BMJ Open

A Protocol for a scoping review of how people with ME/CFS use the internet.

Journal:	*BMJ Open*
Manuscript ID	bmjopen-2023-076904
Article Type:	Protocol
Date Submitted by the Author:	20-Jun-2023
Complete List of Authors:	Shortland, Diane; University of Birmingham Edgbaston Campus, school of medicine and dentistry; Fazil, Qulsom; The University of Birmingham Hallett, Nutmeg; University of Birmingham, School of Nursing and Midwifery Lavis, Anna; University of Birmingham, Institute of Applied Health Research
Keywords:	Fatigue, GENERAL MEDICINE (see Internal Medicine), Health Education, Protocols & guidelines < HEALTH SERVICES ADMINISTRATION & MANAGEMENT, MEDICAL EDUCATION & TRAINING

SCOPING REVIEW PROTOCOL for publication

A protocol for a scoping review of how people with ME/CFS use the internet.

Authors:

Lead researcher - Diane Shortland PhD. University of Birmingham: DLS760@student.bham.ac.uk

Supervisor – Dr. Qulsom Fazil, University of Birmingham: Q.A.Fazil@bham.ac.uk

Supervisor – Dr Anna Lavis, University of Birmingham: A.C.Lavis@bham.ac.uk

Supervisor – Dr Nutmeg Hallett, University of Birmingham: N.N.Hallett@bham.ac.uk

Abstract

Introduction:

Myalgic Encephalomyelitis (ME) is a chronic neurological illness affecting many bodily systems, commonly the nervous and immune systems. Also known as Chronic Fatigue Syndrome (CFS), key symptoms are extreme fatigue, post-exertional malaise, cognitive problems and sleep disturbance (Jason et al 2015).[1] With reported higher levels of online activity for people with ME/CFS than other patient groups (Westerby 2013 cited in Ytre-Arne 2016), [2] it's crucial to gain more knowledge of usage characteristics and experience of online use, and it's integration into everyday life. This scoping review protocol details the proposed methods for gaining insight into this little known phenomenon.

Methods & Analysis:

This review uses the methodological framework for conducting a scoping review by Arksey & O'Malley, [3] with further guidance by Levac, Colquhoun & O'Brien, [4] and the Joanna Briggs Institute. [5] It also refers to the PRISMA-Preporting guidelines. [6] The following bibliographic databases will be searched: Embase, Medline, PsychINFO, Cinahl, AMED, and ASSIA, plus Web of Science, ProQuest Dissertations & Theses Global, Scopus, and Google Scholar for grey literature. Reference lists of included papers will be studied. Two reviewers will independently screen title-abstracts, and then full text of studies against inclusion criteria. Remaining studies will be quality assessed using appropriate critical appraisal tools. Findings will be charted and mapped to gain in-depth knowledge of the use of the internet in people with ME/CFS.

Ethics and Dissemination:

The findings from this review will be disseminated through peer-reviewed publication and a report for leading charities of ME/CFS. The review will collect secondary data only and therefore does not need ethical approval.

Article Summary:

Strengths & Limitations of this study

- To our knowledge this is the first scoping review to map out the online usage and experience of people with ME/CFS.
- A strength of the review will be the rigorous and transparent approach based on a solid methodological framework and the Preferred Reporting Items for Systematic reviews and Meta-analysis extension for Scoping Reviews checklist (PRISMA-ScR).
- The quality of the scoping review will be enhanced by the use of a second reviewer for study selection and charting of results.
- Eligible studies will be quality assessed in accordance with their study design.
- The review is confined to English language which may exclude other language studies that may contain valuable data.

Introduction

Myalgic encephalomyelitis (ME) – meaning inflammation of the brain and spinal cord – is a long term chronic neurological illness, often fluctuating in nature, that causes many symptoms affecting many bodily systems, most commonly the nervous and immune systems (Action for ME 2022). [7] Since 1988, the illness has also been known as Chronic Fatigue Syndrome (CFS). Many publications and researchers use both ME and CFS terms interchangeably and so we have operationalised both names as suitable for inclusion in this review. It is worth noting however, that debate exists in defining and classifying the two and there is evidence of distinct historical trajectories of ME and CFS, with distinguishing features of diagnosis and as such, including both names together could create additional issues surrounding the illness (Grue 2014). [8] Studies have reported however, that despite this debate, certain core symptoms of the illness do appear to be consistently present

across both classifications, namely extreme fatigue, post-exertional malaise, neuro-cognitive difficulties and sleep disturbance (Jason et al 2015). [1]

People with ME/CFS are significantly more impaired in both physical and social functioning than other long term illnesses (Kingod et al 2018; Hvidberg et al 2015; Pendergrast et al 2016).[9, 10, 11] The illness has a negative impact on people's relationships and social networks, with suicide ideation endorsed more frequently in those experiencing unsupportive interactions and social distancing (Clarke & James 2003; McManimen et al 2018). [12, 13] Due to the contested nature of the condition (there is yet no available biomarker for the condition and its diagnosis is therefore subjective, raising a debate over the decades between the medical and psychological realms as to its aetiology and treatment), legitimacy of the illness is often questioned in immediate social support networks, causing additional stress (Harris et al 2016; McManimen et al 2018). [14, 13] As Bowling (2005) states, [15] lack of social support, participation and contact is associated with increased mortality risk and delayed recovery from disease. In a survey by Action for ME (2019), [16] 94% of participants had stopped or reduced social contact, and up to 97% of the 4038 participants said they felt socially isolated because of their condition. Patients describe feeling overwhelmed and let down when disbelieved. When seeking help was unsatisfactory, sufferers responded to this by taking more responsibility for their illness management via 'self-help' tactics (Edwards, Thompson & Blair 2007). [17]

There are reported higher levels of online activity among people with ME than other patient groups (Westerby 2013 cited in Ytre-Arne 2016). [2] Online peer-to-peer support in the form of interactive websites and social media, is now highly valued in chronic illness as a way to connect to others who share the same illness (Van der Eijk et al 2013; Lian & Nettleton 2015). [18, 19] Transcending geographic boundaries, the internet is convenient to those with limited mobility (Lasker et al 2006; Eichhorn 2008). [20, 21] Online communities provide support for people with long term illness with a growing reliance on social media in patients experiencing social isolation and who fear marginalisation because of their illness (Loane & D'Alessandro 2014; Perkins, Coulson & Davies 2020). [22, 23] It also offers support to people otherwise limited by disability or stigma when accessing support offline (Drentea & Moren-Cross 2005). [24]

Uncertainty surrounding illness appears to be a driving factor for internet use (Conrad & Stults 2010) [25] with internet itself being an increasingly public experience as people share personal information and interact in public spheres (Conrad, Bandini & Vasquez 2016). [26] As Beck, Gurion & Sheva (2004) state, [27] "users of the world wide web are no longer passive audiences of data consumers… but are active participants controlling the content of the information. They shape the quality of the data… (facilitating) the expression of emotions (output) and the input of emotional messages, thus developing and reinforcing important social ties between users, forming a system of relationships similar to ties of family and friendship" (p.46). Receiving problem-focused and emotion-focused support from others aids coping and thus becomes a primary driver of willingness to offer such support to others (Lin et al 2015). [28] Online users describe 'social overload' however, where people feel they're giving too much social support to others and experience online group exhaustion (Maier et al 2015). [29]

So how does online usage interplay with the 'real world', particularly for ME patients who are often housebound due to the chronicity of their condition? In general, there is a "sharp distinction between concepts from the virtual world online and the 'real world' offline" but "technology enters and is gradually integrated into people's daily lives" (Lie & Sorensen 1996 cited in Beck, Gurion & Sheva 2004), [27] by a process of 'domestication' where people adapt new technologies and bring them into their home, transferring elements of the physical world into the virtual environment, merging the two worlds and creating a much broader definition of reality. Understanding how people with ME use the internet to aid their illness management and enhance their experience of daily life, is crucial in gaining insight into how informational and social support is found and utilised online and offline. It will shed light on how people's overall support networks are created and maintained, as well as identify the benefits of such illness behaviour.

Kingod et al (2016) [30] studied how people with chronic conditions experience online peer-to-peer social support and its influence in everyday life, in a systematic review of 13 papers, but none of them covered the illness ME. They found four main themes: identity, social support and connectivity, experiential knowledge that both strengthened social ties and

supported offline ties, and collective voice and mobilisation. Allen et al (2020) [31] also looked at chronic illnesses which included ME in a primary study of 30 people across varying conditions. They found that online support was sought in response to deficits in offline support; it was used to assist offline ties as well as substitute offline support.

Both Kingod et al (2016) [30] and Allen et al (2020) [31] stressed the need for further research into understanding the boundaries of online and offline social dimensions and relevance in daily life; how the role of online ties serve within personal networks. Essentially how do people decide who to turn to now they have greater choice in who contributes to their everyday illness management and coping? Having further advancement of knowledge in this area will inform healthcare practice social support initiatives and aim to improve services to those housebound with ME/CFS. It will also gain knowledge into the lack of support present in the home life of people with ME/CFS.

Initial searches revealed a lack of studies conducted in this area that focused on ME/CFS. Studies on other chronic illnesses have an element of transferability of their findings to ME/CFS and several papers were found here highlighting a topic worthy of attention. Preference for online support over offline support was highlighted in cancer and diabetes patients (Chung 2013), [32] with a lack of real world social support predicting active participation in online groups (Cummings, Sproul & Kiesler 2002). [33] The benefits of using social media in health communication include interaction with others, the availability of shared knowledge, widened access to health information, social and emotional support, and empowerment in their healthcare process (Moorhead et al 2013; Huang, Chengalur-Smith & Pinsonneault 2014). [34, 35] Investigating the perceived impact of online participation, Morehouse et al (2021) [36] found people gained a sense of belonging, validation and supportive friendships, decreasing feelings of depression and increasing quality of life. As much as 75% of a sample studied by Kummervold et al (2009) [37] found it easier to discuss personal problems online than face-to-face. Virtual communities appear to play an important role in meeting patients social needs; sense of community is positively associated with cancer patients well-being in areas of personal relations and personal growth (Leimeister et al 2008). [38]

Caplan (2003) [39] found that depressed people may develop preferences for online social interaction but this in turn leads to negative outcomes associated with internet use, and Allen et al (2018) [40] concluded that internet use may indicate an avoidance or absence of offline support. Moreover, Chung (2013) [32] found those dissatisfied with their offline relationships were more likely to develop preference for online social interaction and this can become problematic when excessive reliance increases disengagement from offline interaction. Findings are largely positive in that using the internet appears to improve everyday life, however there are studies that highlight negative impacts. Given that ME/CFS is a contested illness with legitimacy issues and increased stigma, will data collected from the ME/CFS community produce similar or different themes to other conditions?

This scoping review will aim to explore the online usage characteristics of ME/CFS patients and inter-relatedness within everyday life of their online and offline worlds. To prevent unnecessary duplication, a preliminary search for existing scoping and systematic reviews on the subject was carried out in May 2022. To our knowledge, a comprehensive synthesis of related studies on ME/CFS in this field remains absent.

Methods/Design

In order to capture the broadest scope of literature on the topic of online usage in people with ME/CFS, we decided to use a scoping review method. A scoping review is ideal for mapping out the scope or coverage of a body of literature on a given topic when the emerging evidence is still unclear and more specific questions cannot presently be posed (Peters et al 2020). [5] They give a clear indication of available literature, regardless of study design, and an overview of its focus, identifying characteristics of studies to provide an overall picture of current evidence (Munn et al 2018). [41] To map our field of study and examine the extent, range and nature of research activity to date, as well as identify any knowledge gaps in research, our protocol was developed using a framework set out by Arksey & O'Malley (2005). [3] We have also incorporated later improvements to this framework by the work of Levac and collegaues (2010), [4] and the Joanna Briggs Institute (2015). [5]

Traditionally a scoping review concerns itself with summary of results and does not evaluate the quality of included studies. Revisions have noted the value of quality assessment for future researchers however (Daudt, Mossel & Scott 2013 p.6) [42] and so we intend to incorporate this into our review. Guidance from the Preferred Reporting Items for Systematic Reviews and Meta-Analyses Extension for Scoping Reviews checklist (PRISMA - ScR) [6] has been consulted to yield greater transparency and reproducibility. Arksey & O'Malley's framework proposes five mandatory stages (outlined below) and a sixth optional stage: consultation with stakeholders. [3] Our current review does not involve this due to the nascent stage of the project. However, the findings of this review will inform a translation of knowledge engagement exercise which will involve consultation with stakeholders.

Stage One: Identifying the Research Question

The research question for this proposed scoping review aims for comprehensiveness and so will be broad to cover the full breadth of evidence in the field. We aim to answer the following question: **How do people with ME/CFS use the internet?** This aim will be achieved by addressing the following objectives:

- Examine the usage characteristics of people with ME/CFS utilising the internet. (What do they do online, when, and for how long?)
- Examine people with ME/CFS' experiences of online usage. (Why do they go online and what do they gain from going online?)
- Examine people with ME/CFS' online usage inter-relating with their offline lives. (e.g. how does using the internet fit alongside offline informational and social support?)

Arksey & O'Malley (2005) saw scoping as an iterative methodological skill and as such it may be appropriate and acceptable to add supplementary questions based on emerging findings during the review process. [3] We may notice other important data that could be useful to extract (Peters et al 2015). [5] Any changes or amendments will be clearly stated and explained.

Stage Two: Identifying relevant studies

Study eligibility:

We will aim to find both published and grey literature studies. Loosely using a PCC (Population, Concept, Context) framework to develop our inclusion criteria, to align with our objectives and research questions, our population will be adults identified as having ME/CFS. Our concept of interest is internet use. We define 'internet use' as the computer network that allows users to connect with other users and content from all over the world (Collins 2022). [44] Online information, content and social support exists through many various technological avenues nowadays. Kaplan & Haenlein (2010) operationalised 'social media' usage in five main categories: collaborative projects, blogs, content communities, social networking sites and virtual worlds. [43] Gaming is also an online social experience now as it is shared live with other users. The context is loosely any available knowledge that involves personal use of the internet and not organised institutionalised treatment agendas.

Search strategy:

Guidance by the Joanna Briggs Institute (Peters et al 2015) [5] recommends a three-stage process to searching the literature of which we have included all advised stages. An initial search of limited databases has been conducted and from analysis of these results, key words and index terms have been identified. A reference librarian was consulted in preparation, and a systematic search plan was formed with search terms incorporating medical subject headings (MeSH) as well as text words combining comprehensive terms for contemporary social media, and Boolean operators 'AND' and 'OR' (Table 1 shows an example search strategy of keywords for Web of Science. This will be adapted to suit individual databases). Since scoping is an iterative process (Arksey & O'Malley 2005), [3] a pilot of searching will take place and terms will be refined if deemed necessary.

