given the importance of social support to well-being this suggests yet further mechanisms by which writing in its various formats may improve health.

It is a popular assumption that creative writing helps overcome life's stresses, and some professional writers have noted this. Notwithstanding the epistemological, methodological and ethical challenges of studying the impact of creative writing on mental health, it seems appropriate to evaluate whether, the wider field of therapeutic writing might help with mental and physical LTCs.

There have been several systematic reviews and meta-analyses on emotional disclosure/expressive writing, one of which was undertaken by our own team.<sup>8, 12-15</sup> The Cochrane Library lists a protocol for evaluation of written emotional disclosure for asthma.<sup>16</sup> Our preliminary scoping searches did not find any previous economic evaluations of any forms of therapeutic writing. None of the systematic reviews or meta-analyses incorporates a qualitative systematic review or a realist review to make sense of how and why therapeutic writing might work.

### LTCs

The prevalence of LTCs increases with ageing populations. In 2002, the leading chronic diseases (cardiovascular disease, cancer, chronic respiratory disease and diabetes) were responsible for 29 million deaths worldwide.<sup>17</sup> According to the UK Department of Health (DoH),<sup>18</sup> more than 15 million people in England (including half of all those aged over 60 years) are living with at least one LTC, and the risk of death is particularly high in those with three or more conditions occurring concurrently.<sup>19</sup> LTCs also result in a huge burden on NHS resources. Although some are preventable, for most LTCs continuing care is the only realistic management strategy, as biological and psychosocial mechanisms regulating disease progression are not yet fully understood. Since LTCs are difficult to improve, especially for elderly populations, health-care programmes such as self-management support and patient education, often combined with structured clinical follow-up, have been suggested as a way to improve the quality of life of such patients.<sup>20</sup> New therapeutic approaches may help to extend the lifespan and quality of life in people with LTCs.

### Realist Reviews

These reviews ask "what works for whom in what circumstances?" and consider the interaction between context, mechanism and outcome (sometimes abbreviated as C-M-O). i.e. how particular contexts have 'triggered' (or, conversely, interfered with) mechanisms to generate the observed outcomes.<sup>21</sup> The philosophical basis is realism, which assumes the existence of an external reality (a 'real world') but one that is 'filtered' (i.e. perceived, interpreted and responded to) through human senses, volitions, language and culture. Such human processing initiates a constant process of self-

1  
2  
3 generated change in all social intuitions, a vital process that has to be accommodated in  
4 evaluating social programmes.  
5

6  
7 In order to understand how outcomes are generated, the roles of both external reality  
8 and human understanding and response need to be incorporated. Realism does this  
9 through the concept of mechanisms, whose precise definition is contested but for which  
10 a working definition is ‘...underlying entities, processes, or structures which operate in  
11 particular contexts to generate outcomes of interest’.<sup>22</sup> Different contexts interact with  
12 different mechanisms to make particular outcomes more or less likely – hence a realist  
13 review produces recommendations of the general format “In situations [X], complex  
14 intervention [Y], modified in this way and taking account of these contingencies, may be  
15 appropriate”. This approach, when done well, is widely recognised as a robust  
16 methodology which is particularly appropriate when seeking to explore the interaction  
17 between C-M-O in a complex intervention. (See for example Berwick’s editorial in JAMA  
18 explaining why experimental (RCT / meta-analysis) designs may need to be  
19 supplemented (or perhaps in some circumstances replaced) by realist studies aimed at  
20 elucidating C-M-O configurations).<sup>23</sup>  
21  
22  
23  
24

25 A realist approach is particularly useful for this project because therapeutic writing is a  
26 complex intervention which could be useful in a variety of patient groups and currently it  
27 is unclear whether it is effective for all or some, and how and why it might be effective.  
28  
29

