Seeing the Invisible: A Photovoice exploration of living with, and managing the Invisible Symptoms of Multiple Sclerosis

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Thesis Abstract

Background: Multiple Sclerosis (MS) is a chronic inflammatory disease of the central nervous system, characterised by an abnormal immune system response which causes axonal demyelination. MS presents a range of debilitating cognitive, sensory, motor and affective symptoms. Some of the symptoms of MS are classified as ‘invisible’, due to the fact that they are not easily observable to others and are ‘hidden’. Common invisible symptoms include cognitive impairment, fatigue, pain, bladder and bowel dysfunction, sexual problems, and sensory dysfunction. Research shows that living with invisible symptoms causes distress for people with MS that differs from that of visible symptoms, not only because of their physical impacts, but because of their invisible nature.

Studies highlight the issues associated with invisible symptoms, whereby people with MS do not feel understood or believed by others in relation to their invisible symptoms and attempt to navigate their difficulties brought about by this. There is a dearth of research offering in-depth exploration of lived experience of a cluster of invisible MS symptoms.

Aims: This research aimed to explore how people with MS experience *living with* and *managing* invisible symptoms in daily life.

Method: Photovoice is a participatory visual research method, in which people are asked to produce their own images in order to share their experiences of a particular aspect of health and illness. Photovoice was used in this study to explore people’s lived experience of MS invisible symptoms. Twelve people with MS produced digital images over a two-week period to capture their experiences of living with and managing their invisible symptoms. Participants discussed their images and respective meanings in semi-structured interviews. The semi-structured interviews were analysed using an Inductive-Deductive Thematic Analysis.

Results: Three main themes and eight subthemes were developed. The first main theme encompasses the difficulties that exist around conceptualising invisible symptoms, not only due to their invisibility, but because people find it difficult to use language to accurately communicate their abstract symptom experiences. The second theme describes the conflicts of legitimacy this presents for people with MS, whereby they struggle to validate the reality of their invisible symptoms and this reality is also invalidated by others. The third theme elucidated how participants navigate these issues in dynamic ways, choosing to fit their symptoms to their lives or make space for their symptoms depending upon the context, often influenced by their desire for their symptoms to ‘stay invisible’ or to ‘be seen’. Choosing how to navigate invisible symptoms places a continuous burden on people with MS.

Discussion: This research provides insight into the challenges faced by people living with invisible MS symptoms, and the ways they negotiate daily life. It is important that clinicians and healthcare professionals are aware of the conflicts created by symptom invisibility so that they can offer appropriate support to people with MS. Further research is suggested to explore determinants of particular navigation styles, and also to explore whether improved psychological flexibility could support people with MS to navigate their invisible symptoms and associated impacts.

Statement of Contribution

I, Le-Sharn Parker, declare that this research is the product of my own original work. As the primary researcher, I was responsible for undertaking and writing the systematic literature review, the planning and design of the research, establishing approved sponsorship from the University of Nottingham, applying for and ensuring all relevant ethical approval for the project, recruitment, data collection and transcription, data analysis, the write-up of the research, and undertaking and writing the small-scale research project.

Professor Roshan das Nair, as Primary Research Supervisor, offered support and guidance in all of the areas specified above. Roshan introduced me to the original research idea and contributed to the subsequent planning and design of the study. Roshan supported with establishing avenues for recruitment and provided guidance on potential ethical issues. Roshan was also involved in data analysis and cross-checking of themes and supported with the study write-up. Roshan supported with the design, analysis and write-up of the systematic literature review and small-scale research project.

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Dr Clare Bale provided advice and feedback around the initial study design, ethical issues and participant-facing materials, analysis of findings and write-up of the research.