Secondly we intend to include extensive electronic searches of the following bibliographic databases (conducted in Aug to November 2022): EMBASE, Medline, Cinahl, PsychINFO, AMED. And ASSIA. Bramer et al (2017) [45] found that optimal searching to ensure a minimum risk of missing studies, should use four key databases: Embase, Medline, Web of Science and Google Scholar. This produced a 98.3% recall of studies. Because online communities in relation to health have been explored across a range of professional, theoretical, sociological, psychological and healthcare settings however, additional databases have also been covered. Thirdly, Grey literature will be searched (during

November 2022) via Scopus, Web of Science, and ProQuest Dissertations & Theses Global. Web searching via Google Scholar will also take place as well as citations and references of key papers searched by hand.

All eligible studies that meet our inclusion criteria (adults over 18 years of age, located anywhere in the world, identified as having ME/CFS, found in English language peer-reviewed primary studies, on internet use) will be saved on an Excel spreadsheet. Our exclusion criteria are children under 18 years of age, those not identified as having ME/CFS, and systematic reviews since their content is already secondary in nature, so analysis would further dilute and potentially bias findings. Consideration was given to restricting studies to a date limitation since the advancement of internet-based platforms such as social media is a relatively new and still growing area. However it is not possible to confidently put a time limit on when such social media support truly began, so doing so would risk losing valuable studies.

Stage Three: Study selection

The primary researcher will run the initial searches, retrieving titles and abstracts, removing duplicates, and saving all files into a suitable data management storage. Two reviewers will go through the title and abstract of each study and screen them to identify studies that meet the inclusion criteria and will document all results in a 'screening' form. Any uncertainty regarding if a study is eligible or not, will be included at this stage to ensure nothing is missed. If multiple papers are found that describe the same data, we will include the paper that describes the most comprehensive findings. By citation chaining, reference lists of included studies will then be examined by the lead reviewer to identify any eligible studies that meet the inclusion criteria and added to the database findings. Forward searching of papers, via Scopus, that have been cited, will also be checked. To further minimise location bias, authors and researchers of studies will be contacted.

Two independent reviewers will then read the full text of all provisionally included studies, to assess further against the inclusion and exclusion criteria. The devised extraction form will be piloted to ensure it is containing all relevant information needed. Studies will be included or excluded against the pre-determined eligibility criteria. Any missing data will

attempt to be found by contacting the study authors for additional information. Any discrepancies will be resolved through consultation with the wider research team. All reviewers will agree on the final list of included studies. A PRISMA-ScR flow diagram following the process of the scoping review will be used to demonstrate the selection process (Figure 1).

Critical appraisal:

Contrary to the methodological framework originally set out by Arksey & O'Malley (2005), [3] we intend to appraise the remaining eligible studies for quality assessment. This will take place after the data extraction of full text studies. Pham et al (2014) reported only 22.38% of studies included an element of quality assessment. [46] McColl et al (2009) argue that the emphasis of a scoping review is on comprehensive coverage and not standard of evidence. [47] More recent refinements to guidelines however, support the use of some form of critical appraisal (Levac et al 2010, Peters et al 2015). [4,5] Brien et al (2010) believe a lack of quality assessment makes results more challenging to interpret and Grant & Booth (2009) believe it limits uptake of findings into policy and practice. [48, 49] Daudt (2013) considers quality assessment a necessary component of any scoping review and encourages the use of validated tools since use of reporting checklists increases transparency of methods and allows the reader to use the research appropriately. [42] Pham et al (2014) also recognises that some form of quality assessment would enable the identification of gaps in the evidence base rather than just where research is lacking. [46]

A quality assessment form will be used to extract relevant data for appraisal. Since it is expected that the majority of studies will be qualitative in nature, we have chosen the Critical Appraisal Skills Programme (CASP 2018). [50] If we identify any mixed methods studies then we will use the Mixed Methods Appraisal Tool (MMAT 2018) (51). If any quantitative data is found we will use a checklist suited to the study design from the selection available at JBI, most likely the Checklist for Analytical Cross Sectional Studies (52). Any discrepancies between reviewers on quality assessment will be discussed with the wider research team. No exclusion of eligible studies will take place as a result of appraisal since such studies can still contain rich and useful qualitative narrative. Poor quality studies will be highlighted and reflected upon within the data summaries. The outcome of each

study assessment, along with all study files will be included in an Excel spreadsheet alongside other data extraction details.

Stage Four: Charting the data

Two independent reviewers will perform a full-text review of provisionally included studies. Piloting of a small sample will take place, in accordance with advice from Levac et al (2010) to ensure agreement is reached on extraction consistency. [4] Charted data extracted and documented in a designed extraction form will include, but not be limited to, the following:

- Article title, authors, year of publication
- Study research aims
- Study design and setting
- Number of participants
- Characteristics of the population
- Study inclusion criteria
- Online usage information
- Data collection and analysis methods
- Study findings/outcome

As previously mentioned the review will take an iterative approach and so the content of extraction can be updated with discussion of the research team. This allows for the variables and themes to be included to best be able to answer the review question and meet its objectives.

Stage Five: Summarising and reporting the results

Levac and colleagues (2010) encourage a rigorous approach to analysis that includes descriptive numerical summary as well as thematic analysis. [4] Using the information extracted, data charting will involve visual summaries as well as narrative that describes the aims of included studies, their areas of focus, online user characteristics and findings to determine how the studies to date inform the current knowledge base. Any quantitative or mixed method studies will be 'qualitized' by extracting data from quantitative or mixed method studies and transforming it into textual descriptions to integrate with qualitative data and form a single summary comprising themes of narrative across the review studies

(Peters et al 2015). [5] Developed in an inductive manner without a set of a priori themes, these scoping study summary methods, in accordance with Braun & Clarke (2006), [53] will enable us to ascertain broad themes of what is known about how people with ME/CFS use the internet, their experience of doing so, and how this fits within their daily lives as per our research objectives. Two reviewers will perform all analysis independently before reaching consensus of themes and any discrepancies will be resolved with the wider research team.

Ethics and Dissemination:

All data generated will be stored on pass-protected computers. The authors will disseminate the findings through submission for publication in a peer-reviewed academic journal and a report will be written for leading charities of ME/CFS. The review itself will only deal with secondary data and therefore ethical approval is not required. Our findings will be used to inform the design of a future study aiming to gain greater knowledge of online social support in people with ME/CFS. Patient and Public Involvement will take place in the dissemination stages of this review and will guide all future research plans.

Patient and Public Involvement

Since this is a protocol only, it does not have any involvement with patients. Any data regarding patient participation is secondary through already published papers.

Discussion

A global comprehensive systematic scoping summary of primary data on internet use in people with ME/CFS, in terms of both usage and characteristics, and in relation to offline daily life, will be conducted to fill a gap in knowledge surrounding this under-researched area. In relation to online worlds, we frequently find ourselves asking research questions that contain complex medical, sociological, and social concepts. This type of research resists easy quantification and by aiming to critically reflect on the material found via this scoping review, we aim to capture the complexity inherent in such questions involving people's experiences (Kingod 2016). [30] In writing up the research findings we will be guided by enhancing transparency in reporting the synthesis of qualitative research (ENTREQ) (Tong et al 2012). [54] Dissemination will be relevant to academic knowledge sharing, charities for

ME/CFS that offer support and online services, as well as healthcare professionals and patients.

[3,783 words]

Keywords:

Myalgic Encephalomyelitis (M.E), Chronic Fatigue Syndrome (CFS), internet use, online usage, peer support.

Correspondence to:

Diane Shortland

University of Birmingham

DLS760@student.bham.ac.uk

07984 944683

Conflict of Interest:

No conflicts of interest were declared.

Funding:

The authors have not declared any specific grants for this research from any funding agency in public, commercial or not-for-profit sectors.

Author Contributions:

Diane Shortland – responsible for all areas of the study from protocol design, screening, analysis, discussion and writing up of study results.

Dr. Qulsom Fazil, Dr. Anna Lavis & Dr. Nutmeg Hallett – contribution to the design of study, drafting and revising of work, final approval and agreement

Contributor acknowledgement:

Michelle Minns-Sykes has contributed to this paper by independently screening papers for the systematic review.

Data Statement: Open access. Study available by contacting lead author.

Table One: Search strategy example for Web of Science.

Figure One: PRISMA Flow Chart of systematic scoping review process

References

[1] Jason, L; Kot, B; Sunnquist, M; Brown, A; Evans, M; Jantke, R; Williams, Y; Furst, J; Vernon, S. (2015) 'Chronic fatigue syndrome and myalgic encephalomyelitis: towards an empirical case definition'. *Health Psychology and Behavioral Medicine*. 3 (1) 82-93.

[2] Ytre-Arne, B. (2016) 'The social media experiences of long-term patients: Illness, identity and participation'. *Nordicom Review*. 37 (1) 57-70.

[3] Arksey, H., O'Malley, L. (2005) 'Scoping studies: towards a methodological framework'. *International Journal of Social Research Methodology*. 8:1 19-32.

[4] Levac, D., Colquhoun, H., O'Brien, K.K (2010) 'Scoping studies: advancing the methodology'. *Implementation Science* 5; 69.

[5] Peters, MDJ., Godfrey, C., McInerney, P., Munn, Z., TRicco, AC., Khalil, H. (2020) 'Scoping Reviews' in: Aromataris, E., Munn, Z (eds) JBI *Manual for Evidence Synthesis*. Available at: https://synthesismanual.jbi.global.

[6] Tricco, A.C., Lillie, E., Zarin, E., O'Brien, K.K., Colquhoun, H., Levac, D., Moher, D., Peters, M.D., Horsley, T., Weeks, T., Hempel, S. (2018) 'PRISMA extension for scoping reviews (PRISMA-ScR): checklist and explanation'. *Ann Intern Med*. 167 (7) 467-473.

[7] Action for ME (2022) *What is M.E.?* Available at: https://www.actionforme.org.uk/get-information/what-is-me/what-does-me-feel-like/ (Accessed 6th June 2022).

[8] Grue, J. (2014) 'A Garden of Forking Paths: A discourse perspective on 'Myalgic Encephalomyelitis' and 'Chronic Fatigue Syndrome'. *Critical Discourse Studies*. 11:1, 35-48.

[] Kingod, C; Bowman, E; Curran, H; Nacul, L; Lacerda, E. (2018) 'Functional Status and Well-Being in People with Myalgic Encephalomyelitis/Chronic Fatigue Syndrome Compared with People with Multiple Sclerosis and Healthy Controls'. *Pharmaco Economics Open*. 2: 381-392.

[10] Hvidberg, F., Brinth, M., Schouborg, L., Olesen, A.V ., Petersen, K.D., Ehlers, L. (2015) 'The health-related quality of life for patients with myalgic encephalomyelitis/chronic fatigue syndrome (ME/CFS)', PLoS One, 10 (7): e0132421.

[11] Pendergrast, T., Brown, A., Sunnquist, M., Jantke, R., Newton, J.L., Strand, E.B., Jason, L.A. (2016) 'Housebound versus non housebound patients with myalgic encephalomyelitis and chronic fatigue syndrome', *Chronic Illness*, 12 (4), pp.292-307

[12] Clarke, J., James, S. (2003) 'The radicalized self: the impact on the self of the contested nature of the diagnosis of chronic fatigue syndrome'. *Social Science & Medicine*. 1387-1395.

[13] McManimen, S., McClellan, D., Stoothoff, J., Jason, L. (2018) 'Effects of unsupportive social interactions, stigma, and symptoms on patients with myalgic encephalomyelitis'. *Journal of Community Psychology*. 46 (8) 959-971.

[14] Harris, K., Band, R., Cooper, H., Macintyre, G., Mejia, A., Wearden, A. (2016) 'Distress in significant others of patients with chronic fatigue syndrome: a systematic review of the literature'. *British Journal of Health Psychology*. 21 (4) 881-893.

[15] [15] Bowling, A. (2005) *Measuring Health: A review of quality of life measurement scales*. Maidenhead: Open University Press.

[16] Action for ME. (2019) '*Five year Big Survey*'. Available at: http://actionforme.org.uk/research-and-campaign/five-year-big-survey/ (Accessed 9th August 2022)

[17] Edwards, C.R., Thompson, A.R., Blair, A. (2007) 'An 'overwhelming illness': women's experiences of learning to live with chronic fatigue syndrome/myalgic encephalomyelitis', *Journal of Health Psychology*, 12 (2), p.203-215

[18] Van der Eijk, M., Faber, M., Aarts, J., Kremer, J., Munneke, M., Bloem, B. (2013) 'Using health communities to deliver patient-centred care to people with chronic conditions'. *Journal of Medical Internet Research*. 15 (6) 115.

[19] Lian, O., Nettleton, S. (2015) 'United we stand": Framing Myalgic Encephalomyelitis in a virtual symbolic community'. *Qualitative Health Research*. 25 (10) 1383-1394.

[20] Lasker, J., Sogolow, E., Sharim, R. (2006)' The role of an online community for people with a rare disease: Content analysis of messages posted on a primary biliary cirrhosis mailing list'.. 7 (1).

[21] Eichhorn, K. (2008) 'Soliciting and providing social support over the internet: An investigation of online eating disorder support groups'. *Journal of Computer Mediated Communication*. 14 (1) 67-78.

[22] Loane, S., D'Alessandro, S. (2014) 'Empowered and knowledgeable health consumers: the impact of online support groups on the doctor-patient relationship'. *Australian Marketing Journal*. 22: 238-245.

[23] Perkins, V., Coulson, N. S., and Davies, E.B. (2020) 'Using Online Support Communities for Tourette Syndrome and TIC Disorders: Online Survey of User's Experiences'. *Journal of Medical Internet Research.* 22 (11): e18099.

[24] Drentea, P., Moren-Cross, J. (2005) 'Social capital and social support on the web: The case of an internet mother site'. *Sociology of health & Illness*. 27: 920-943.

[25] Conrad, P., Stults, C., (2010) 'The Internet and the Experience of Illness' in Bird, C, E., Conrad, P., Fremont, A, M., Timmermans, S., *Handbook of Medical Sociology*, 6th edition. Vanderbilt University Press, p.179

[26] Conrad, P., Bandini, J., Vasquez, A. (2016) 'Illness and the internet: from private to public experience'. *Health*. 20 (1) 22-32.