### 30 **Objectives of the project**

- 31  
32 1. What are the different types of therapeutic writing that have been evaluated in  
33 comparative studies? What are their defining characteristics? How are they  
34 delivered? What underlying theories have been proposed for their effect/s?  
35
- 36 2. What is the clinical effectiveness of the different types of therapeutic writing for  
37 LTCs compared to no writing or other suitable comparators?  
38
- 39 3. How is heterogeneity in results of empirical studies accounted for in terms of  
40 patient and/or contextual factors, and what are the mechanisms and moderators  
41 responsible for the success, failure or partial success of interventions (i.e. what  
42 works for whom in what circumstances and why)?  
43  
44
- 45 4. What is the cost effectiveness of one or more types of therapeutic writing in one  
46 or more representative LTCs where there is sufficient information on the  
47 intervention, comparator and outcomes to conduct an economic evaluation.  
48  
49  
50  
51  
52  
53  
54  
55  
56  
57  
58  
59  
60

## Research Methods

### Systematic review of effectiveness and realist review

We will undertake two interlinked reviews simultaneously (see Table 1 for inclusion criteria). No language restrictions will be applied.

Table 1: Review question components

Question components	Systematic review	
	Inclusion criteria	Exclusion criteria
<b>Population</b>	Any LTC as per DoH definition. <sup>18</sup>	Acute conditions, stress, bereavement etc.
<b>Exposure / Intervention</b>	Any form of therapeutic writing including emotional disclosure/ expressive writing, poetry, diaries etc.	Talking to a listener, counselling, psychotherapy, talking into a tape recorder, mobile phone or similar where this the primary mode of delivering the intervention, expressive drama, dance, film-making. Evaluation of other people's writing.
<b>Comparison</b>	Non writing, waiting list, inexpressive writing, attention controls, any control thought to be inactive.	Any active or possibly active control including therapeutic writing or talking into a tape recorder or mobile phone.
<b>Outcome</b>	Any relevant clinical outcomes including both disease specific outcomes and generic outcomes such as: quality of life; health service use; psychological outcomes; behavioural outcomes; social functioning; adverse events; adherence to therapies; costs.	Intermediate physiological outcomes such as salivary cortisol, immune parameters not routinely measured in the management of LTCs.
<b>Study designs</b>	Any comparative studies including RCTs, cohort or case-control studies. Economic evaluations.	Single case reports, case series, studies where results for intervention and control groups not presented separately.



## Search Strategy

The searches inform both reviews, find any previous models that have been conducted in therapeutic writing and provide some inputs to the decision-analytic model. A database of published and unpublished literature will be assembled from searches using a comprehensive search strategy, examination of reference lists in systematic reviews, hand searching journals and contact with experts in the area. The wider searches will map the extent of relevant literature (mapping review). From this list of studies, appropriately includable studies for the systematic review will be selected, according to the inclusion criteria in Table 1.

The following databases will be searched: MEDLINE, EMBASE, PsychInfo, CAB Abstracts, PEDro, PILOTS, Zetoc, Science Citation Index, Social Sciences Citation Index, Linguistics and Language Behavior Abstracts, Periodicals Index Online, Applied Social Sciences Index and Abstracts (ASSIA), ERIC, AMED, CINHAI, Cochrane Central Register of Controlled Trials (CENTRAL) and Database of Abstracts of Reviews of Effects (DARE) for primary studies. Grey literature searching is important because of the possibility that effect size estimates may have been overestimated due to selective reporting bias and unpublished studies are known to be less likely to have statistically significant results compared to published studies.<sup>24</sup> Information on studies in progress and unpublished research or research reported in the grey literature will be sought by searching a range of relevant databases including the Inside Conferences, Systems for Information in Grey Literature (OpenSIGLE), Dissertation Abstracts, Current Controlled Trials database and Clinical Trials.gov. Cochrane Database of Systematic Reviews (CDSR), Health Technology Assessment Database (HTA) and the Campbell Library will be searched for systematic reviews and economic evaluations. In addition, Internet searches will also be carried out using specialist search gateways (such as OMNI: <http://www.omni.ac.uk/>), general search engines (such as Google: <http://www.google.co.uk/>) and meta-search engines (such as Copernic: <http://www.copernic.com/>).