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Systematic Literature Review

The notion of ‘invisibility’ in people’s experiences of the symptoms of multiple sclerosis: a systematic meta-synthesis

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Purpose: Invisible symptoms, such as cognitive problems, have a negative impact on people living with Multiple Sclerosis (MS), related to the very notion that they are 'unseen'. It is important to understand the notion of 'invisibility' in MS, as invisible symptoms are particularly distressing, and there is a paucity of research focussing on their invisible nature and its specific impact. We aimed to systematically identify, appraise and synthesise qualitative research regarding the notion of 'invisibility' in relation to people's lived experience of symptoms of MS.

Methods and Materials: Articles meeting inclusion criteria were critically appraised and synthesised using a meta-ethnographic approach.

Results: 17 articles were identified from six electronic databases. Three third-order themes were presented as a *line of argument*. 'Invisibility' was conceptualised by people with MS as a discrepancy between the internal experience of symptoms and what is observed externally. 'Invisibility' of MS symptoms was found to have a number of impacts, including not feeling understood or validated by others, issues around the perceived legitimacy of their illness, and living with needs which are hidden. We found that 'invisibility' by its nature offers people a choice of strategies they use to navigate it. This choice introduces a dilemma: disclose the diagnosis to be 'seen', or remain 'invisible'.

Conclusions: This review uncovered ways in which people with MS are affected by the invisibility of their symptoms, and some of the ways they navigate these in their lives. We highlight the need to better understand and respond to these experiences, both at a clinical level and in the general public. Focussed exploration of people's experiences of 'invisibility' in MS, including the ways in which 'invisibility' is managed on a day-to-day basis could raise clinical and public awareness of the impacts of 'invisibility', easing the dilemmas faced by those with MS.

Keywords: multiple sclerosis; meta-ethnography; meta-synthesis; invisible symptoms; invisibility

Introduction

Multiple Sclerosis (MS) is a chronic disease of the central nervous system, in which an abnormal response of the immune system results in damage to nerve fibres and myelin sheaths [1]. The disease causes cognitive, sensory and motor impairments that significantly impact the individual's life [2]. These impairments can be classified into 'visible' (e.g., motor and mobility problems) and 'invisible' symptoms [3].

The term ‘invisible symptoms of MS’ has been variously defined in the research literature, however there is some agreement that these refer to symptoms that are ‘hidden’ or not visible or overtly seen as debilitating to others [4, 5]. Commonly cited invisible symptoms of MS include fatigue, cognitive impairment (memory loss and problem-solving deficits), pain, bladder and bowel dysfunction, blurred vision, sexual problems, and sensory dysfunction such as numbness or tingling [3,6].

Invisible symptoms of MS have been found to impact the lives of people with MS in a variety of ways, and are described in research papers, patient accounts and journalistic pieces. In a study [7] investigating the relationship between symptom severity and perceptions of health in 1865 outpatients, invisible symptoms of MS (specifically pain, fatigue and cognitive impairment) were predictive of negative health perception, with pain being the most predictive, even compared to ‘visible’ symptoms. The authors concluded that invisible symptoms of MS are as important to the individual’s sense of wellbeing as visible symptoms, and the long-term impact of living with invisible symptoms should not be underestimated. In fact, another study of 145 people with MS [8] found that invisible symptoms were more predictive of health distress than visible symptoms.

‘Research matters’ [4], a periodical published by the UK MS Society for people with MS, released an edition focussed on invisible symptoms of MS, highlighting how these symptoms were distinct in their impact from those that are visible. This was also apparent in ‘But you look so good!’, a patient-produced brochure published by the National MS Society of the USA [9]. While not a research publication, this work covered specific invisible symptoms using a selection of quotes from people with MS and highlighted the psychological distress the ‘invisibility’ of the illness causes them. It emphasised people with MS’ experiences of stigma related to their invisible symptoms, including perceptions of not having a ‘real’ illness, reduced confidence in own perception of symptoms, discouragement from help-seeking, and social isolation.

Despite there being evidence to suggest that invisible symptoms are a particularly distressing aspect of living with MS, and indication in patient-produced materials that this is related to their invisible nature, there is a paucity of empirical research that offers an in-depth understanding of the experience of ‘invisibility’ in MS. In the absence of research that focuses specifically on ‘invisibility’, we can draw out some inferences about this phenomenon based on existing qualitative research that explores people with MS’ lived experiences of specific, commonly cited invisible symptoms.