[27] Beck, L, S., Gurion, B., Sheva, Beer, (2004) 'Internet Ethnography: Online and Offline', *International Journal of Qualitative Methods*, 3 (2) p.45-51.

[28] Lin, T.C., Hsu, J.S.C., Cheng, H.L., Chiu, C.M. (2015) 'Exploring the relationship between receiving and offering online social support: A dual social support model', *Information & Management*, 52 (3), p.371-383

[29] Maier, C., Laumer, S., Eckhardt, A., Weitzel, T., (2015) 'Giving too much social support: social overload on social networking sites', *European Journal of Information Systems*, 24 (5).

[30] Kingod, N., Cleal, B., Wahlberg, A., Husted, G. (2016) 'Online peer-to-peer communities in the daily lives of people with Chronic Illness: A qualitative systematic review'. *Qualitative Health Research*. 27 (1) 89-99.

[31] Allen, C., Vassilev, I., Kennedy, A., Rogers, A. (2020) 'The work and relatedness of ties mediated online in supporting long-term condition self-management', *Sociology of Health & Illness*, 42 (3) p.579-595.

[32] Chung, J. (2013) 'Social interaction in online support groups: preference for online social interaction over offline social interaction'. *Computers in Human Behaviour*. 29: 1408-1414.

[33] Cummings, J.N., Sproull, L., Kiesler, S.B. (2002) 'Beyond hearing: Where real-world and online support meet', *Group dynamics*, 6 (1), p.78-88

[34] Moorhead, S.A., Hazlett, D.E., Harrison, L., Carroll, J.K., Irwin, A., Hoving, C. (2013) 'A New Dimension of Health Care: Systematic Review of the Uses, Benefits, and Limitations of Social Media for Health Communication', *Journal of medical Internet research*, 15 (4), p.85

[35] Huang, K-Y., Chengalur-Smith, I., Ran, W. (2014) 'Not just for support: Companionship activities in healthcare virtual support communities', *Communications of the Association for Information Systems*, 34 (1), p.561-594

[36] Morehouse, S., Schaible, K., Williams, O., Herlache-Pretzer, E., Webster, S. (2021) 'Impacts of online support groups on quality of life, and perceived anxiety and depression in those with ME/CFS: a survey', *Fatigue*, 9 (2), p.113-122

[37] Kummervold, p., Gammon, D., Bergvik, S., Johnsen, J., Hasvold, T., Rosenvinge, J., (2002) 'Social support in a wired world: Use of online mental health forums in Norway', *Nordic Journal of Psychiatry*, 56 (1) p.59-65.

[38] Leimeister, J.M., Schweizer, K., Leimeister, S., Krcmar, H. (2008) 'Do virtual communities matter for the social support of patients? Antecedents and effects of virtual relationships in online communities', *Information Technology & People*, 21 (4), p.350-374

[39] Caplan, S.E. (2003) 'Preference for Online Social Interaction: A Theory of Problematic Internet Use and Psychosocial Well-Being', *Communication Research*, 30 (6), p.625-649

[40] Allen, C. Vassilev, I., Lin, S., Culliford, D., Rogers, A. (2018) 'The contribution of internet use in personal networks of support for long-term condition self-management'. *Chronic Illness*. (0) 1-16.

[41] Munn, Z., Peters, M.D.J., Stern, C., Tufanaru, C., McArthur, A., Aromataris, E. (2018) 'Systematic review or scoping review? Guidance for authors when choosing between a systematic or scoping review approach'. *BMC Medical Research Methodology*. 18: 143.

[42] Daudt, H.ML., van Mossel, C., Scott, S.J. (2013) 'Enhancing the scoping study methodology: a large, inter-professional team's experience with Arksey and O'Malley's framework'. *BMC Medical Research Methodology*, 13:48.

[43] Kaplan, A. M., Haenlein, M., (2010) 'Users of the world, unite! The challenges and opportunities of Social Media, *Business Horizons*, 53 p.59-68.

[44] Collins (2022) *'Internet'* Accessed at: www.collinsdictionary.com/dictionary/english/internet

[45] Bramer, W.M., Rethlefsen, M,L., Kleijnen, J., Franco, O.H., (2017) 'Optimal database combinations for literature searches in systematic reviews: a prospective exploratory study', *Systematic Reviews*, 6 (245).

[46] Pham, M.T., Rajic, A., Greig, J.D., Sargeant, J.M., Papadopoulos, A., McEwan, S.A. (2014) 'A scoping review of scoping reviews: advancing the approach and enhancing the consistency'. *Research Synthesis Methods*. 5 371-385.

[47] McColl, M.A., Shortt, S., Godwin, M., Smith, K., Rowe, K., O'Brien, P/. Donnelly. (2009) 'Models for integrating rehabilitation and primary care: a scoping review'. *Archives of Physical Medicine and Rehabilitation*. 90(9): 1523-1531.

[48] Brien, S.E., Lorenzetti, D.L., Lewis, S., Kennedy, J., Ghsli, W.A. (2010) 'Overview of a formal scoping review on health system report cards'. *Implementation Science*. 5 (1): 2

[49] Grant, M., Booth, A. (2009) 'A typology of reviews: an analysis of 14 review types and associated methodologies'. *Health Info Libr J*.26 920: 91-108

[50] Critical Appraisal Skills Programme (2018) CASP Qualitative Checklist. Available at: https://casp-uk.b-cdn.net/wp-content/uploads/2018/03/CASP-Qualitative-Checklist-2018_fillable_form.pdf. (Accessed 9th August 2022).

[51] Mixed Methods Appraisal Tool (2018) MMAT Mixed Methods Checklist. Available at: http://mixedmethodsappraisaltoolpublic.pbworks.com/w/page/24607821/FrontPage. (Accessed 20th June 2023).

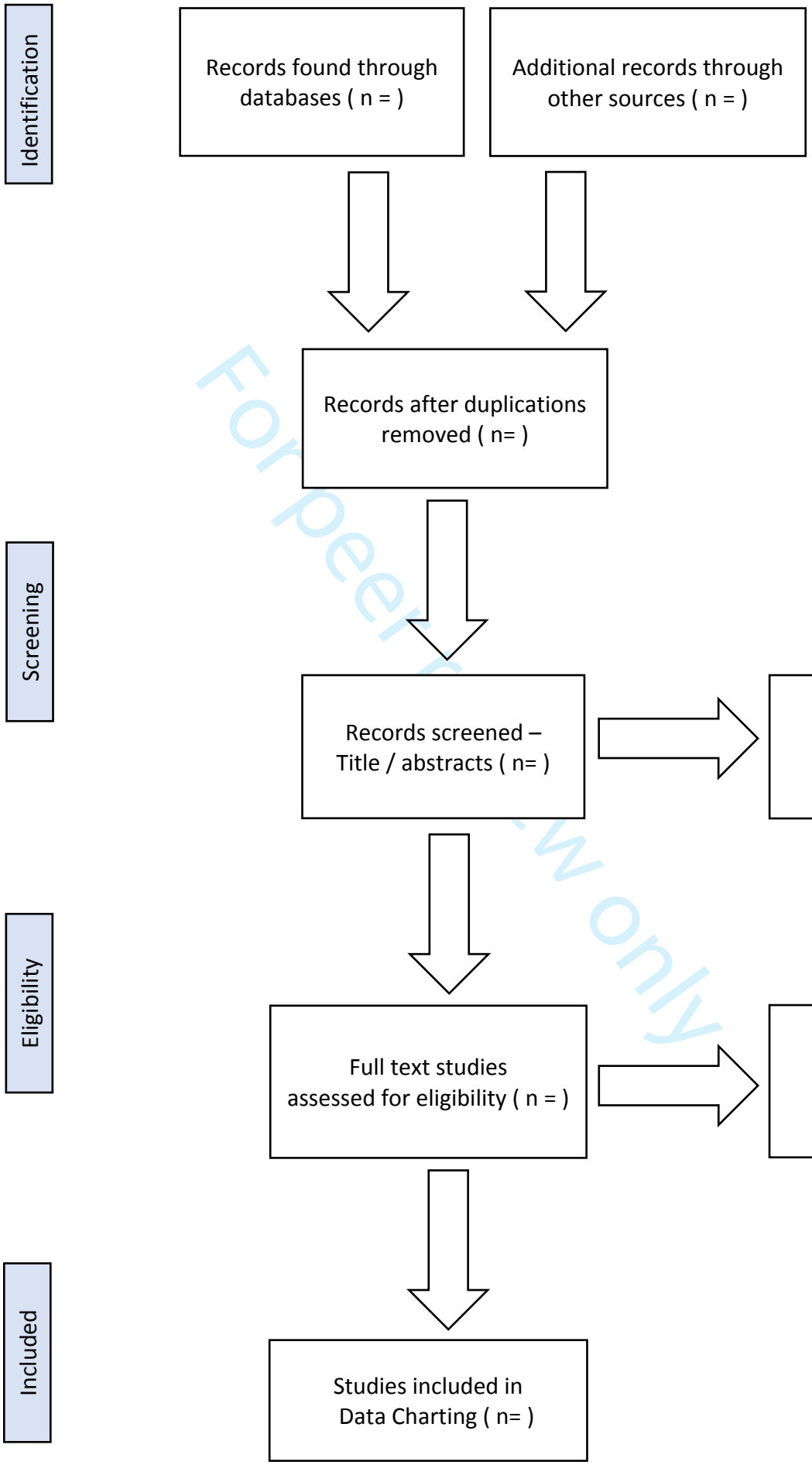
[52] Joanna Briggs Institute (2020) Critical Appraisal Tools. Available at: https://jbi.global/critical-appraisal-tools. (Accessed 20th June 2023).

[53] Braun, V., Clarke, V. (2006) 'Using thematic analysis in psychology'. *Qualitative Research Psychology*. 3: 77-101.

[54] Tong, A., Remming, K., McInnes, E., Oliver, S/, Craig, J. (2012) 'Enhancing transparency in reporting the synthesis of qualitative research: ENTREQ'. BMC Medical Research Methodology. 12: 181.

TABLE ONE - Search Strategy Example for Web of Science	
POPULATION	'ME' *OR* 'M.E.' *OR* 'Myalgic Encephalomyelitis' *OR* 'CFS' *OR* 'Chronic Fatigue Syndrome' *OR* 'ME/CFS' *OR* 'CFS/ME'
AND	
CONCEPT OF INTEREST	Online OR 'online us*' OR 'online activit*' OR 'online platform' OR 'online discussion' OR 'online social media' OR 'online communit*' OR 'online social network' OR 'online group' OR 'online health communit*' OR 'online support' OR 'online peer-to-peer' OR 'online user experience' OR 'online virtual' OR internet OR 'internet us*' OR 'internet activit*' OR 'internet discussion' OR 'internet communit*' OR 'internet-based' OR 'internet forum' OR 'internet communication' OR 'internet group' OR 'internet support' OR 'internet peer-to-peer' OR 'internet user experience' OR 'internet virtual' OR Facebook OR YouTube OR gaming OR Instagram OR TikTok OR 'message boards'.

PRISMA Flow Chart of Systematic Scoping Review Process



Reporting checklist for protocol of a systematic review and meta analysis.

Based on the PRISMA-P guidelines.

Instructions to authors

Complete this checklist by entering the page numbers from your manuscript where readers will find each of the items listed below.

Your article may not currently address all the items on the checklist. Please modify your text to include the missing information. If you are certain that an item does not apply, please write "n/a" and provide a short explanation.

Upload your completed checklist as an extra file when you submit to a journal.

In your methods section, say that you used the PRISMA-Preporting guidelines, and cite them as:

Moher D, Shamseer L, Clarke M, Ghersi D, Liberati A, Petticrew M, Shekelle P, Stewart LA. Preferred Reporting Items for Systematic Review and Meta-Analysis Protocols (PRISMA-P) 2015 statement. Syst Rev. 2015;4(1):1.

		Reporting Item	Page Number
Title			
Identification	#1a	Identify the report as a protocol of a systematic review	1
Update	#1b	If the protocol is for an update of a previous systematic review, identify as such	N/A
Registration			
	#2	If registered, provide the name of the registry (such as PROSPERO) and registration number	N/A
Authors			
Contact	#3a	Provide name, institutional affiliation, e-mail address of all protocol authors; provide physical mailing address of corresponding author	1
Contribution	#3b	Describe contributions of protocol authors and identify the guarantor of the review	13

Amendments			
	#4	If the protocol represents an amendment of a previously completed or published protocol, identify as such and list changes; otherwise, state plan for documenting important protocol amendments	N/A
Support			
Sources	#5a	Indicate sources of financial or other support for the review	13
Sponsor	#5b	Provide name for the review funder and / or sponsor	N/A
Role of sponsor or funder	#5c	Describe roles of funder(s), sponsor(s), and / or institution(s), if any, in developing the protocol	1
Introduction			
Rationale	#6	Describe the rationale for the review in the context of what is already known	2-6
Objectives	#7	Provide an explicit statement of the question(s) the review will address with reference to participants, interventions, comparators, and outcomes (PICO)	6-7
Methods			
Eligibility criteria	#8	Specify the study characteristics (such as PICO, study design, setting, time frame) and report characteristics (such as years considered, language, publication status) to be used as criteria for eligibility for the review	7
Information sources	#9	Describe all intended information sources (such as electronic databases, contact with study authors, trial registers or other grey literature sources) with planned dates of coverage	8
Search strategy	#10	Present draft of search strategy to be used for at least one electronic database, including planned limits, such that it could be repeated	8
Study records - data management	#11a	Describe the mechanism(s) that will be used to manage records and data throughout the review	8, 15
Study records - selection process	#11b	State the process that will be used for selecting studies (such as two independent reviewers) through each phase of the review (that is, screening, eligibility and inclusion in meta-analysis)	9
Study records - data	#11c	Describe planned method of extracting data from reports (such as	9-10

collection process		piloting forms, done independently, in duplicate), any processes for obtaining and confirming data from investigators	
Data items	#12	List and define all variables for which data will be sought (such as PICO items, funding sources), any pre-planned data assumptions and simplifications	7
Outcomes and prioritization	#13	List and define all outcomes for which data will be sought, including prioritization of main and additional outcomes, with rationale	10-11
Risk of bias in individual studies	#14	Describe anticipated methods for assessing risk of bias of individual studies, including whether this will be done at the outcome or study level, or both; state how this information will be used in data synthesis	9-10
Data synthesis	#15a	Describe criteria under which study data will be quantitatively synthesised	N/A
Data synthesis	#15b	If data are appropriate for quantitative synthesis, describe planned summary measures, methods of handling data and methods of combining data from studies, including any planned exploration of consistency (such as I2, Kendall's τ)	N/A
Data synthesis	#15c	Describe any proposed additional analyses (such as sensitivity or subgroup analyses, meta-regression)	N/A
Data synthesis	#15d	If quantitative synthesis is not appropriate, describe the type of summary planned	10-11
Meta-bias(es)	#16	Specify any planned assessment of meta-bias(es) (such as publication bias across studies, selective reporting within studies)	10-11
Confidence in cumulative evidence	#17	Describe how the strength of the body of evidence will be assessed (such as GRADE)	10

CASP Critical Appraisal Skills Programme

www.casp-uk.net
info@casp-uk.net
Summertown Pavilion, Middle Way Oxford OX2 7LG

CASP Checklist: 10 questions to help you make sense of a Qualitative research

How to use this appraisal tool: Three broad issues need to be considered when appraising a qualitative study:

- Are the results of the study valid? (Section A)
- What are the results? (Section B)
- Will the results help locally? (Section C)

The 10 questions on the following pages are designed to help you think about these issues systematically. The first two questions are screening questions and can be answered quickly. If the answer to both is "yes", it is worth proceeding with the remaining questions. There is some degree of overlap between the questions, you are asked to record a "yes", "no" or "can't tell" to most of the questions. A number of italicised prompts are given after each question. These are designed to remind you why the question is important. Record your reasons for your answers in the spaces provided.