Citations will be selected for inclusion in each review in a two-stage process using the criteria in Table 1 by one reviewer with a random 10% of citations independently checked by a second reviewer. Copies of full manuscripts of all citations that are likely to meet the selection criteria will be obtained. Two reviewers will then independently select studies that meet the predefined criteria. Disagreements will be resolved by consensus and/or arbitration involving a third reviewer. Authors of conference abstracts will be contacted for fully published articles. If an abstract only is available, results will be in an appendix. Once the final sample for the systematic review has been identified, each paper will be tracked in Science Citation Index and titles screened independently to identify sister papers of these documents.

## Definition of LTCs

There is no definitive list of LTCs and the potential range of diseases of interest is both



1  
2  
3 extensive and diverse. For the purposes of this review we will adopt the UK Department  
4 of Health definition of a LTC: "Long term conditions are those conditions that cannot, at  
5 present, be cured, but can be controlled by medication and other therapies. They include  
6 diabetes, asthma, and chronic obstructive pulmonary disease." Our working definition of  
7 LTCs also includes mental health problems, including eating disorders, and chronic  
8 infections such as HIV. All cancer studies will be included because previous reviewing  
9 experience has shown that patients may receive palliative care for prolonged periods  
10 and terminally ill patients in hospices may still be receiving active treatment. Thus the  
11 distinction between active treatment and palliation may become difficult to distinguish  
12 and furthermore disease trajectories are not always predictable. There is a debate  
13 around whether obesity in the absence of any co-morbidity is a disease<sup>25</sup> and we will  
14 exclude studies in people with uncomplicated overweight and obesity We will also  
15 evaluate addictive conditions (alcohol, smoking, illegal drugs, legal drugs) and learning  
16 disability because the results would be useful to the NHS, although these may not meet  
17 the current definition of LTCs. We have excluded the following:

- 22 •personality traits such as alexithymia, body dissatisfaction
- 23 •people who have undergone stressful life events such as bereavement, domestic  
24 violence, child sex abuse
- 25 •people found to be at increased risk of developing a LTC

### 28 **Systematic review of effectiveness**

29  
30  
31 Studies' findings will be extracted in duplicate using pre-designed and piloted data  
32 extraction forms, based on those previously developed. Any disagreements will be  
33 resolved by consensus and/or arbitration involving a third reviewer. Missing information  
34 will be obtained from investigators if it is crucial to subsequent analysis. To avoid  
35 introducing bias, unpublished information will be coded in the same fashion as published  
36 information. In addition to using multiple coders to ensure the reproducibility of the  
37 overview, sensitivity analyses around important or questionable judgments regarding the  
38 inclusion or exclusion of studies, the validity assessments and data extraction will be  
39 performed.

40  
41  
42  
43 Quality of selected studies will be assessed based on accepted contemporary standards  
44 such as the Newcastle Ottawa scale for cohort and case control studies<sup>26</sup> and GRADE.  
45 The GRADE methodology<sup>27-29</sup> will guide us when assessing the quality of the evidence  
46 overall and summarising the results.<sup>30, 31</sup>

47  
48  
49 Meta-analyses will be conducted using standard software packages such as STATA. A  
50 special problem that we are likely to face is very little RCT evidence, which is why  
51 observational studies will be included. Separate analyses will be performed on  
52 randomised and non-randomised data. Any heterogeneity of results between studies will  
53 be statistically assessed using  $I^2$  and graphically assessed, including use of funnel plots.  
54 We will explore causes of the heterogeneity and proceed to perform meta-analysis if  
55 appropriate.<sup>31</sup> To explore causes of heterogeneity subgroup analyses will be planned a