Commonly cited invisible symptoms that have high prevalence rates in people with MS are fatigue, pain, cognitive impairment, and bladder and bowel dysfunction [4,10]. Cognitive impairment affects up to 60% of people with MS [9] and is a highly debilitating invisible symptom [11]. Fatigue is one of the most researched invisible symptoms of MS, likely due to the detrimental effects it has on daily functioning [12], and it is considered the most common invisible symptom [13]. Pain is reported as a central feature of MS by more than half of patients [14] and over 80% of people with MS experience some degree of bladder dysfunction [15]. Around 50% of people with MS experience neurogenic bowel dysfunction [16], and themes around humiliation and distress have been found in the research relating to this symptom [17].

Given the prevalence of these invisible symptoms in MS, and the fact that experiential accounts of these symptoms or how they are managed in daily life has not been well-researched, we felt it was an important first step to collate the extant research evidence, to uncover the narratives around this concept, in order to determine how researchers and clinicians can better help people manage these invisible symptoms.

Aims

This review aimed to explore the notion of 'invisibility' in relation to people's lived experience of MS symptoms.

Materials and Methods

This meta-synthesis was prospectively registered with PROSPERO International Register of Systematic Reviews (Registration number: CRD42018107183, date 20.08.2018).

Inclusion Criteria

Inclusion criteria were defined *a priori*. Research mainly concerning fatigue, pain, cognitive impairment, and bowel and bladder problems were captured in the review. This decision was based on the high prevalence rates of these symptoms in comparison to other invisible symptoms of MS such as sexual dysfunction [4], and because the impact of these symptoms on people with MS appear to be greater than some other invisible symptoms. For example, over 60% of 4,660 participants in a study classified the impact of their dizziness as mild and not particularly problematic [18]. However, studies focussing on pain, fatigue, cognitive impairment, and bowel and bladder problems have found significant negative impacts on mental health, social roles, quality of life, employment and daily living for people with MS [19,20,21,22,23].

The search strategy, however, was not limited to these symptoms. Articles concerning the general lived experience of MS were also captured so as not to restrict our inferences about 'invisibility' to specific individual symptoms, when we know that people with MS often experience symptoms in clusters [6]. Therefore, articles were included in the review if they:

- Were written in the English language.

- Included adult participants with a diagnosis of MS.
- Were empirical papers that used qualitative methods of data collection and analysis.
- Focussed on participants' lived experience of fatigue, pain, cognitive impairment, or bladder and/or bowel problems, *or* general lived experience of MS.
- Discussed the experience of 'invisibility' in relation to participants' symptoms (individual symptoms or general clusters) of MS. This included any appraisal or acknowledgment by participants or authors that others cannot 'see' participants' MS/symptoms.

Articles that used mixed methods were considered for inclusion provided they had a clear qualitative component, identifiable from their title or abstract. Articles that used participants with different diagnoses were considered provided that it was made explicit which data or outcomes were linked specifically to people with MS, to allow for separate data extraction.

Exclusion criteria

Articles were excluded from the review if they:

- Used the same participant data as any other articles that were already included.
- Were secondary reviews of studies that already met inclusion criteria.

Searching

A systematic search was conducted on Embase, MEDLINE, AMED, PsycINFO, CINAHL and Web of Science from their inception until 3rd May 2019.

Search terms were developed according to recommendations made by Shaw et al. [24]. Subject headings and free-text terms were combined to give a high search yield and to ensure potential articles would not be excluded by the process. The search terms used in Medline are presented in table 1. Equivalent searches were used in each of the other databases to ensure a broad and consistent search.