About: These checklists were designed to be used as educational pedagogic tools, as part of a workshop setting, therefore we do not suggest a scoring system. The core CASP checklists (randomised controlled trial & systematic review) were based on JAMA 'Users' guides to the medical literature 1994 (adapted from Guyatt GH, Sackett DL, and Cook DJ), and piloted with health care practitioners.

For each new checklist, a group of experts were assembled to develop and pilot the checklist and the workshop format with which it would be used. Over the years overall adjustments have been made to the format, but a recent survey of checklist users reiterated that the basic format continues to be useful and appropriate.

Referencing: we recommend using the Harvard style citation, i.e.: *Critical Appraisal Skills Programme (2018). CASP (insert name of checklist i.e. Qualitative) Checklist. [online] Available at: URL. Accessed: Date Accessed.*

CASP Critical Appraisal Skills Programme

Paper for appraisal and reference:

Section A: Are the results valid?

1. Was there a clear statement of the aims of the research?

Yes ☐ Can't Tell ☐ No ☐

HINT: Consider
- what was the goal of the research
- why it was thought important
- its relevance

Comments:

2. Is a qualitative methodology appropriate?

Yes ☐ Can't Tell ☐ No ☐

HINT: Consider
- If the research seeks to interpret or illuminate the actions and/or subjective experiences of research participants
- Is qualitative research the right methodology for addressing the research goal

Comments:

Is it worth continuing?

3. Was the research design appropriate to address the aims of the research?

Yes ☐ Can't Tell ☐ No ☐

HINT: Consider
- if the researcher has justified the research design (e.g. have they discussed how they decided which method to use)

Comments:

CASP Critical Appraisal Skills Programme

4. Was the recruitment strategy appropriate to the aims of the research?

Yes ☐
Can't Tell ☐
No ☐

HINT: Consider
- If the researcher has explained how the participants were selected
- If they explained why the participants they selected were the most appropriate to provide access to the type of knowledge sought by the study
- If there are any discussions around recruitment (e.g. why some people chose not to take part)

Comments:

5. Was the data collected in a way that addressed the research issue?

Yes ☐
Can't Tell ☐
No ☐

HINT: Consider
- If the setting for the data collection was justified
- If it is clear how data were collected (e.g. focus group, semi-structured interview etc.)
- If the researcher has justified the methods chosen
- If the researcher has made the methods explicit (e.g. for interview method, is there an indication of how interviews are conducted, or did they use a topic guide)
- If methods were modified during the study. If so, has the researcher explained how and why
- If the form of data is clear (e.g. tape recordings, video material, notes etc.)
- If the researcher has discussed saturation of data

Comments:

CASP Critical Appraisal Skills Programme

6. Has the relationship between researcher and participants been adequately considered?

Yes ☐
Can't Tell ☐
No ☐

HINT: Consider

- If the researcher critically examined their own role, potential bias and influence during (a) formulation of the research questions (b) data collection, including sample recruitment and choice of location
- How the researcher responded to events during the study and whether they considered the implications of any changes in the research design

Comments:

Section B: What are the results?

7. Have ethical issues been taken into consideration?

Yes ☐
Can't Tell ☐
No ☐

HINT: Consider

- If there are sufficient details of how the research was explained to participants for the reader to assess whether ethical standards were maintained
- If the researcher has discussed issues raised by the study (e.g. issues around informed consent or confidentiality or how they have handled the effects of the study on the participants during and after the study)
- If approval has been sought from the ethics committee

Comments:

CASP Critical Appraisal Skills Programme

8. Was the data analysis sufficiently rigorous?

Yes ☐
Can't Tell ☐
No ☐

HINT: Consider
- If there is an in-depth description of the analysis process
- If thematic analysis is used. If so, is it clear how the categories/themes were derived from the data
- Whether the researcher explains how the data presented were selected from the original sample to demonstrate the analysis process
- If sufficient data are presented to support the findings
- To what extent contradictory data are taken into account
- Whether the researcher critically examined their own role, potential bias and influence during analysis and selection of data for presentation

Comments:

9. Is there a clear statement of findings?

Yes ☐
Can't Tell ☐
No ☐

HINT: Consider whether
- If the findings are explicit
- If there is adequate discussion of the evidence both for and against the researcher's arguments
- If the researcher has discussed the credibility of their findings (e.g. triangulation, respondent validation, more than one analyst)
- If the findings are discussed in relation to the original research question

Comments:

CASP Critical Appraisal Skills Programme

Section C: Will the results help locally?

10. How valuable is the research?

HINT: Consider

- If the researcher discusses the contribution the study makes to existing knowledge or understanding (e.g. do they consider the findings in relation to current practice or policy, or relevant research-based literature
- If they identify new areas where research is necessary
- If the researchers have discussed whether or how the findings can be transferred to other populations or considered other ways the research may be used

Comments:

Part I: Mixed Methods Appraisal Tool (MMAT), version 2018

Category of study designs	Methodological quality criteria	Responses			
		Yes	No	Can't tell	Comments
Screening questions (for all types)	S1. Are there clear research questions?				
	S2. Do the collected data allow to address the research questions?				
	Further appraisal may not be feasible or appropriate when the answer is 'No' or 'Can't tell' to one or both screening questions.				
1. Qualitative	1.1. Is the qualitative approach appropriate to answer the research question?				
	1.2. Are the qualitative data collection methods adequate to address the research question?				
	1.3. Are the findings adequately derived from the data?				
	1.4. Is the interpretation of results sufficiently substantiated by data?				
	1.5. Is there coherence between qualitative data sources, collection, analysis and interpretation?				
2. Quantitative randomized controlled trials	2.1. Is randomization appropriately performed?				
	2.2. Are the groups comparable at baseline?				
	2.3. Are there complete outcome data?				
	2.4. Are outcome assessors blinded to the intervention provided?				
	2.5 Did the participants adhere to the assigned intervention?				
3. Quantitative non-randomized	3.1. Are the participants representative of the target population?				
	3.2. Are measurements appropriate regarding both the outcome and intervention (or exposure)?				
	3.3. Are there complete outcome data?				
	3.4. Are the confounders accounted for in the design and analysis?				
	3.5. During the study period, is the intervention administered (or exposure occurred) as intended?				
4. Quantitative descriptive	4.1. Is the sampling strategy relevant to address the research question?				
	4.2. Is the sample representative of the target population?				
	4.3. Are the measurements appropriate?				
	4.4. Is the risk of nonresponse bias low?				
	4.5. Is the statistical analysis appropriate to answer the research question?				
5. Mixed methods	5.1. Is there an adequate rationale for using a mixed methods design to address the research question?				
	5.2. Are the different components of the study effectively integrated to answer the research question?				
	5.3. Are the outputs of the integration of qualitative and quantitative components adequately interpreted?				
	5.4. Are divergences and inconsistencies between quantitative and qualitative results adequately addressed?				
	5.5. Do the different components of the study adhere to the quality criteria of each tradition of the methods involved?				

Hong QN, Pluye P, Fàbregues S, Bartlett G, Boardman F, Cargo M, Dagenais P, Gagnon M-P, Griffiths F, Nicolau B, O'Cathain A, Rousseau M-C, Vedel I. Mixed Methods Appraisal Tool (MMAT), version 2018. Registration of Copyright (#1148552), Canadian Intellectual Property Office, Industry Canada.

CHECKLIST FOR ANALYTICAL CROSS SECTIONAL STUDIES

Critical Appraisal tools for use in JBI Systematic Reviews

INTRODUCTION

JBI is an JBI is an international research organisation based in the Faculty of Health and Medical Sciences at the University of Adelaide, South Australia. JBI develops and delivers unique evidence-based information, software, education and training designed to improve healthcare practice and health outcomes. With over 70 Collaborating Entities, servicing over 90 countries, JBI is a recognised global leader in evidence-based healthcare.

JBI Systematic Reviews

The core of evidence synthesis is the systematic review of literature of a particular intervention, condition or issue. The systematic review is essentially an analysis of the available literature (that is, evidence) and a judgment of the effectiveness or otherwise of a practice, involving a series of complex steps. JBI takes a particular view on what counts as evidence and the methods utilised to synthesise those different types of evidence. In line with this broader view of evidence, JBI has developed theories, methodologies and rigorous processes for the critical appraisal and synthesis of these diverse forms of evidence in order to aid in clinical decision-making in healthcare. There now exists JBI guidance for conducting reviews of effectiveness research, qualitative research, prevalence/incidence, etiology/risk, economic evaluations, text/opinion, diagnostic test accuracy, mixed-methods, umbrella reviews and scoping reviews. Further information regarding JBI systematic reviews can be found in the JBI Evidence Synthesis Manual.

JBI Critical Appraisal Tools

All systematic reviews incorporate a process of critique or appraisal of the research evidence. The purpose of this appraisal is to assess the methodological quality of a study and to determine the extent to which a study has addressed the possibility of bias in its design, conduct and analysis. All papers selected for inclusion in the systematic review (that is – those that meet the inclusion criteria described in the protocol) need to be subjected to rigorous appraisal by two critical appraisers. The results of this appraisal can then be used to inform synthesis and interpretation of the results of the study. JBI Critical appraisal tools have been developed by the JBI and collaborators and approved by the JBI Scientific Committee following extensive peer review. Although designed for use in systematic reviews, JBI critical appraisal tools can also be used when creating Critically Appraised Topics (CAT), in journal clubs and as an educational tool.

JBI CRITICAL APPRAISAL CHECKLIST FOR ANALYTICAL CROSS SECTIONAL STUDIES

Reviewer_____Date_____

Author_____Year_____ Record Number_____

	Yes	No	Unclear	Not applicable
1. Were the criteria for inclusion in the sample clearly defined?	☐	☐	☐	☐
2. Were the study subjects and the setting described in detail?	☐	☐	☐	☐
3. Was the exposure measured in a valid and reliable way?	☐	☐	☐	☐
4. Were objective, standard criteria used for measurement of the condition?	☐	☐	☐	☐
5. Were confounding factors identified?	☐	☐	☐	☐
6. Were strategies to deal with confounding factors stated?	☐	☐	☐	☐
7. Were the outcomes measured in a valid and reliable way?	☐	☐	☐	☐
8. Was appropriate statistical analysis used?	☐	☐	☐	☐

Overall appraisal: Include ☐ Exclude ☐ Seek further info ☐

Comments (Including reason for exclusion)

_____

_____

_____

EXPLANATION OF ANALYTICAL CROSS SECTIONAL STUDIES CRITICAL APPRAISAL

How to cite: Moola S, Munn Z, Tufanaru C, Aromataris E, Sears K, Sfetcu R, Currie M, Qureshi R, Mattis P, Lisy K, Mu P-F. Chapter 7: Systematic reviews of etiology and risk . In: Aromataris E, Munn Z (Editors). *JBI Manual for Evidence Synthesis.* JBI, 2020. Available from https://synthesismanual.jbi.global

Analytical cross sectional studies Critical Appraisal Tool

Answers: Yes, No, Unclear or Not/Applicable

1. Were the criteria for inclusion in the sample clearly defined?

The authors should provide clear inclusion and exclusion criteria that they developed prior to recruitment of the study participants. The inclusion/exclusion criteria should be specified (e.g., risk, stage of disease progression) with sufficient detail and all the necessary information critical to the study.

2. Were the study subjects and the setting described in detail?

The study sample should be described in sufficient detail so that other researchers can determine if it is comparable to the population of interest to them. The authors should provide a clear description of the population from which the study participants were selected or recruited, including demographics, location, and time period.

3. Was the exposure measured in a valid and reliable way?

The study should clearly describe the method of measurement of exposure. Assessing validity requires that a 'gold standard' is available to which the measure can be compared. The validity of exposure measurement usually relates to whether a current measure is appropriate or whether a measure of past exposure is needed.

Reliability refers to the processes included in an epidemiological study to check repeatability of measurements of the exposures. These usually include intra-observer reliability and inter-observer reliability.

4. Were objective, standard criteria used for measurement of the condition?

It is useful to determine if patients were included in the study based on either a specified diagnosis or definition. This is more likely to decrease the risk of bias. Characteristics are another useful approach to matching groups, and studies that did not use specified diagnostic methods or definitions should provide evidence on matching by key characteristics

5. Were confounding factors identified?

Confounding has occurred where the estimated intervention exposure effect is biased by the presence of some difference between the comparison groups (apart from the exposure investigated/of interest). Typical confounders include baseline characteristics, prognostic factors, or concomitant exposures (e.g. smoking). A confounder is a difference between the comparison groups and it influences the direction of the study results. A high quality study at the level of cohort design will identify the potential confounders and measure them (where possible). This is difficult for studies where behavioral, attitudinal or lifestyle factors may impact on the results.

6. Were strategies to deal with confounding factors stated?

Strategies to deal with effects of confounding factors may be dealt within the study design or in data analysis. By matching or stratifying sampling of participants, effects of confounding factors can be adjusted for. When dealing with adjustment in data analysis, assess the statistics used in the study. Most will be some form of multivariate regression analysis to account for the confounding factors measured.