1  
2  
3 *priori* to see whether variations in clinical factors e.g. populations, interventions,  
4 outcomes or study quality affect the estimation of effects. Individual factors explaining  
5 heterogeneity will also be analysed using meta-regression, to determine their unique  
6 contribution to the heterogeneity, if possible.<sup>32</sup> Conclusions regarding the typical  
7 estimate of an effect size of the intervention will be interpreted cautiously if there is  
8 significant heterogeneity. If necessary, we will use indirect comparisons to inform the  
9 economic model.  
10  
11

### 12 **Health economic evaluation**

13  
14  
15 This is a broad term to describe a variety of approaches that can be used to illustrate the  
16 economic consequences of a therapeutic strategy. In a project such as this, it is  
17 important to be flexible with regards to planning an economic evaluation because the  
18 depth of complexity of any economic modelling, for example, will be driven partially by  
19 the data available. If no health-related quality of life information is found it may not be  
20 appropriate to state in advance that a cost utility analysis will be conducted. However,  
21 value for money is an important consideration in the current economic climate so any  
22 information on costs and cost effectiveness compared to an appropriate comparator will  
23 be presented. If any economic evaluations were found, they will be evaluated for quality  
24 using the NICE Reference Case and for publication standards using CHEERS  
25 statement.<sup>33</sup>  
26  
27  
28

29  
30 If there are well-powered RCTs with homogenous outcomes in the same disease area or  
31 areas, we will then associate improvements in outcomes with gains in health related  
32 quality of life where possible. This may include use of decision-analytic modelling either  
33 by using an existing disease-specific model available in the literature or by constructing  
34 a de-novo model. The aim will be to estimate the incremental cost-effectiveness of  
35 incorporating therapeutic writing into the currently recommended NHS treatment  
36 regimen for the particular disease area. Results will be presented in terms of cost per  
37 quality adjusted life year (QALY) gained, with the uncertainty in both the RCT evidence  
38 and the modelling incorporated. If the information in the literature found in the systematic  
39 review is not sufficient then we will carry out more general forms of economic evaluation  
40 such as cost-consequences or cost minimisation analyses. All economic evaluations will  
41 follow the reference case used by the National Institute for Health and Clinical  
42 Excellence (NICE) as far as the available evidence permits, for example discounting  
43 costs and benefits at 3% per annum, and using the perspective of the NHS and social  
44 services.  
45  
46  
47  
48

49 The economic modelling component of the work is likely to be challenging. Some of  
50 these challenges are likely to include the following: (i) the intervention is likely to affect a  
51 wide range of disease areas and clinical outcomes, which cannot be captured in a single  
52 disease model; (ii) there may be a need to combine evidence that is heterogeneous in  
53 terms of quality (mixture of observational studies, trials and qualitative studies) as well  
54 as the nature of the intervention and population studied; (iii) there may not be sufficient  
55  
56  
57  
58  
59  
60

1  
2  
3 disease natural history and quality of life information in the literature to conduct a cost-  
4 utility analysis to the specifications of the NICE reference case.  
5

6  
7 To address these challenges, we have deliberately left the precise nature of the health  
8 economic evaluation open ended until after the literature reviews are completed.  
9 Whenever possible, we will conduct a “gold standard” cost-utility analysis, using a  
10 decision analytic model to examine the impact of the intervention on disease  
11 progression, with its parameters informed by a synthesis of the highest quality RCT  
12 evidence, and with outcomes presented in terms of costs per QALY. However, we  
13 envision that the opportunities for this kind of analysis are likely to be slim. In most  
14 cases, we will make the most of any evidence available, for instance conducting cost-  
15 consequences analyses if there is no quality of life information or if there are a range of  
16 different outcomes that cannot be captured within a single model. Such analyses will still  
17 provide useful information to guide decision making, and will also highlight the gaps in  
18 evidence that can be addressed by future studies.  
19  
20  
21