From the initial search, duplicates were removed using 'RefWorks' software [25]. Titles and abstracts of each article were screened first by LSP to select relevant articles for full text reviews. If insufficient information was provided in an article's title or abstract (or abstract was unavailable), then the full text was reviewed. The reference lists of all included papers were checked by title for relevance by LSP and abstracts obtained where relevance was ambiguous from the title. In instances where relevance or inclusion was uncertain, this was discussed with the rest of the research team who independently reviewed the titles and abstracts, arriving at a decision unanimously. Discrepancies in judgment did not arise, however if the research team had been unable to reach a decision about the inclusion of an article, an external researcher would be consulted.

Epistemology

The review was approached from a critical realist epistemological position, encompassing an assumption that objective knowledge exists, however the ability to access this is tentative and mediated by our perceptions and beliefs [26]. The assumption was made that people's experiences of their invisible symptoms are subjective and constructed, however there is a physical, objective reality to these symptoms. We acknowledge that the studies reviewed contain secondary-level interpretations (made by the researchers) of the first-level original data, which are influenced by the researchers' own biases and experiences. The current review generates a triple hermeneutic, a third-level interpretation which will in turn be influenced by the reviewer's epistemological position and beliefs [27]. A reflexive log was kept by the primary author to note their responses to the data, frequently returning to the review aims throughout the review process.

Table 1. Search strategy used in Medline in review search process

Search step	Search Terms
1	(exp multiple sclerosis/) OR (“multiple sclerosis” OR MS).mp.
2	(exp qualitative research/) OR (qualitative or interview* or focus group* or (grounded adj (theor* or analy*)) or narrative* or theme* or thematic or content analy* or phenomenolog* or conversational analy* or (framework adj (approach or analy* or method*))).mp. OR (experience OR “living with” OR “patient perspective”).mp
3	(exp fatigue/) OR (exp pain/) OR (exp cognition) OR (exp cognitive function) OR (exp urinary bladder/) OR (fatigue or pain or cogniti* or bladder or bowel).mp
4	(invisib* or "invisible symptoms" or hidden or "not visible").mp.
5	3 OR 4
6	1 AND 2 AND 5

Critical appraisal

There appears to be no consensus on suitable methods used for appraisal, or if indeed it is appropriate to appraise qualitative research at all [28]. However, as this review included studies that used different methodologies, we decided to use the Critical Appraisal Skills Programme (CASP) [29] tool to appraise the quality of the articles. The CASP encompasses the considerations recommended by Dixon-Woods et al [30] for appraising qualitative research at a generic level, unspecific to methodology. Sandelowski, Docherty and Emden [31] state that interesting findings of studies in reviews should not be discounted on account of surface mistakes or 'incorrect' terminology. The CASP therefore was not used to exclude studies from the meta-synthesis, but rather to consider any quality issues, and to have an awareness of the way that these issues may distort the synthesis [30].

The 10 quality criteria outlined in the CASP were applied to each article and were marked according to a 'key' (see table 3). Studies were not given a 'score' in relation to quality, as this is not advisable as stated in the CASP.

Judgements about the methodological quality of the studies were made by one author (LSP) and cross checked independently by two other members of the research team (GT and DDB). Levitt, Wertz, Motulsky, Morrow and Ponterotto [32] highlight that it is rare for a single reviewer to have the required expertise to equally critically appraise qualitative research from a range of epistemological positions and methodologies. Thus, multiple reviewers add reliability to the quality appraisals.

Data extraction

A data extraction pro-forma was created to facilitate the identification and organisation of the key characteristics of each included study. Data deemed as relevant to the review aims were direct comments about the symptoms of MS being ‘invisible’ ‘hidden’ or ‘unseen’, and descriptions that were conceptually analogous to ‘invisibility’ in relation to the symptoms of MS. First-order and second-order constructs relating to the notion of ‘invisibility’ in MS were recorded from each study- these constructs are described in table 2 using definitions from Malpass, [27] and based on Schutz’s [33] original concepts. Findings that were not relevant were excluded so as not to compromise fidelity to the aims of the review [34]. Second-order themes that were not accompanied by a supporting quotation were also extracted. This allowed all eligible data in the studies to be included and avoided omission of potentially valuable information from the synthesis [35]. We acknowledge however, that in doing so it we make some interpretations from the interpretations of the authors of the included studies, rather than directly from quotations.