7. Were the outcomes measured in a valid and reliable way?

Read the methods section of the paper. If for e.g. lung cancer is assessed based on existing definitions or diagnostic criteria, then the answer to this question is likely to be yes. If lung cancer is assessed using observer reported, or self-reported scales, the risk of over- or under-reporting is increased, and objectivity is compromised. Importantly, determine if the measurement tools used were validated instruments as this has a significant impact on outcome assessment validity.

Having established the objectivity of the outcome measurement (e.g. lung cancer) instrument, it's important to establish how the measurement was conducted. Were those involved in collecting data trained or educated in the use of the instrument/s? (e.g. radiographers). If there was more than one data collector, were they similar in terms of level of education, clinical or research experience, or level of responsibility in the piece of research being appraised?

8. Was appropriate statistical analysis used?

As with any consideration of statistical analysis, consideration should be given to whether there was a more appropriate alternate statistical method that could have been used. The methods section should be detailed enough for reviewers to identify which analytical techniques were used (in particular, regression or stratification) and how specific confounders were measured.

For studies utilizing regression analysis, it is useful to identify if the study identified which variables were included and how they related to the outcome. If stratification was the analytical approach used, were the strata of analysis defined by the specified variables? Additionally, it is also important to assess the appropriateness of the analytical strategy in terms of the assumptions associated with the approach as differing methods of analysis are based on differing assumptions about the data and how it will respond.

BMJ Open

A Protocol for a scoping review of how people with ME/CFS use the internet.

Journal:	*BMJ Open*
Manuscript ID	bmjopen-2023-076904.R1
Article Type:	Protocol
Date Submitted by the Author:	06-Dec-2023
Complete List of Authors:	Shortland, Diane; University of Birmingham Edgbaston Campus, school of medicine and dentistry; Fazil, Qulsom; The University of Birmingham Hallett, Nutmeg; University of Birmingham, School of Nursing and Midwifery Lavis, Anna; University of Birmingham, Institute of Applied Health Research
Primary Subject Heading:	Qualitative research
Secondary Subject Heading:	Research methods
Keywords:	Fatigue, GENERAL MEDICINE (see Internal Medicine), Protocols & guidelines < HEALTH SERVICES ADMINISTRATION & MANAGEMENT, Chronic Disease, Social Interaction

SCOPING REVIEW PROTOCOL for publication

A protocol for a scoping review of how people with ME/CFS use the internet.

Authors:

Lead researcher - Diane Shortland PhD. University of Birmingham: DLS760@student.bham.ac.uk

Supervisor – Dr. Qulsom Fazil, University of Birmingham: Q.A.Fazil@bham.ac.uk

Supervisor – Dr Nutmeg Hallett, University of Birmingham: N.N.Hallett@bham.ac.uk

Supervisor – Dr Anna Lavis, University of Birmingham: A.C.Lavis@bham.ac.uk

Abstract

Introduction:

Myalgic Encephalomyelitis (ME) is a chronic neurological illness affecting many bodily systems, commonly the nervous and immune systems. Also known as Chronic Fatigue Syndrome (CFS), key symptoms are extreme fatigue, post-exertional malaise, cognitive problems and sleep disturbance (Jason et al 2015).[1] With reported higher levels of online activity for people with ME/CFS than other patient groups (Westerby 2013 cited in Ytre-Arne 2016), [2] it's crucial to gain more knowledge of usage characteristics and experience of online use, and it's integration into everyday life. This scoping review protocol details the proposed methods for gaining insight into this little known phenomenon.

Methods & Analysis:

This review uses the methodological framework for conducting a scoping review by Arksey & O'Malley, [3] with further guidance by Levac, Colquhoun & O'Brien, [4] and the Joanna Briggs Institute. [5] It also refers to the PRISMA-Preporting guidelines. [6] The following bibliographic databases will be searched: Embase, Medline, PsychINFO, Cinahl, AMED, and ASSIA, plus Web of Science, ProQuest Dissertations & Theses Global, Scopus, and Google Scholar for grey literature. Reference lists of included papers will be studied. Two reviewers will independently screen title-abstracts, and then full text of studies against inclusion criteria. Remaining studies will be quality assessed using appropriate critical appraisal tools. Findings will be charted and mapped to gain in-depth knowledge of the use of the internet in people with ME/CFS.

Ethics and Dissemination:

The findings from this review will be disseminated through peer-reviewed publication and a report for leading charities of ME/CFS. The review will collect secondary data only and therefore does not need ethical approval.

Article Summary:

Strengths & Limitations of this study

- To our knowledge this is the first scoping review to map out the online usage and experience of people with ME/CFS.
- A strength of the review will be the rigorous and transparent approach based on a solid methodological framework and the Preferred Reporting Items for Systematic reviews and Meta-analysis extension for Scoping Reviews checklist (PRISMA-ScR).
- The quality of the scoping review will be enhanced by the use of a second reviewer for study selection and charting of results.
- Eligible studies will be quality assessed in accordance with their study design.
- The review is confined to English language which may exclude other language studies that may contain valuable data.

Introduction

Myalgic encephalomyelitis (ME) – meaning inflammation of the brain and spinal cord – is a long term chronic neurological illness, often fluctuating in nature, that causes many symptoms affecting many bodily systems, most commonly the nervous and immune systems (Action for ME 2022). [7] Since 1988, the illness has also been known as Chronic Fatigue Syndrome (CFS). Many publications and researchers use both ME and CFS terms interchangeably and so we have operationalised both names as suitable for inclusion in this review. It is worth noting however, that debate exists in defining and classifying the two and there is evidence of distinct historical trajectories of ME and CFS, with distinguishing features of diagnosis and as such, including both names together could create additional issues surrounding the illness (Grue 2014). [8] Studies have reported however, that despite this debate, certain core symptoms of the illness do appear to be consistently present

across both classifications, namely extreme fatigue, post-exertional malaise, neuro-cognitive difficulties and sleep disturbance (Jason et al 2015). [1]

People with ME/CFS are significantly more impaired in both physical and social functioning than other long term illnesses (Kingod et al 2018; Hvidberg et al 2015; Pendergrast et al 2016).[9, 10, 11] The illness has a negative impact on people's relationships and social networks, with suicide ideation endorsed more frequently in those experiencing unsupportive interactions and social distancing (Clarke & James 2003; McManimen et al 2018). [12, 13] Due to the contested nature of the condition (there is yet no available biomarker for the condition and its diagnosis is therefore subjective, raising a debate over the decades between the medical and psychological realms as to its aetiology and treatment), legitimacy of the illness is often questioned in immediate social support networks, causing additional stress (Harris et al 2016; McManimen et al 2018). [14, 13] As Bowling (2005) states, [15] lack of social support, participation and contact is associated with increased mortality risk and delayed recovery from disease. In a survey by Action for ME (2019), [16] 94% of participants had stopped or reduced social contact, and up to 97% of the 4038 participants said they felt socially isolated because of their condition. Patients describe feeling overwhelmed and let down when disbelieved. When seeking help was unsatisfactory, sufferers responded to this by taking more responsibility for their illness management via 'self-help' tactics (Edwards, Thompson & Blair 2007). [17]

There are reported higher levels of online activity among people with ME than other patient groups (Westerby 2013 cited in Ytre-Arne 2016). [2] Online peer-to-peer support in the form of interactive websites and social media, is now highly valued in chronic illness as a way to connect to others who share the same illness (Van der Eijk et al 2013; Lian & Nettleton 2015). [18, 19] Transcending geographic boundaries, the internet is convenient to those with limited mobility (Lasker et al 2006; Eichhorn 2008). [20, 21] Online communities provide support for people with long term illness with a growing reliance on social media in patients experiencing social isolation and who fear marginalisation because of their illness (Loane & D'Alessandro 2014; Perkins, Coulson & Davies 2020). [22, 23] It also offers support to people otherwise limited by disability or stigma when accessing support offline (Drentea & Moren-Cross 2005). [24]

Uncertainty surrounding illness appears to be a driving factor for internet use (Conrad & Stults 2010) [25] with internet itself being an increasingly public experience as people share personal information and interact in public spheres (Conrad, Bandini & Vasquez 2016). [26] As Beck, Gurion & Sheva (2004) state, [27] "users of the world wide web are no longer passive audiences of data consumers… but are active participants controlling the content of the information. They shape the quality of the data… (facilitating) the expression of emotions (output) and the input of emotional messages, thus developing and reinforcing important social ties between users, forming a system of relationships similar to ties of family and friendship" (p.46). Receiving problem-focused and emotion-focused support from others aids coping and thus becomes a primary driver of willingness to offer such support to others (Lin et al 2015). [28] Online users describe 'social overload' however, where people feel they're giving too much social support to others and experience online group exhaustion (Maier et al 2015). [29]

So how does online usage interplay with the 'real world', particularly for ME patients who are often housebound due to the chronicity of their condition? In general, there is a "sharp distinction between concepts from the virtual world online and the 'real world' offline" but "technology enters and is gradually integrated into people's daily lives" (Lie & Sorensen 1996 cited in Beck, Gurion & Sheva 2004), [27] by a process of 'domestication' where people adapt new technologies and bring them into their home, transferring elements of the physical world into the virtual environment, merging the two worlds and creating a much broader definition of reality. Understanding how people with ME use the internet to aid their illness management and enhance their experience of daily life, is crucial in gaining insight into how informational and social support is found and utilised online and offline. It will shed light on how people's overall support networks are created and maintained, as well as identify the benefits of such illness behaviour.

Kingod et al (2016) [30] studied how people with chronic conditions experience online peer-to-peer social support and its influence in everyday life, in a systematic review of 13 papers, but none of them covered the illness ME. They found four main themes: identity, social support and connectivity, experiential knowledge that both strengthened social ties and

supported offline ties, and collective voice and mobilisation. Allen et al (2020) [31] also looked at chronic illnesses which included ME in a primary study of 30 people across varying conditions. They found that online support was sought in response to deficits in offline support; it was used to assist offline ties as well as substitute offline support.

Both Kingod et al (2016) [30] and Allen et al (2020) [31] stressed the need for further research into understanding the boundaries of online and offline social dimensions and relevance in daily life; how the role of online ties serve within personal networks. Essentially how do people decide who to turn to now they have greater choice in who contributes to their everyday illness management and coping? Having further advancement of knowledge in this area will inform healthcare practice social support initiatives and aim to improve services to those housebound with ME/CFS. It will also gain knowledge into the lack of support present in the home life of people with ME/CFS.

Initial searches revealed a lack of studies conducted in this area that focused on ME/CFS. Studies on other chronic illnesses have an element of transferability of their findings to ME/CFS and several papers were found here highlighting a topic worthy of attention. Preference for online support over offline support was highlighted in cancer and diabetes patients (Chung 2013), [32] with a lack of real world social support predicting active participation in online groups (Cummings, Sproul & Kiesler 2002). [33] The benefits of using social media in health communication include interaction with others, the availability of shared knowledge, widened access to health information, social and emotional support, and empowerment in their healthcare process (Moorhead et al 2013; Huang, Chengalur-Smith & Pinsonneault 2014). [34, 35] Investigating the perceived impact of online participation, Morehouse et al (2021) [36] found people gained a sense of belonging, validation and supportive friendships, decreasing feelings of depression and increasing quality of life. As much as 75% of a sample studied by Kummervold et al (2009) [37] found it easier to discuss personal problems online than face-to-face. Virtual communities appear to play an important role in meeting patients social needs; sense of community is positively associated with cancer patients well-being in areas of personal relations and personal growth (Leimeister et al 2008). [38]

Caplan (2003) [39] found that depressed people may develop preferences for online social interaction but this in turn leads to negative outcomes associated with internet use, and Allen et al (2018) [40] concluded that internet use may indicate an avoidance or absence of offline support. Moreover, Chung (2013) [32] found those dissatisfied with their offline relationships were more likely to develop preference for online social interaction and this can become problematic when excessive reliance increases disengagement from offline interaction. Findings are largely positive in that using the internet appears to improve everyday life, however there are studies that highlight negative impacts. Given that ME/CFS is a contested illness with legitimacy issues and increased stigma, will data collected from the ME/CFS community produce similar or different themes to other conditions?

This scoping review will aim to explore the online usage characteristics of ME/CFS patients and inter-relatedness within everyday life of their online and offline worlds. To prevent unnecessary duplication, a preliminary search for existing scoping and systematic reviews on the subject was carried out in May 2022. To our knowledge, a comprehensive synthesis of related studies on ME/CFS in this field remains absent.

Methods/Design

In order to capture the broadest scope of literature on the topic of online usage in people with ME/CFS, we decided to use a scoping review method. A scoping review is ideal for mapping out the scope or coverage of a body of literature on a given topic when the emerging evidence is still unclear and more specific questions cannot presently be posed (Peters et al 2020). [5] They give a clear indication of available literature, regardless of study design, and an overview of its focus, identifying characteristics of studies to provide an overall picture of current evidence (Munn et al 2018). [41] To map our field of study and examine the extent, range and nature of research activity to date, as well as identify any knowledge gaps in research, our protocol was developed using a framework set out by Arksey & O'Malley (2005). [3] We have also incorporated later improvements to this framework by the work of Levac and collegaues (2010), [4] and the Joanna Briggs Institute (2015). [5]

Traditionally a scoping review concerns itself with summary of results and does not evaluate the quality of included studies. Revisions have noted the value of quality assessment for future researchers however (Daudt, Mossel & Scott 2013 p.6) [42] and so we intend to incorporate this into our review. Guidance from the Preferred Reporting Items for Systematic Reviews and Meta-Analyses Extension for Scoping Reviews checklist (PRISMA - ScR) [6] has been consulted to yield greater transparency and reproducibility. Arksey & O'Malley's framework proposes five mandatory stages (outlined below) and a sixth optional stage: consultation with stakeholders. [3] Our current review does not involve this due to the nascent stage of the project. However, the findings of this review will inform a translation of knowledge engagement exercise which will involve consultation with stakeholders.

Stage One: Identifying the Research Question

The research question for this proposed scoping review aims for comprehensiveness and so will be broad to cover the full breadth of evidence in the field. We aim to answer the following question: **How do people with ME/CFS use the internet?** This aim will be achieved by addressing the following objectives:

- Examine the usage characteristics of people with ME/CFS utilising the internet. (What do they do online, when, and for how long?)
- Examine people with ME/CFS' experiences of online usage. (Why do they go online and what do they gain from going online?)
- Examine people with ME/CFS' online usage inter-relating with their offline lives. (e.g. how does using the internet fit alongside offline informational and social support?)

Arksey & O'Malley (2005) saw scoping as an iterative methodological skill and as such it may be appropriate and acceptable to add supplementary file 1 questions based on emerging findings during the review process. [3] We may notice other important data that could be useful to extract (Peters et al 2015). [5] Any changes or amendments will be clearly stated and explained.