## 22 **Realist review**

23  
24 The realist review will cover the papers in the above sampling frame (Table 1) along with  
25 “sister papers” i.e. any qualitative or mixed-method studies linked to these index papers,  
26 but published as a separate paper. The review team will begin by attempting to develop  
27 a ‘generic’ initial programme theory for therapeutic writing. That is, to build a model that  
28 tries to explain how therapeutic writing is meant to go about producing its potential  
29 benefits. When analysing the findings from the effectiveness review we will use  
30 interpretive cross-case comparison to understand and explain how and why observed  
31 outcomes have occurred in the studies included in our review. In other words we will  
32 compare interventions where therapeutic writing has been ‘successful’ against those  
33 which have not; to try to understand how context has (or has not) influenced the reported  
34 findings. Specifically, when analysing the findings from the effectiveness review we will  
35 be using a realist logic that seeks to construct context-mechanism-outcome  
36 configurations (CMOCs) for the findings in the ‘successful’ and ‘less successful’  
37 therapeutic writing interventions. The purpose of this process is to construct for each  
38 finding (outcome) an explanation of what has caused the outcome to occur (mechanism)  
39 and the conditions (context) that have triggered this putative mechanism. If necessary,  
40 we will seek to iteratively develop one or more explanatory theories to account for these  
41 CMOCs. An important process is to build an understanding of how the CMOCs we have  
42 constructed fit in with our initial programme theory. In other words, for any one CMOC,  
43 how does it (if at all) explain how therapeutic writing is meant to go about producing its  
44 potential benefits? For example, does the CMOC’s tell us anything about how we might  
45 need to refine our initial programme theory? Thus throughout the realist review, we  
46 move iteratively between the analysis of particular examples, a refinement of the over-  
47 arching programme theory, and (if necessary) further iterative searching for data to test  
48 particular theories or sub-theories. The pursuit of rigour in realist research reflects  
49 principles usually seen in qualitative research, although it may draw on qualitative,  
50 quantitative or mixed methods. Much rests on achieving immersion (i.e. reading and  
51  
52  
53  
54  
55  
56  
57  
58  
59  
60

10

1  
2  
3 re-reading papers to really understand what was done and why), thinking reflexively  
4 about findings, developing theory iteratively as emerging data are analysed, seeking  
5 disconfirming cases and alternative explanations, and defending one's interpretations to  
6 researchers within and outside one's own team.<sup>34</sup>  
7  
8

9 Two co-applicants (TG & GW) have recently led an international project, the RAMESES  
10 study, which has developed guidance for realist review. This involved a systematically  
11 recruited group of international experts who have produced methodological guidance  
12 and publication standards ([www.ramesesproject.org](http://www.ramesesproject.org)). We will ensure that the new  
13 RAMESES standards (which we anticipate will become the gold standard for realist  
14 review internationally) are strictly followed in the realist component of this study.  
15  
16

### 17 **Narrative synthesis of systematic review of effectiveness, realist review and** 18 **health economic evaluation** 19

20  
21 There are two published examples of combining conventional systematic review  
22 methods with realist review methods. One addressed the question 'What is the impact of  
23 school feeding programmes on growth and educational achievement in deprived  
24 children, and what explains variation in the findings across studies?'.<sup>35, 36</sup> The other  
25 addressed the question 'What are the components and statistical properties of different  
26 risk scores for type 2 diabetes, and what explains whether and how these scores were  
27 used?'.<sup>34</sup> This review considered longitudinal and cross-sectional population cohort  
28 studies and papers describing case studies of attempts to implement the risk score. The  
29 therapeutic writing will use a similar approach to these two projects, using joining text  
30 and commentary ('narrative synthesis'). This 'narrative summary' or 'narrative synthesis'  
31 is a very well established and robust way of linking two sets of review findings, especially  
32 when those sets of findings are philosophically incommensurable and/or address  
33 different research questions within a single study (hence do not lend themselves to a  
34 'technical' approach to combining findings).<sup>37, 38</sup>  
35  
36  
37  
38