Meta-synthesis

A meta-ethnographic approach was used as this is a leading method for synthesising qualitative research in the healthcare field [36]. The approach was developed by Noblit and Hare [37], who outlined seven steps to this type of synthesis: ‘getting started’, deciding what is relevant, reading the studies, determining how the studies are related, ‘translating’ the studies, synthesising the translations, and expressing the synthesis. This review replicated the specific methods used by Malpass et al. [27]. The process of meta-ethnography seeks to identify second-order constructs and develop from them third-order constructs through a process of ‘translating’ the meanings between studies and identifying similarities and differences. These third-order constructs are new interpretations that aim to extend beyond the findings of the original studies.

Table 2. Levels of constructs in meta-ethnography

First-order	Participants' views, accounts and interpretations of their experiences of 'invisibility' related to their MS symptoms- "Interpretations of experience" [27, p.158].
Second-order	The original study authors' views and interpretations (expressed as themes and concepts) of participants'' experiences of 'invisibility' in related to their MS symptoms- "Interpretations of interpretations of experience" [27, p.158].
Third-order	The views and interpretations of those conducting the synthesis, (expressed in terms of themes and key concepts)- "Interpretations of interpretations of interpretations of experience" [27, p.158].

All papers were read carefully in chronological order and re-read several times for familiarity with the data. First and second-order constructs were extracted from each study and recorded verbatim on the data extraction pro-forma. We recognise that first-order constructs are still influenced by the interpretations of the original authors as they are selected to emphasise their themes [38]. 'Conceptual maps' were created for each study to show how each of the studies' second order themes related to one another [27] and to preserve these contextual meanings. Second-order themes were entered into a matrix to juxtapose the constructs from each study and allow us to later 'translate them into one another' (the process of generating third-order themes whereby we compare and contrast themes from across the articles to identify relationships between them). The original wording from the authors of the studies were used wherever possible to remain faithful to the meanings of each study [39]. However, some reinterpretation was necessary to shape the original authors' themes to the review aims (as some of these captured the notion of 'invisibility' under generic theme labels/ was not the main focus of the theme), as recommended by Levitt [34]. Second-order themes were compared and contrasted across studies in a reciprocal synthesis to generate third-order themes. To facilitate the reciprocal synthesis, the second-order themes in each study were compared to those of others and grouped into broader categories with labels that captured the overall meaning of those themes (second order summaries). We ensured that each secondary concept from each paper was included in one of the second order summaries. During this process, again the original wording from the authors of the studies were used wherever possible to remain faithful to the meanings of each study. A narrative translation of each of the second order summaries was created to encompass the meaning from all the studies they related to. Finally, these translations were synthesised into third-order themes by comparing and contrasting the second order summaries of the included studies, identifying commonalities and differences between them, and extending beyond the original interpretations of the primary study authors. Due to the interpretative and iterative processes involved in developing and naming third order constructs, it can be difficult to describe this in a fully transparent and structured way [40]. LSP demonstrated how the third-order constructs had emerged in a such a way that they could be cross-checked by the other members of the research team.