Stage Two: Identifying relevant studies

Study eligibility:

We will aim to find both published and grey literature studies. Loosely using a PCC (Population, Concept, Context) framework to develop our inclusion criteria, to align with our objectives and research questions, our population will be adults with a formal diagnosis of ME/CFS, from any symptom classification criteria in operation (Oxford, Fukuda, Canadian Consensus Criteria, NICE, International Consensus Criteria, SEID), as well as those without an official diagnosis but who self-identify as having ME/CFS. This is included since historically the illness has suffered endless definition, classification and standardisation issues which have resulted in many people with the condition not receiving a correct diagnosis. In an attempt to avoid missing any relevant data, this broad use of the term ME/CFS will be used. All levels of severity will also be included in data collection as it is anticipated that many studies may not specify severity, plus those that do will provide a useful means of comparison against internet usage frequency and type. Our concept of interest is internet use. We define 'internet use' as the computer network that allows users to connect with other users and content from all over the world (Collins 2022). [43] Online information, content and social support exists through many various technological avenues nowadays. Kaplan & Haenlein (2010) operationalised 'social media' usage in five main categories: collaborative projects, blogs, content communities, social networking sites and virtual worlds. [44] Gaming is also an online social experience now as it is shared live with other users. The context is loosely any available knowledge that involves personal use of the internet and not organised institutionalised treatment agendas. Internet use therefore, for the purposes of this review, will only be relevant if it has some direct relation to ME/CFS, for instance, searching for information and guidance of the illness, the sharing of ME/CFS related knowledge, social participation online with other people who have ME/CFS, or the use of social media and forums linked to the illness in some way.

Search strategy:

Guidance by the Joanna Briggs Institute (Peters et al 2015) [5] recommends a three-stage process to searching the literature of which we have included all advised stages. An initial search of limited databases has been conducted and from analysis of these results, key words and index terms have been identified. A reference librarian was consulted in preparation, and a systematic search plan was formed with search terms incorporating medical subject headings (MeSH) as well as text words combining comprehensive terms for

contemporary social media, and Boolean operators 'AND' and 'OR' (Table 1 shows an example search strategy of keywords for Web of Science. This will be adapted to suit individual databases). Since scoping is an iterative process (Arksey & O'Malley 2005), [3] a pilot of searching will take place and terms will be refined if deemed necessary.

TABLE ONE - Search Strategy Example for Web of Science	
POPULATION	'ME' *OR* 'M.E.' *OR* 'Myalgic Encephalomyelitis' *OR* 'CFS' *OR* 'Chronic Fatigue Syndrome' *OR* 'ME/CFS' *OR* 'CFS/ME'
AND	
CONCEPT OF INTEREST	Online OR 'online us*' OR 'online activit*' OR 'online platform' OR 'online discussion' OR 'online social media' OR 'online communit*' OR 'online social network' OR 'online group' OR 'online health communit*' OR 'online support' OR 'online peer-to-peer' OR 'online user experience' OR 'online virtual' OR internet OR 'internet us*' OR 'internet activit*' OR 'internet discussion' OR 'internet communit*' OR 'internet-based' OR 'internet forum' OR 'internet communication' OR 'internet group' OR 'internet support' OR 'internet peer-to-peer' OR 'internet user experience' OR 'internet virtual' OR Facebook OR YouTube OR gaming OR Instagram OR TikTok OR 'message boards'.

Secondly we intend to include extensive electronic searches of the following bibliographic databases (conducted in Aug to November 2022): EMBASE, Medline, Cinahl, PsychINFO, AMED. And ASSIA. Bramer et al (2017) [45] found that optimal searching to ensure a minimum risk of missing studies, should use four key databases: Embase, Medline, Web of Science and Google Scholar. This produced a 98.3% recall of studies. Because online communities in relation to health have been explored across a range of professional, theoretical, sociological, psychological and healthcare settings however, additional databases have also been covered. Thirdly, Grey literature will be searched (during November 2022) via Scopus, Web of Science, and ProQuest Dissertations & Theses Global.

Web searching via Google Scholar will also take place as well as citations and references of key papers searched by hand.

All eligible studies that meet our inclusion criteria (adults over 18 years of age, located anywhere in the world, identified as having ME/CFS, found in English language peer-reviewed primary studies, on internet use) will be saved on an Excel spreadsheet. Our exclusion criteria are children under 18 years of age, those not identified as having ME/CFS, and systematic reviews since their content is already secondary in nature, so analysis would further dilute and potentially bias findings. Consideration was given to restricting studies to a date limitation since the advancement of internet-based platforms such as social media is a relatively new and still growing area. However it is not possible to confidently put a time limit on when such social media support truly began, so doing so would risk losing valuable studies.

Stage Three: Study selection

The primary researcher will run the initial searches, retrieving titles and abstracts, removing duplicates, and saving all files into a suitable data management storage. Two reviewers will go through the title and abstract of each study and screen them to identify studies that meet the inclusion criteria and will document all results in a 'screening' form. Any uncertainty regarding if a study is eligible or not, will be included at this stage to ensure nothing is missed. If multiple papers are found that describe the same data, we will include the paper that describes the most comprehensive findings. By citation chaining, reference lists of included studies will then be examined by the lead reviewer to identify any eligible studies that meet the inclusion criteria and added to the database findings. Forward searching of papers, via Scopus, that have been cited, will also be checked. To further minimise location bias, authors and researchers of studies will be contacted.

Two independent reviewers will then read the full text of all provisionally included studies, to assess further against the inclusion and exclusion criteria. The devised extraction form will be piloted to ensure it is containing all relevant information needed. Studies will be included or excluded against the pre-determined eligibility criteria. Any missing data will attempt to be found by contacting the study authors for additional information. Any

discrepancies will be resolved through consultation with the wider research team. All reviewers will agree on the final list of included studies. A PRISMA-ScR flow diagram following the process of the scoping review will be used to demonstrate the selection process (Supplementary file 2).

Critical appraisal:

Contrary to the methodological framework originally set out by Arksey & O'Malley (2005), [3] we intend to appraise the remaining eligible studies for quality assessment. This will take place after the data extraction of full text studies. Pham et al (2014) reported only 22.38% of studies included an element of quality assessment. [46] McColl et al (2009) argue that the emphasis of a scoping review is on comprehensive coverage and not standard of evidence. [47] More recent refinements to guidelines however, support the use of some form of critical appraisal (Levac et al 2010, Peters et al 2015). [4,5] Brien et al (2010) believe a lack of quality assessment makes results more challenging to interpret and Grant & Booth (2009) believe it limits uptake of findings into policy and practice. [48, 49] Daudt (2013) considers quality assessment a necessary component of any scoping review and encourages the use of validated tools since use of reporting checklists increases transparency of methods and allows the reader to use the research appropriately. [42] Pham et al (2014) also recognises that some form of quality assessment would enable the identification of gaps in the evidence base rather than just where research is lacking. [46]

A quality assessment form will be used to extract relevant data for appraisal. Since it is expected that the majority of studies will be qualitative in nature, we have chosen the Critical Appraisal Skills Programme (see supplementary file 3) (CASP 2018). [50] If we identify any mixed methods studies then we will use the Mixed Methods Appraisal Tool (see supplementary file 4) (MMAT 2018) [51]. If any quantitative data is found we will use a checklist suited to the study design from the selection available at JBI, most likely the Checklist for Analytical Cross Sectional Studies (see supplementary file 5) [52]. Any discrepancies between reviewers on quality assessment will be discussed with the wider research team. No exclusion of eligible studies will take place as a result of appraisal since such studies can still contain rich and useful qualitative narrative. Poor quality studies will be highlighted and reflected upon within the data summaries. The outcome of each study

assessment, along with all study files will be included in an Excel spreadsheet alongside other data extraction details.

Stage Four: Charting the data

Two independent reviewers will perform a full-text review of provisionally included studies. Piloting of a small sample will take place, in accordance with advice from Levac et al (2010) to ensure agreement is reached on extraction consistency. [4] Charted data extracted and documented in a designed extraction form will include, but not be limited to, the following:

- Article title, authors, year of publication
- Study research aims
- Study design and setting
- Number of participants
- Characteristics of the population
- Study inclusion criteria
- Online usage information
- Data collection and analysis methods
- Study findings/outcome

As previously mentioned the review will take an iterative approach and so the content of extraction can be updated with discussion of the research team. This allows for the variables and themes to be included to best be able to answer the review question and meet its objectives.

Stage Five: Summarising and reporting the results

Levac and colleagues (2010) encourage a rigorous approach to analysis that includes descriptive numerical summary as well as thematic analysis. [4] Using the information extracted, data charting will involve visual summaries as well as narrative that describes the aims of included studies, their areas of focus, online user characteristics and findings to determine how the studies to date inform the current knowledge base. Any quantitative or mixed method studies will be 'qualitized' by extracting data from quantitative or mixed method studies and transforming it into textual descriptions to integrate with qualitative data and form a single summary comprising themes of narrative across the review studies

(Peters et al 2015). [5] Developed in an inductive manner without a set of a priori themes, these scoping study summary methods, in accordance with Braun & Clarke (2006), [53] will enable us to ascertain broad themes of what is known about how people with ME/CFS use the internet, their experience of doing so, and how this fits within their daily lives as per our research objectives. Two reviewers will perform all analysis independently before reaching consensus of themes and any discrepancies will be resolved with the wider research team.

Ethics and Dissemination:

All data generated will be stored on pass-protected computers. The authors will disseminate the findings through submission for publication in a peer-reviewed academic journal and a report will be written for leading charities of ME/CFS. The review itself will only deal with secondary data and therefore ethical approval is not required. Our findings will be used to inform the design of a future study aiming to gain greater knowledge of online social support in people with ME/CFS. Patient and Public Involvement will take place in the dissemination stages of this review and will guide all future research plans.

Patient and Public Involvement

Since this is a protocol only, it does not have any involvement with patients. Any data regarding patient participation is secondary through already published papers.

Discussion

A global comprehensive systematic scoping summary of primary data on internet use in people with ME/CFS, in terms of both usage and characteristics, and in relation to offline daily life, will be conducted to fill a gap in knowledge surrounding this under-researched area. In relation to online worlds, we frequently find ourselves asking research questions that contain complex medical, sociological, and social concepts. This type of research resists easy quantification and by aiming to critically reflect on the material found via this scoping review, we aim to capture the complexity inherent in such questions involving people's experiences (Kingod 2016). [30] In writing up the research findings we will be guided by enhancing transparency in reporting the synthesis of qualitative research (ENTREQ) (Tong et al 2012). [54] Dissemination will be relevant to academic knowledge sharing, charities for

ME/CFS that offer support and online services, as well as healthcare professionals and patients.

[3,783 words]

Keywords:
Myalgic Encephalomyelitis (M.E), Chronic Fatigue Syndrome (CFS), internet use, online usage, peer support.

Correspondence to:
Diane Shortland
University of Birmingham
DLS760@student.bham.ac.uk
07984 944683

Conflict of Interest:
No conflicts of interest were declared.

Funding:
The authors have not declared any specific grants for this research from any funding agency in public, commercial or not-for-profit sectors.

Author Contributions:
Diane Shortland – responsible for all areas of the study from protocol design, screening, analysis, discussion and writing up of study results.
Dr. Qulsom Fazil, Dr. Anna Lavis & Dr. Nutmeg Hallett – contribution to the design of study, drafting and revising of work, final approval and agreement

Contributor acknowledgement:
Michelle Minns-Sykes has contributed to this paper by independently screening papers for the systematic review.

Data Statement: Open access. Study available by contacting lead author.

Table One: Search strategy example for Web of Science.

Figure One: PRISMA Flow Chart of systematic scoping review process

References

[1] Jason, L; Kot, B; Sunnquist, M; Brown, A; Evans, M; Jantke, R; Williams, Y; Furst, J; Vernon, S. (2015) 'Chronic fatigue syndrome and myalgic encephalomyelitis: towards an empirical case definition'. *Health Psychology and Behavioral Medicine*. 3 (1) 82-93.

[2] Ytre-Arne, B. (2016) 'The social media experiences of long-term patients: Illness, identity and participation'. *Nordicom Review*. 37 (1) 57-70.

[3] Arksey, H., O'Malley, L. (2005) 'Scoping studies: towards a methodological framework'. *International Journal of Social Research Methodology*. 8:1 19-32.

[4] Levac, D., Colquhoun, H., O'Brien, K.K (2010) 'Scoping studies: advancing the methodology'. *Implementation Science* 5; 69.

[5] Peters, MDJ., Godfrey, C., McInerney, P., Munn, Z., TRicco, AC., Khalil, H. (2020) 'Scoping Reviews' in: Aromataris, E., Munn, Z (eds) JBI *Manual for Evidence Synthesis*. Available at: https://synthesismanual.jbi.global.

[6] Tricco, A.C., Lillie, E., Zarin, E., O'Brien, K.K., Colquhoun, H., Levac, D., Moher, D., Peters, M.D., Horsley, T., Weeks, T., Hempel, S. (2018) 'PRISMA extension for scoping reviews (PRISMA-ScR): checklist and explanation'. *Ann Intern Med*. 167 (7) 467-473.

[7] Action for ME (2022) *What is M.E.?* Available at: https://www.actionforme.org.uk/get-information/what-is-me/what-does-me-feel-like/ (Accessed 6th June 2022).

[8] Grue, J. (2014) 'A Garden of Forking Paths: A discourse perspective on 'Myalgic Encephalomyelitis' and 'Chronic Fatigue Syndrome'. *Critical Discourse Studies*. 11:1, 35-48.

[] Kingod, C; Bowman, E; Curran, H; Nacul, L; Lacerda, E. (2018) 'Functional Status and Well-Being in People with Myalgic Encephalomyelitis/Chronic Fatigue Syndrome Compared with People with Multiple Sclerosis and Healthy Controls'. *Pharmaco Economics Open*. 2: 381-392.

[10] Hvidberg, F., Brinth, M., Schouborg, L., Olesen, A.V ., Petersen, K.D., Ehlers, L. (2015) 'The health-related quality of life for patients with myalgic encephalomyelitis/chronic fatigue syndrome (ME/CFS)', PLoS One, 10 (7): e0132421.

[11] Pendergrast, T., Brown, A., Sunnquist, M., Jantke, R., Newton, J.L., Strand, E.B., Jason, L.A. (2016) 'Housebound versus non housebound patients with myalgic encephalomyelitis and chronic fatigue syndrome', *Chronic Illness*, 12 (4), pp.292-307

[12] Clarke, J., James, S. (2003) 'The radicalized self: the impact on the self of the contested nature of the diagnosis of chronic fatigue syndrome'. *Social Science & Medicine*. 1387-1395.