39 We will also be integrating the results of the economic evaluation with the realist and  
40 conventional systematic reviews. Combining economic evaluation with systematic  
41 reviews is very commonly done for HTA reports so needs no further explanation here.  
42 However, what is frequently not discussed is how the clinical context is combined with  
43 the economic evaluation to enable the HTA report to have clinical credibility. This is  
44 frequently done by ensuring that the clinicians on the project are present when  
45 discussing the economic model, fully understand it and can appreciate the implications  
46 of the assumptions made. Their insights into the patient experience often result in the  
47 structure of the model needing to be changed, or different numerical inputs being used  
48 to reflect clinical reality. In our project we will be fully involving patient representatives  
49 and therapeutic writing experts in all aspects of the project.  
50  
51  
52

53  
54 The reviews are currently underway and this 18 month project will be complete in  
55 summer 2014 with per reviewed results available by the end of 2014.  
56  
57  
58  
59  
60

1  
2  
3 The reviews are currently underway and this 18 month project will be complete in  
4 Summer 2014 (see Figure 1 - Gantt chart) with peer reviewed results available by the  
5 end of 2014.  
6  
7  
8  
9  
10  
11  
12  
13  
14  
15  
16  
17  
18  
19  
20  
21  
22  
23  
24  
25  
26  
27  
28  
29  
30  
31  
32  
33  
34  
35  
36  
37  
38  
39  
40  
41  
42  
43  
44  
45  
46  
47  
48  
49  
50  
51  
52  
53  
54  
55  
56  
57  
58  
59  
60

For peer review only

## Figure legend

Figure 1: **Timetable**

## Contributorship

All authors have contributed to writing of this protocol.

## Competing Interests

None

## Funding

None

## References

1. Pearson L. The Use of Written Communications in Psychotherapy. Springfield, IL: Charles C Thomas; 1965.
2. Jolly M. What I never wanted to tell you: therapeutic letter writing in cultural context. *J Med Humanit.* 2011; **32**(1): 47-59.
3. Pennebaker JW, Beall SK. Confronting a traumatic event: toward an understanding of inhibition and disease. *J Abnorm Psychol.* 1986; **95**(3): 274-81.
4. Nicholls S. Beyond expressive writing: Evolving models of developmental creative writing. *Journal of Health Psychology.* 2009; **14**(2): 171-80.
5. Baker JR, Moore SM. Blogging as a social tool: A psychosocial examination of the effects of blogging. *CyberPsychology & Behavior.* 2008; **11**(6): 747-9.
6. Bolton G, Field V, Thompson K, foreword by Morrison B. *Writing Works: a Resource Handbook for Therapeutic Workshops and Activities.* London and Philadelphia; 2006.
7. Meads CA. *Emotional disclosure (expressive writing) and health.* Saarbrücken:



1  
2  
3 VDM-Verlag; 2009.  
4

5  
6 8.Meads C, Nouwen A. Does emotional disclosure have any effects? A systematic  
7 review of the literature with meta-analyses. *International Journal of Technology*  
8 *Assessment in Health Care*. 2005; **21**(02): 153-64.  
9

10  
11 9.Meads C, Sheffield D. Issues Regarding Systematic Review Reports and Trial  
12 Heterogeneity. *The Journal of Nervous and Mental Disease*. 2005; **193**(6): 424-5.  
13

14  
15 10.Ogden J. *Health psychology: A textbook*. Maidenhead, BRK, England: Open  
16 University Press; 1996.  
17

18  
19 11.Greenhalgh T. Writing as therapy. Effects on immune mediated illness need  
20 substantiation in independent studies. *BMJ*. 1999; **319**(7205): 270-1.  
21

22  
23 12.Smyth JM. Written emotional expression: effect sizes, outcome types, and  
24 moderating variables. *J Consult Clin Psychol*. 1998; **66**(1): 174-84.  
25