Results

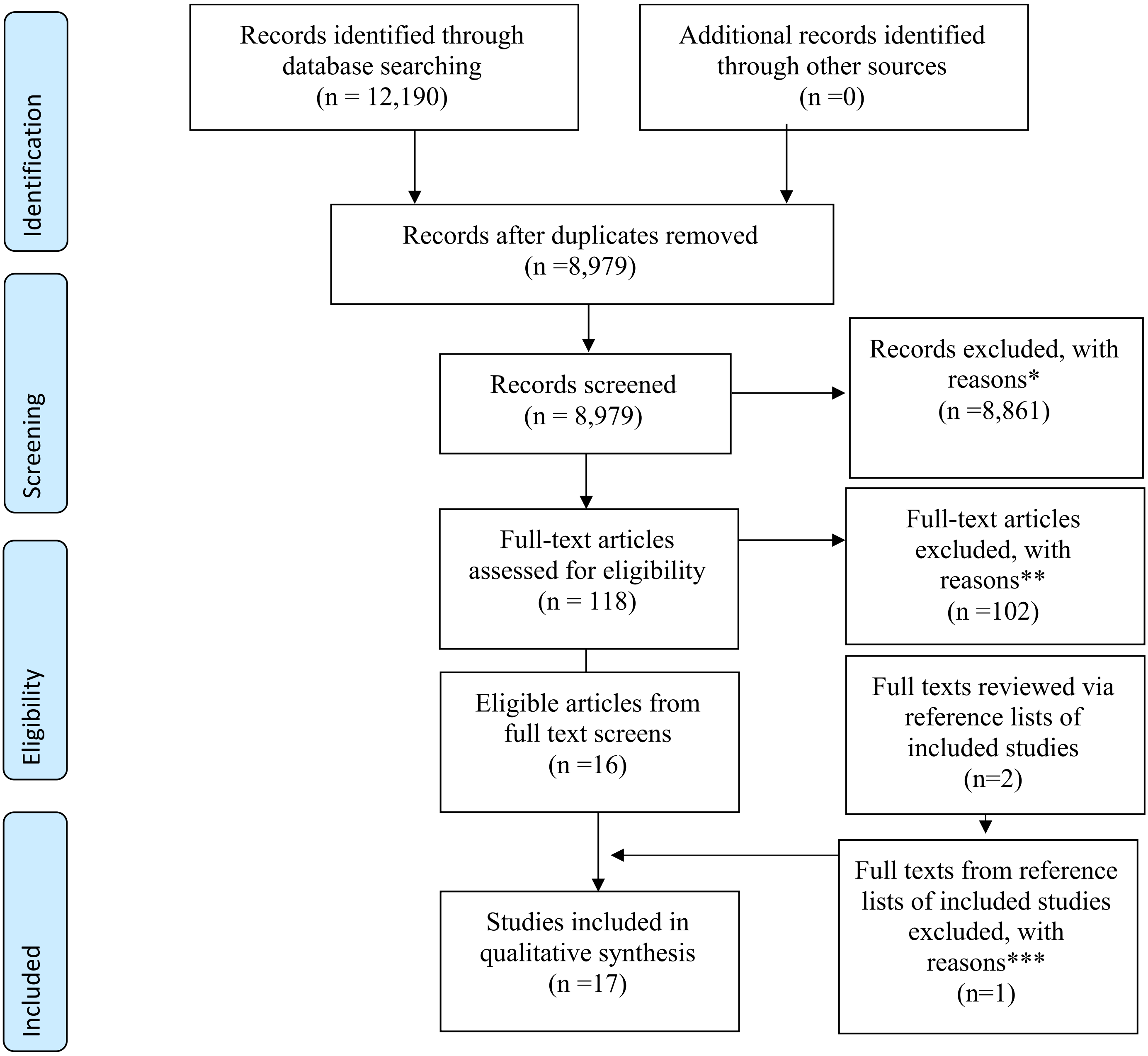
The PRISMA [41] flowchart outlining the search process is presented in figure 1. The searches yielded 12,447 results. Duplicates were removed, leaving 8,979 articles for screening, and 122 articles were identified for full text reading. Four texts identified for full screening remained unavailable after requesting inter-library loans (these were unpublished doctoral dissertations from non-UK institutions) and were therefore excluded. One hundred and eighteen full texts were reviewed against inclusion criteria, and 16 articles included in the review as a result. One additional article was found from searching the reference lists that met the inclusion criteria bringing the total of included papers to 17.

Study Characteristics

Table 3 summarises the general characteristics of the 17 articles included in the review. Articles were arranged chronologically, and each study was assigned an ID number [42-58], by which they will be referred to throughout. Study [55] used a mixed methods approach, and therefore only the qualitative component of this has been tabulated.