[13] McManimen, S., McClellan, D., Stoothoff, J., Jason, L. (2018) 'Effects of unsupportive social interactions, stigma, and symptoms on patients with myalgic encephalomyelitis'. *Journal of Community Psychology*. 46 (8) 959-971.

[14] Harris, K., Band, R., Cooper, H., Macintyre, G., Mejia, A., Wearden, A. (2016) 'Distress in significant others of patients with chronic fatigue syndrome: a systematic review of the literature'. *British Journal of Health Psychology*. 21 (4) 881-893.

[15] [15] Bowling, A. (2005) *Measuring Health: A review of quality of life measurement scales*. Maidenhead: Open University Press.

[16] Action for ME. (2019) '*Five year Big Survey*'. Available at: http://actionforme.org.uk/research-and-campaign/five-year-big-survey/ (Accessed 9th August 2022)

[17] Edwards, C.R., Thompson, A.R., Blair, A. (2007) 'An 'overwhelming illness': women's experiences of learning to live with chronic fatigue syndrome/myalgic encephalomyelitis', *Journal of Health Psychology*, 12 (2), p.203-215

[18] Van der Eijk, M., Faber, M., Aarts, J., Kremer, J., Munneke, M., Bloem, B. (2013) 'Using health communities to deliver patient-centred care to people with chronic conditions'. *Journal of Medical Internet Research*. 15 (6) 115.

[19] Lian, O., Nettleton, S. (2015) 'United we stand": Framing Myalgic Encephalomyelitis in a virtual symbolic community'. *Qualitative Health Research*. 25 (10) 1383-1394.

[20] Lasker, J., Sogolow, E., Sharim, R. (2006)' The role of an online community for people with a rare disease: Content analysis of messages posted on a primary biliary cirrhosis mailing list'.. 7 (1).

[21] Eichhorn, K. (2008) 'Soliciting and providing social support over the internet: An investigation of online eating disorder support groups'. *Journal of Computer Mediated Communication*. 14 (1) 67-78.

[22] Loane, S., D'Alessandro, S. (2014) 'Empowered and knowledgeable health consumers: the impact of online support groups on the doctor-patient relationship'. *Australian Marketing Journal*. 22: 238-245.

[23] Perkins, V., Coulson, N. S., and Davies, E.B. (2020) 'Using Online Support Communities for Tourette Syndrome and TIC Disorders: Online Survey of User's Experiences'. *Journal of Medical Internet Research.* 22 (11): e18099.

[24] Drentea, P., Moren-Cross, J. (2005) 'Social capital and social support on the web: The case of an internet mother site'. *Sociology of health & Illness*. 27: 920-943.

[25] Conrad, P., Stults, C., (2010) 'The Internet and the Experience of Illness' in Bird, C, E., Conrad, P., Fremont, A, M., Timmermans, S., *Handbook of Medical Sociology*, 6th edition. Vanderbilt University Press, p.179

[26] Conrad, P., Bandini, J., Vasquez, A. (2016) 'Illness and the internet: from private to public experience'. *Health*. 20 (1) 22-32.

[27] Beck, L, S., Gurion, B., Sheva, Beer, (2004) 'Internet Ethnography: Online and Offline', *International Journal of Qualitative Methods*, 3 (2) p.45-51.

[28] Lin, T.C., Hsu, J.S.C., Cheng, H.L., Chiu, C.M. (2015) 'Exploring the relationship between receiving and offering online social support: A dual social support model', *Information & Management*, 52 (3), p.371-383

[29] Maier, C., Laumer, S., Eckhardt, A., Weitzel, T., (2015) 'Giving too much social support: social overload on social networking sites', *European Journal of Information Systems*, 24 (5).

[30] Kingod, N., Cleal, B., Wahlberg, A., Husted, G. (2016) 'Online peer-to-peer communities in the daily lives of people with Chronic Illness: A qualitative systematic review'. *Qualitative Health Research*. 27 (1) 89-99.

[31] Allen, C., Vassilev, I., Kennedy, A., Rogers, A. (2020) 'The work and relatedness of ties mediated online in supporting long-term condition self-management', *Sociology of Health & Illness*, 42 (3) p.579-595.

[32] Chung, J. (2013) 'Social interaction in online support groups: preference for online social interaction over offline social interaction'. *Computers in Human Behaviour*. 29: 1408-1414.

[33] Cummings, J.N., Sproull, L., Kiesler, S.B. (2002) 'Beyond hearing: Where real-world and online support meet', *Group dynamics*, 6 (1), p.78-88

[34] Moorhead, S.A., Hazlett, D.E., Harrison, L., Carroll, J.K., Irwin, A., Hoving, C. (2013) 'A New Dimension of Health Care: Systematic Review of the Uses, Benefits, and Limitations of Social Media for Health Communication', *Journal of medical Internet research*, 15 (4), p.85

[35] Huang, K-Y., Chengalur-Smith, I., Ran, W. (2014) 'Not just for support: Companionship activities in healthcare virtual support communities', *Communications of the Association for Information Systems*, 34 (1), p.561-594

[36] Morehouse, S., Schaible, K., Williams, O., Herlache-Pretzer, E., Webster, S. (2021) 'Impacts of online support groups on quality of life, and perceived anxiety and depression in those with ME/CFS: a survey', *Fatigue*, 9 (2), p.113-122

[37] Kummervold, p., Gammon, D., Bergvik, S., Johnsen, J., Hasvold, T., Rosenvinge, J., (2002) 'Social support in a wired world: Use of online mental health forums in Norway', *Nordic Journal of Psychiatry*, 56 (1) p.59-65.

[38] Leimeister, J.M., Schweizer, K., Leimeister, S., Krcmar, H. (2008) 'Do virtual communities matter for the social support of patients? Antecedents and effects of virtual relationships in online communities', *Information Technology & People*, 21 (4), p.350-374

[39] Caplan, S.E. (2003) 'Preference for Online Social Interaction: A Theory of Problematic Internet Use and Psychosocial Well-Being', *Communication Research*, 30 (6), p.625-649

[40] Allen, C. Vassilev, I., Lin, S., Culliford, D., Rogers, A. (2018) 'The contribution of internet use in personal networks of support for long-term condition self-management'. *Chronic Illness*. (0) 1-16.

[41] Munn, Z., Peters, M.D.J., Stern, C., Tufanaru, C., McArthur, A., Aromataris, E. (2018) 'Systematic review or scoping review? Guidance for authors when choosing between a systematic or scoping review approach'. *BMC Medical Research Methodology*. 18: 143.

[42] Daudt, H.ML., van Mossel, C., Scott, S.J. (2013) 'Enhancing the scoping study methodology: a large, inter-professional team's experience with Arksey and O'Malley's framework'. *BMC Medical Research Methodology*, 13:48.

[43] Collins (2022) *'Internet'* Accessed at: www.collinsdictionary.com/dictionary/english/internet

[44] Kaplan, A. M., Haenlein, M., (2010) 'Users of the world, unite! The challenges and opportunities of Social Media, *Business Horizons*, 53 p.59-68.

[45] Bramer, W.M., Rethlefsen, M,L., Kleijnen, J., Franco, O.H., (2017) 'Optimal database combinations for literature searches in systematic reviews: a prospective exploratory study', *Systematic Reviews*, 6 (245).

[46] Pham, M.T., Rajic, A., Greig, J.D., Sargeant, J.M., Papadopoulos, A., McEwan, S.A. (2014) 'A scoping review of scoping reviews: advancing the approach and enhancing the consistency'. *Research Synthesis Methods*. 5 371-385.

[47] McColl, M.A., Shortt, S., Godwin, M., Smith, K., Rowe, K., O'Brien, P/. Donnelly. (2009) 'Models for integrating rehabilitation and primary care: a scoping review'. *Archives of Physical Medicine and Rehabilitation*. 90(9): 1523-1531.

[48] Brien, S.E., Lorenzetti, D.L., Lewis, S., Kennedy, J., Ghsli, W.A. (2010) 'Overview of a formal scoping review on health system report cards'. *Implementation Science*. 5 (1): 2

[49] Grant, M., Booth, A. (2009) 'A typology of reviews: an analysis of 14 review types and associated methodologies'. *Health Info Libr J*.26 920: 91-108

[50] Critical Appraisal Skills Programme (2018) CASP Qualitative Checklist. Available at: https://casp-uk.b-cdn.net/wp-content/uploads/2018/03/CASP-Qualitative-Checklist-2018_fillable_form.pdf. (Accessed 9th August 2022).

[51] Mixed Methods Appraisal Tool (2018) MMAT Mixed Methods Checklist. Available at: http://mixedmethodsappraisaltoolpublic.pbworks.com/w/file/fetch/127916259/MMAT_2018_criteria-manual_2018-08-01_ENG.pdf (Accessed 6th December 2023).

[52] Joanna Briggs Institute (2020) Critical Appraisal Tools. Available at: https://jbi.global/sites/default/files/2019-05/JBI_Critical_Appraisal-Checklist_for_Analytical_Cross_Sectional_Studies2017_0.pdf (Accessed 6th December 2023).

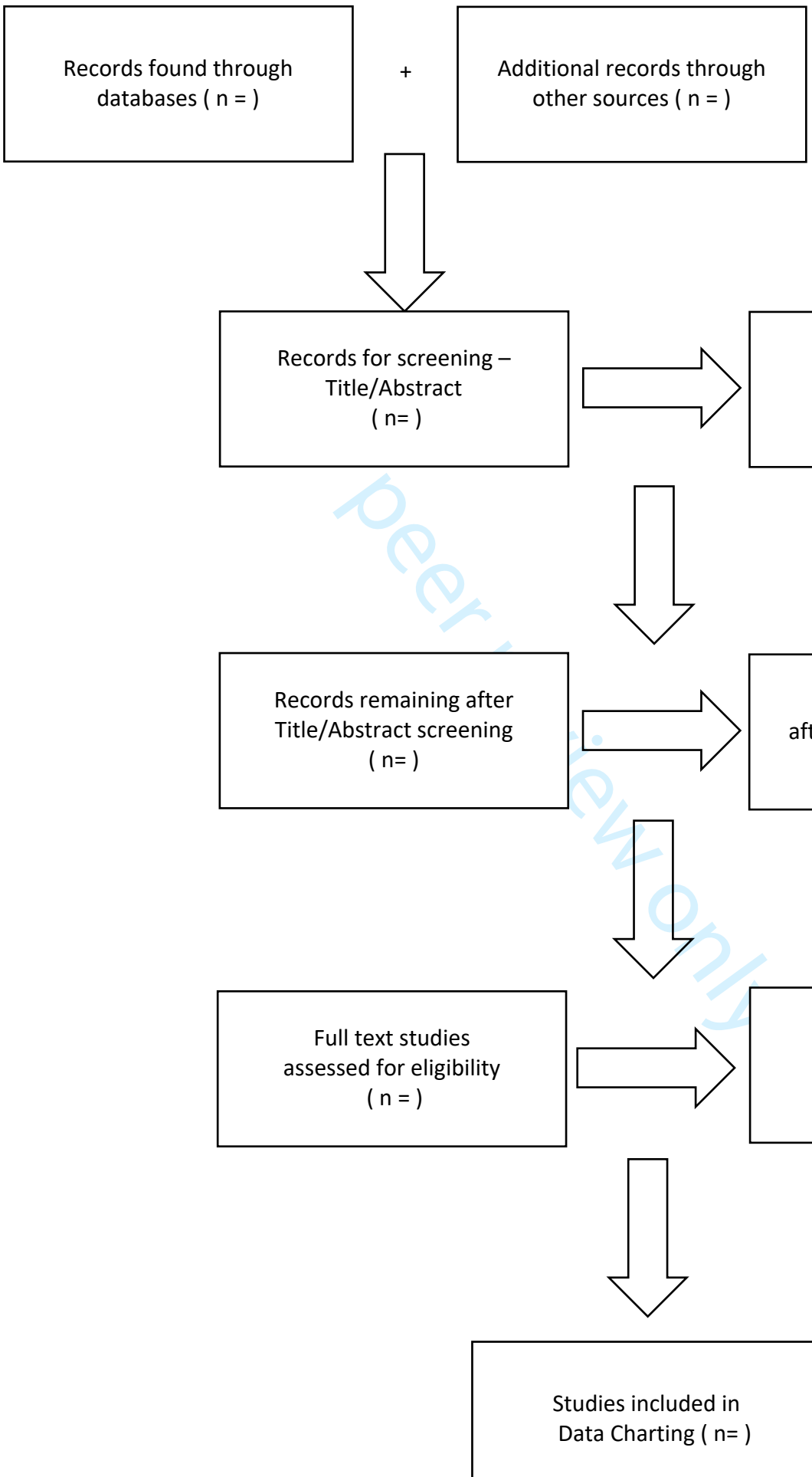
[53] Braun, V., Clarke, V. (2006) 'Using thematic analysis in psychology'. *Qualitative Research Psychology*. 3: 77-101.

[54] Tong, A., Remming, K., McInnes, E., Oliver, S/, Craig, J. (2012) 'Enhancing transparency in reporting the synthesis of qualitative research: ENTREQ'. BMC Medical Research Methodology. 12: 181.

FULL TEXT DATA EXTRACTION

Study ID ……	Author ……	Year …….	Assessor Initials …….	Date Assessed ……
Population: Adults ☐		Identified as having ME/CFS? ☐		Concept: Personal internet use? ☐
Institutional use? ☐		Country …….	Date of study conducted …	Peer reviewed ……
Study aims:				
Study design and setting:				
No. of participants in the study:				
Characteristics of population:				
Study's inclusion criteria:				
Online usage information:				
Data collection and analysis methods:				
Study findings:				
Included as full text	Yes ☐ No. ☐		Exclusion reason:	
Author contact details: RESEARCHER COMMENTS:				

PRISMA Flow Chart



www.casp-uk.net

info@casp-uk.net

Summertown Pavilion, Middle Way Oxford OX2 7LG

CASP Checklist: 10 questions to help you make sense of a Qualitative research

How to use this appraisal tool: Three broad issues need to be considered when appraising a qualitative study:

- Are the results of the study valid? (Section A)
- What are the results? (Section B)
- Will the results help locally? (Section C)

The 10 questions on the following pages are designed to help you think about these issues systematically. The first two questions are screening questions and can be answered quickly. If the answer to both is "yes", it is worth proceeding with the remaining questions. There is some degree of overlap between the questions, you are asked to record a "yes", "no" or "can't tell" to most of the questions. A number of italicised prompts are given after each question. These are designed to remind you why the question is important. Record your reasons for your answers in the spaces provided.