26  
27 13.Frisina PG, Borod JC, Lepore SJ. A meta-analysis of the effects of written  
28 emotional disclosure on the health outcomes of clinical populations. *J Nerv Ment Dis*.  
29 2004; **192**(9): 629-34.  
30

31  
32 14.Frattaroli J. Experimental disclosure and its moderators: a meta-analysis. *Psychol*  
33 *Bull*. 2006; **132**(6): 823-65.  
34

35  
36 15.Harris AH. Does expressive writing reduce health care utilization? A meta-  
37 analysis of randomized trials. *J Consult Clin Psychol*. 2006; **74**(2): 243-52.  
38

39  
40 16.Theadom A, Smith HE, Yorke J, Hankins M, Apfelbacher CJ, Jones CJ, et al.  
41 *Written emotional disclosure for asthma*  
42 *Cochrane Database of Systematic Reviews*  
43 2009; (2): Art. No.: CD007676. DOI:10.1002/14651858.CD007676  
44

45  
46 17.Yach D, Hawkes C, Gould CL, et al. The global burden of chronic diseases:  
47 overcoming impediments to prevention and control. *JAMA : the journal of the*  
48 *American Medical Association*. 2004; **291**(21): 2616-22.  
49

50  
51 18.Department of Health U. Ten things you need to know about long term conditions.  
52 2011 [cited March 2011]; Available from:  
53 [http://www.dh.gov.uk/en/Healthcare/Longtermconditions/tenthingsyouneedtoknow/in](http://www.dh.gov.uk/en/Healthcare/Longtermconditions/tenthingsyouneedtoknow/index.htm)  
54 [dex.htm](http://www.dh.gov.uk/en/Healthcare/Longtermconditions/tenthingsyouneedtoknow/index.htm)  
55

56  
57 19.Barnett K, Mercer SW, Norbury M, et al. Epidemiology of multimorbidity and  
58 implications for health care, research, and medical education: a cross-sectional  
59  
60



1  
2  
3 study. *Lancet*. 2012; **380**(9836): 37-43.  
4

5  
6 20.Ouwens M, Wollersheim H, Hermens R, et al. Integrated care programmes for  
7 chronically ill patients: a review of systematic reviews. *International journal for quality*  
8 *in health care : journal of the International Society for Quality in Health Care / ISQua*.  
9 2005; **17**(2): 141-6.  
10

11  
12 21.Pawson R, Greenhalgh T, Harvey G, et al. Realist review--a new method of  
13 systematic review designed for complex policy interventions. *J Health Serv Res*  
14 *Policy*. 2005; **10 Suppl 1**: 21-34.  
15

16  
17 22.Astbury B, Leeuw FL. Unpacking Black Boxes: Mechanisms and Theory Building  
18 in Evaluation. *American Journal of Evaluation*. 2010; **31**(3): 363-81.  
19

20  
21 23.Berwick D. The Science of Improvement. *JAMA : the journal of the American*  
22 *Medical Association*. 2008; **299**(10): 1182-84.  
23

24  
25 24.Egger MD, K. Davey-Smith, G. . Problems and limitations in conducting  
26 systematic reviews. In: Egger MD-S, G. Altman, D.G., editor. *Systematic reviews in*  
27 *healthcare, Meta-analysis in context*. London: BMJ Books; 2001.  
28

29  
30 25.Heshka S, Allison DB. Is obesity a disease? *Int J Obes Relat Metab Disord*. 2001;  
31 **25**(10): 1401-4.  
32

33  
34 26.Wilson A, Lissauer D, Thangaratinam S, et al. A comparison of clinical officers  
35 with medical doctors on outcomes of caesarean section in the developing world:  
36 meta-analysis of controlled studies. *BMJ*. **342**: d2600.  
37