All 17 studies explored people's experiences of living with symptoms of MS to varying degrees. Eight studies' [44-46,49,50,52,55,56] focus was on people's lived experiences of specific commonly cited invisible symptoms of MS. Five studies focussed only on fatigue [44,47,52,55,56], one on pain [46], one on cognitive impairment [45], and one on both fatigue and pain [50]. Seven studies focused on various aspects of participants' lived experience of MS and its associated symptoms more generally [42,43,47,48,51,53,54,56,58].

Figure 1. PRISMA flowchart of searching process



** Not written in English language = 35; Not empirical studies using qualitative methods=4447; Not including adults with MS= 3103; Not focussed on lived experience of MS in general or symptoms specified in inclusion criteria=1272; Paper unavailable=4.*

** *Not written in English language = 1; Not empirical studies using qualitative methods=33; Not focussed on lived experience of MS in general or symptoms specified in inclusion criteria= 4; No discussion of invisibility=59; Review of studies already screened and rejected or included=2; conference papers (abstracts only) =3. *** No discussion of invisibility=1.*

Table 3. General Study Characteristics extracted from included articles

Study Number	Authors (year), Location	Aims	Sample Composition	Qualitative data collection method	Method of data analysis
[42]	Fitzgerald & Paterson (1995) Australia	Not explicitly stated Draws on the narratives of a small group of women with "hidden" MS.	15 women with MS and no obvious outward signs of their illness Age range: 33-56	SSI	Narrative approach but not specified
[43]	Dyck & Jongbloed (2000) British Columbia, Canada	To investigate the everyday work experiences of women diagnosed with MS and the contextual framing of such experience.	31 women with MS and in paid work Age range: 25-49	SSI	Constant comparative analysis
[44]	Olsson, Lexell & Soderberg (2005) Sweden	To elucidate the meaning of fatigue for women with MS.	10 adults with MS (all women) Age range: 43-59	Narrative interviews	Phenomenological hermeneutic interpretation
[45]	Shevil & Finlyason (2006) Location not stated	To explore the impact of cognitive changes on the lives of four individuals with MS with different life roles (worker, parent, retired older adult), and how they interpret and apply meaning to their experience of cognitive change.	4 adults with MS and self-reported cognitive changes (gender not specified) One full time worker; one part one part-time worker; a parent and a retired older adult.	Multiple SSIs (3 per participant at 6-9 week intervals). Modified version of the Experience Sampling Method (called cognitive experience forms). 16 completed per participant across study course	Thematic analysis
[46]	Douglas, Windsor & Wollin (2008) Queensland, Australia	Explore the experiences and perceptions of pain among people with MS.	32 adults with MS related pain (8 men, 24 women) Age range: 26-72	Focus Groups	Thematic analysis

Study Number	Authors (year), Location	Aims	Sample Composition	Qualitative data collection method	Method of data analysis
[47]	Vickers (2009) Australia	To explore the life and work of people with MS.	21 adults with MS who were either working part or full time, or had worked full-time in the past	Interviews	Heideggerian Phenomenology
[48]	Lohne, Aasgaard, Caspari, Slettebo & Naden (2010) Norway	To find out how persons suffering with MS experience and understand dignity and violation in the context of a rehabilitation ward.	14 adults with MS (6 men, 8 women) Age range: 39-66	SSI	Inductive hermeneutic analysis
[49]	Moriya & Kutsumi (2010) Japan	To obtain descriptions of the experiences of fatigue of people with multiple sclerosis, including experiences related to their interpersonal relations and social life.	9 adults with MS who experience fatigue (3 men, 6 women). Age range: 31-57	SSI	Not specified but process described indicates thematic analysis
[50]	Yorkston,Johnson, Boesflug, Skala & Amtmann (2010) Washington, USA	To examine the experiences of people with chronic disabling conditions as they communicate with health care providers about pain and fatigue, and to examine different ways in which living with pain and fatigue impacts the lives of people with chronic health conditions.	23 adults with chronic disabling conditions (5 men, 18 women) Age range:26-60 Of this sample, 5 with MS diagnosis (gender and ages not reported)	Focus groups	Not specified but process described indicates thematic analysis