About: These checklists were designed to be used as educational pedagogic tools, as part of a workshop setting, therefore we do not suggest a scoring system. The core CASP checklists (randomised controlled trial & systematic review) were based on JAMA 'Users' guides to the medical literature 1994 (adapted from Guyatt GH, Sackett DL, and Cook DJ), and piloted with health care practitioners.

For each new checklist, a group of experts were assembled to develop and pilot the checklist and the workshop format with which it would be used. Over the years overall adjustments have been made to the format, but a recent survey of checklist users reiterated that the basic format continues to be useful and appropriate.

Referencing: we recommend using the Harvard style citation, i.e.: *Critical Appraisal Skills Programme (2018). CASP (insert name of checklist i.e. Qualitative) Checklist. [online] Available at: URL. Accessed: Date Accessed.*

CASP Critical Appraisal Skills Programme

Paper for appraisal and reference:

Section A: Are the results valid?

1. Was there a clear statement of the aims of the research?

Yes ☐ Can't Tell ☐ No ☐

HINT: Consider
- what was the goal of the research
- why it was thought important
- its relevance

Comments:

2. Is a qualitative methodology appropriate?

Yes ☐ Can't Tell ☐ No ☐

HINT: Consider
- If the research seeks to interpret or illuminate the actions and/or subjective experiences of research participants
- Is qualitative research the right methodology for addressing the research goal

Comments:

Is it worth continuing?

3. Was the research design appropriate to address the aims of the research?

Yes ☐ Can't Tell ☐ No ☐

HINT: Consider
- if the researcher has justified the research design (e.g. have they discussed how they decided which method to use)

Comments:

CASP Critical Appraisal Skills Programme

4. Was the recruitment strategy appropriate to the aims of the research?

Yes ☐
Can't Tell ☐
No ☐

HINT: Consider
- If the researcher has explained how the participants were selected
- If they explained why the participants they selected were the most appropriate to provide access to the type of knowledge sought by the study
- If there are any discussions around recruitment (e.g. why some people chose not to take part)

Comments:

5. Was the data collected in a way that addressed the research issue?

Yes ☐
Can't Tell ☐
No ☐

HINT: Consider
- If the setting for the data collection was justified
- If it is clear how data were collected (e.g. focus group, semi-structured interview etc.)
- If the researcher has justified the methods chosen
- If the researcher has made the methods explicit (e.g. for interview method, is there an indication of how interviews are conducted, or did they use a topic guide)
- If methods were modified during the study. If so, has the researcher explained how and why
- If the form of data is clear (e.g. tape recordings, video material, notes etc.)
- If the researcher has discussed saturation of data

Comments:

CASP Critical Appraisal Skills Programme

6. Has the relationship between researcher and participants been adequately considered?

Yes ☐
Can't Tell ☐
No ☐

HINT: Consider
- If the researcher critically examined their own role, potential bias and influence during (a) formulation of the research questions (b) data collection, including sample recruitment and choice of location
- How the researcher responded to events during the study and whether they considered the implications of any changes in the research design

Comments:

Section B: What are the results?

7. Have ethical issues been taken into consideration?

Yes ☐
Can't Tell ☐
No ☐

HINT: Consider
- If there are sufficient details of how the research was explained to participants for the reader to assess whether ethical standards were maintained
- If the researcher has discussed issues raised by the study (e.g. issues around informed consent or confidentiality or how they have handled the effects of the study on the participants during and after the study)
- If approval has been sought from the ethics committee

Comments:

CASP Critical Appraisal Skills Programme

8. Was the data analysis sufficiently rigorous?

Yes ☐
Can't Tell ☐
No ☐

HINT: Consider
- If there is an in-depth description of the analysis process
- If thematic analysis is used. If so, is it clear how the categories/themes were derived from the data
- Whether the researcher explains how the data presented were selected from the original sample to demonstrate the analysis process
- If sufficient data are presented to support the findings
- To what extent contradictory data are taken into account
- Whether the researcher critically examined their own role, potential bias and influence during analysis and selection of data for presentation

Comments:

9. Is there a clear statement of findings?

Yes ☐
Can't Tell ☐
No ☐

HINT: Consider whether
- If the findings are explicit
- If there is adequate discussion of the evidence both for and against the researcher's arguments
- If the researcher has discussed the credibility of their findings (e.g. triangulation, respondent validation, more than one analyst)
- If the findings are discussed in relation to the original research question

Comments:

CASP Critical Appraisal Skills Programme

Section C: Will the results help locally?

10. How valuable is the research?

HINT: Consider

- If the researcher discusses the contribution the study makes to existing knowledge or understanding (e.g. do they consider the findings in relation to current practice or policy, or relevant research-based literature
- If they identify new areas where research is necessary
- If the researchers have discussed whether or how the findings can be transferred to other populations or considered other ways the research may be used

Comments:

Part I: Mixed Methods Appraisal Tool (MMAT), version 2018

Category of study designs	Methodological quality criteria	Responses			
		Yes	No	Can't tell	Comments
Screening questions (for all types)	S1. Are there clear research questions?				
	S2. Do the collected data allow to address the research questions?				
	Further appraisal may not be feasible or appropriate when the answer is 'No' or 'Can't tell' to one or both screening questions.				
1. Qualitative	1.1. Is the qualitative approach appropriate to answer the research question?				
	1.2. Are the qualitative data collection methods adequate to address the research question?				
	1.3. Are the findings adequately derived from the data?				
	1.4. Is the interpretation of results sufficiently substantiated by data?				
	1.5. Is there coherence between qualitative data sources, collection, analysis and interpretation?				
2. Quantitative randomized controlled trials	2.1. Is randomization appropriately performed?				
	2.2. Are the groups comparable at baseline?				
	2.3. Are there complete outcome data?				
	2.4. Are outcome assessors blinded to the intervention provided?				
	2.5 Did the participants adhere to the assigned intervention?				
3. Quantitative non-randomized	3.1. Are the participants representative of the target population?				
	3.2. Are measurements appropriate regarding both the outcome and intervention (or exposure)?				
	3.3. Are there complete outcome data?				
	3.4. Are the confounders accounted for in the design and analysis?				
	3.5. During the study period, is the intervention administered (or exposure occurred) as intended?				
4. Quantitative descriptive	4.1. Is the sampling strategy relevant to address the research question?				
	4.2. Is the sample representative of the target population?				
	4.3. Are the measurements appropriate?				
	4.4. Is the risk of nonresponse bias low?				
	4.5. Is the statistical analysis appropriate to answer the research question?				
5. Mixed methods	5.1. Is there an adequate rationale for using a mixed methods design to address the research question?				
	5.2. Are the different components of the study effectively integrated to answer the research question?				
	5.3. Are the outputs of the integration of qualitative and quantitative components adequately interpreted?				
	5.4. Are divergences and inconsistencies between quantitative and qualitative results adequately addressed?				
	5.5. Do the different components of the study adhere to the quality criteria of each tradition of the methods involved?				

Hong QN, Pluye P, Fàbregues S, Bartlett G, Boardman F, Cargo M, Dagenais P, Gagnon M-P, Griffiths F, Nicolau B, O'Cathain A, Rousseau M-C, Vedel I. Mixed Methods Appraisal Tool (MMAT), version 2018. Registration of Copyright (#1148552), Canadian Intellectual Property Office, Industry Canada.

CHECKLIST FOR ANALYTICAL CROSS SECTIONAL STUDIES

Critical Appraisal tools for use in JBI Systematic Reviews

INTRODUCTION

JBI is an JBI is an international research organisation based in the Faculty of Health and Medical Sciences at the University of Adelaide, South Australia. JBI develops and delivers unique evidence-based information, software, education and training designed to improve healthcare practice and health outcomes. With over 70 Collaborating Entities, servicing over 90 countries, JBI is a recognised global leader in evidence-based healthcare.

JBI Systematic Reviews

The core of evidence synthesis is the systematic review of literature of a particular intervention, condition or issue. The systematic review is essentially an analysis of the available literature (that is, evidence) and a judgment of the effectiveness or otherwise of a practice, involving a series of complex steps. JBI takes a particular view on what counts as evidence and the methods utilised to synthesise those different types of evidence. In line with this broader view of evidence, JBI has developed theories, methodologies and rigorous processes for the critical appraisal and synthesis of these diverse forms of evidence in order to aid in clinical decision-making in healthcare. There now exists JBI guidance for conducting reviews of effectiveness research, qualitative research, prevalence/incidence, etiology/risk, economic evaluations, text/opinion, diagnostic test accuracy, mixed-methods, umbrella reviews and scoping reviews. Further information regarding JBI systematic reviews can be found in the JBI Evidence Synthesis Manual.

JBI Critical Appraisal Tools

All systematic reviews incorporate a process of critique or appraisal of the research evidence. The purpose of this appraisal is to assess the methodological quality of a study and to determine the extent to which a study has addressed the possibility of bias in its design, conduct and analysis. All papers selected for inclusion in the systematic review (that is – those that meet the inclusion criteria described in the protocol) need to be subjected to rigorous appraisal by two critical appraisers. The results of this appraisal can then be used to inform synthesis and interpretation of the results of the study. JBI Critical appraisal tools have been developed by the JBI and collaborators and approved by the JBI Scientific Committee following extensive peer review. Although designed for use in systematic reviews, JBI critical appraisal tools can also be used when creating Critically Appraised Topics (CAT), in journal clubs and as an educational tool.

JBI CRITICAL APPRAISAL CHECKLIST FOR ANALYTICAL CROSS SECTIONAL STUDIES

Reviewer_____Date_____

Author_____Year_____ Record Number_____

	Yes	No	Unclear	Not applicable
1. Were the criteria for inclusion in the sample clearly defined?	☐	☐	☐	☐
2. Were the study subjects and the setting described in detail?	☐	☐	☐	☐
3. Was the exposure measured in a valid and reliable way?	☐	☐	☐	☐
4. Were objective, standard criteria used for measurement of the condition?	☐	☐	☐	☐
5. Were confounding factors identified?	☐	☐	☐	☐
6. Were strategies to deal with confounding factors stated?	☐	☐	☐	☐
7. Were the outcomes measured in a valid and reliable way?	☐	☐	☐	☐
8. Was appropriate statistical analysis used?	☐	☐	☐	☐

Overall appraisal: Include ☐ Exclude ☐ Seek further info ☐

Comments (Including reason for exclusion)

_____

_____

_____

EXPLANATION OF ANALYTICAL CROSS SECTIONAL STUDIES CRITICAL APPRAISAL

How to cite: Moola S, Munn Z, Tufanaru C, Aromataris E, Sears K, Sfetcu R, Currie M, Qureshi R, Mattis P, Lisy K, Mu P-F. Chapter 7: Systematic reviews of etiology and risk . In: Aromataris E, Munn Z (Editors). *JBI Manual for Evidence Synthesis.* JBI, 2020. Available from https://synthesismanual.jbi.global

Analytical cross sectional studies Critical Appraisal Tool

Answers: Yes, No, Unclear or Not/Applicable

1. Were the criteria for inclusion in the sample clearly defined?

The authors should provide clear inclusion and exclusion criteria that they developed prior to recruitment of the study participants. The inclusion/exclusion criteria should be specified (e.g., risk, stage of disease progression) with sufficient detail and all the necessary information critical to the study.

2. Were the study subjects and the setting described in detail?

The study sample should be described in sufficient detail so that other researchers can determine if it is comparable to the population of interest to them. The authors should provide a clear description of the population from which the study participants were selected or recruited, including demographics, location, and time period.

3. Was the exposure measured in a valid and reliable way?

The study should clearly describe the method of measurement of exposure. Assessing validity requires that a 'gold standard' is available to which the measure can be compared. The validity of exposure measurement usually relates to whether a current measure is appropriate or whether a measure of past exposure is needed.

Reliability refers to the processes included in an epidemiological study to check repeatability of measurements of the exposures. These usually include intra-observer reliability and inter-observer reliability.

4. Were objective, standard criteria used for measurement of the condition?

It is useful to determine if patients were included in the study based on either a specified diagnosis or definition. This is more likely to decrease the risk of bias. Characteristics are another useful approach to matching groups, and studies that did not use specified diagnostic methods or definitions should provide evidence on matching by key characteristics

5. Were confounding factors identified?

Confounding has occurred where the estimated intervention exposure effect is biased by the presence of some difference between the comparison groups (apart from the exposure investigated/of interest). Typical confounders include baseline characteristics, prognostic factors, or concomitant exposures (e.g. smoking). A confounder is a difference between the comparison groups and it influences the direction of the study results. A high quality study at the level of cohort design will identify the potential confounders and measure them (where possible). This is difficult for studies where behavioral, attitudinal or lifestyle factors may impact on the results.

6. Were strategies to deal with confounding factors stated?

Strategies to deal with effects of confounding factors may be dealt within the study design or in data analysis. By matching or stratifying sampling of participants, effects of confounding factors can be adjusted for. When dealing with adjustment in data analysis, assess the statistics used in the study. Most will be some form of multivariate regression analysis to account for the confounding factors measured.

7. Were the outcomes measured in a valid and reliable way?

Read the methods section of the paper. If for e.g. lung cancer is assessed based on existing definitions or diagnostic criteria, then the answer to this question is likely to be yes. If lung cancer is assessed using observer reported, or self-reported scales, the risk of over- or under-reporting is increased, and objectivity is compromised. Importantly, determine if the measurement tools used were validated instruments as this has a significant impact on outcome assessment validity.

Having established the objectivity of the outcome measurement (e.g. lung cancer) instrument, it's important to establish how the measurement was conducted. Were those involved in collecting data trained or educated in the use of the instrument/s? (e.g. radiographers). If there was more than one data collector, were they similar in terms of level of education, clinical or research experience, or level of responsibility in the piece of research being appraised?

8. Was appropriate statistical analysis used?

As with any consideration of statistical analysis, consideration should be given to whether there was a more appropriate alternate statistical method that could have been used. The methods section should be detailed enough for reviewers to identify which analytical techniques were used (in particular, regression or stratification) and how specific confounders were measured.

For studies utilizing regression analysis, it is useful to identify if the study identified which variables were included and how they related to the outcome. If stratification was the analytical approach used, were the strata of analysis defined by the specified variables? Additionally, it is also important to assess the appropriateness of the analytical strategy in terms of the assumptions associated with the approach as differing methods of analysis are based on differing assumptions about the data and how it will respond.