38  
39 27.Guyatt G, Oxman A, Vist G, et al. GRADE: an emerging consensus on rating  
40 quality of evidence and strength of recommendations. *BMJ*. 2008; **336**: 924-6.  
41

42  
43 28.Guyatt G, Oxman A, Vist G, et al. GRADE: What is "quality of evidence" and why  
44 is it important to clinicians? . *BMJ*. 2008; (336): 995-8.  
45

46  
47 29.Guyatt G, Oxman A, GE V, R K. Rating quality of evidence and strength of  
48 recommendations GRADE: an emerging consensus on rating quality of evidence  
49 and strength of recommendations *BMJ*. 2008; **336**: 924-6.  
50

51  
52 30.Latthe PM, Foon R, Khan K. Nonsurgical treatment of stress urinary incontinence  
53 (SUI): grading of evidence in systematic reviews. *Bjog*. 2008; **115**(4): 435-44.  
54

55  
56 31.Khan KS, Kunz R, Kleijnen J, et al. *Systematic Reviews to Support Evidence-*  
57 *Based Medicine: How to Review and Apply Findings of Healthcare Research* London  
58 *Royal Society of Medicine Press*; 2005.  
59  
60

1  
2  
3 32.Sterne JAC, Egger M. Regression methods to detect publication and other bias in  
4 meta-analysis. . Chichester: Wiley; 2005.  
5

6  
7 33.Husereau D, Drummond M, Petrou S, et al. Consolidated Health Economic  
8 Evaluation Reporting Standards (CHEERS) statement. BMJ. 2013; **346:f1049**.  
9

10  
11 34.Noble D, Mathur R, Dent T, et al. A statistical feast but an impact famine:  
12 Systematic review of risk scores for type 2 diabetes. BMJ. 2011; **343:d7163**.  
13

14  
15 35.Greenhalgh T, Kristjansson E, Robinson V. Realist review to understand the  
16 efficacy of school feeding programmes. BMJ. 2007; **335(7625)**: 858-61.  
17

18  
19 36.Kristjansson E, Robinson V, Petticrew M, et al. School feeding for improving the  
20 physical and psychosocial health of disadvantaged elementary school children. The  
21 Cochrane database of systematic reviews. 2007; (1): CD004676.  
22

23  
24 37.Dixon-Woods M, Agarwal S, Jones D, et al. Synthesising qualitative and  
25 quantitative evidence: a review of possible methods. J Health Serv Res Policy. 2005;  
26 **10(1)**: 45-53.  
27

28  
29 38.Popay Jea. Guidance on the Conduct of Narrative Synthesis in Systematic  
30 Reviews. Available in full text on  
31 <http://citeseerx.ist.psu.edu/viewdoc/download?doi=10.111783100&rep=rep1&type=pdf>.  
32 ESRC 2006.  
33  
34  
35  
36  
37  
38  
39  
40  
41  
42  
43  
44  
45  
46  
47  
48  
49  
50  
51  
52  
53  
54  
55  
56  
57  
58  
59  
60

1  
2  
3  
4  
5  
6  
7  
8  
9  
10  
11  
12  
13  
14  
15  
16  
17  
18  
19  
20  
21  
22  
23  
24  
25  
26  
27  
28  
29  
30  
31  
32  
33  
34  
35  
36  
37  
38  
39  
40  
41  
42  
43  
44  
45  
46  
47  
48  
49  
50  
51  
52  
53  
54  
55  
56  
57  
58  
59  
60

Months	1	2	3	4	5	6	7	8	9	10	11	12	13	14	15	16	17	18
Protocol development	X																	
Protocol peer review	X																	
Study selection		X	X	X	X													
Effectiveness review		X	X	X	X	X	X	X										
Realist Review						X	X	X	X	X	X							
Evidence synthesis									X	X	X	X						
Economic modelling									X	X	X	X	X	X				
Report production												X	X	X	X	X	X	X
Internal peer review																	X	X

224x90mm (300 x 300 DPI)

Peer review only