Study Number	Authors (year), Location	Aims	Sample Composition	Qualitative data collection method	Method of data analysis
[51]	Olsson, Skar & Soderberg (2011) Sweden	To elucidate meanings of being received and met by others as experienced by women with MS	15 women with MS diagnosis Age range: 35-70 All with SPMS	Narrative Interviews	Phenomenological hermeneutic interpretation
[52]	Smith, Olson, Hale & Baxter (2011) New Zealand	To describe the experiences of people with MS-related fatigue, who engaged in community-based exercise activities in order to discover how fatigue influenced their exercise participation.	9 women with MS who experienced a decline in energy levels since diagnosis, and exercised within their community a minimum of once a week over the previous two months. Age Range: 28-70 3 had SPMS; 5 RRMS;	SSI (in person and over phone)	Interpretive description
[53]	Blundell Jones & Walsh (2014) UK	To develop an understanding of the lived experience of emotions in MS, to reveal how feelings are coped with and understood, and to uncover factors influencing help-seeking for emotional difficulties.	10 women with MS Age range: 30-65 7 with RRMS; 1 with SPMS; 1 SPMS with relapse; 1 unsure.	SSI	Interpretative Phenomenological Analysis

Study Number	Authors (year), Location	Aims	Sample Composition	Qualitative data collection method	Method of data analysis
[54]	Pretorius & Joubert (2014) South Africa	To explore the personal experiences of individuals with MS in the South African context, with a special focus on the challenges faced by individuals living with MS and the support and/or resources that help these individuals cope with their condition.	10 adults with MS diagnosis (3 men, 7 women). Age Range: 38-71 4 with RRMS; one with PPMS; 5 with SPMS.	SSI	Thematic analysis
[55]	Wendebourg et al (2016) Germany	To assess people's interest in and need for fatigue self-management training and to develop a cognitive-behavioural group intervention based on this.	16 adults with MS and fatigue (3 men, 13 women)	Interviews (4 participants) Focus groups (12 participants)	Inductive but not specified
[56]	Turpin et al (2018) Queensland, Australia	To explore how people became aware of and understood their MS fatigue, and how they accommodate it in their daily lives	13 adults with MS fatigue (2 men, 11 women) Age range- 25-67 11 RMMS; one with PPMS and one with SPMS.	SSI	Thematic analysis

Study Number	Authors (year), Location	Aims	Sample Composition	Qualitative data collection method	Method of data analysis
[57]	van der Meide, van Gorp, van der Hiele & Visser (2018) The Netherlands	To gain insight into the meaning of work in the everyday lives of people with RRMS, and barriers/facilitators to staying in work.	19 adults with RRMS (11 women, 6 men, 2 unspecified) Age range: 29-55	SSI (Narrative)	Thematic Analysis
[58]	van der Meide, Teunissen, Collard, Visse & Visser (2018) The Netherlands	To explore how people who had been diagnosed with MS for some time, experience their body in daily life.	13 women with RRMS	SSI	Phenomenological Analysis

The total number of participants across studies was 246; 183 of these were women, 31 were men, and gender was not specified for 32 participants. Seven studies [42-44,51-53, 58] only had women in their samples. Sample sizes ranged from 4-32 participants, and ages of participants ranged from 25-72, although four studies [45,47,55,58] did not report participant age. The studies were conducted between 1995-2018. The majority of the studies did not report participants' ethnicity; therefore, it is uncertain how culturally diverse these samples were.

Quality Appraisal

Quality appraisals for each study are displayed in table 4. The four articles [44,53,54, 58] that were deemed the highest quality according to the CASP [30] met all 10 criteria. Overall, study quality was good, and the majority of the studies fulfilled at least nine criteria. Study [42] met the fewest criteria, and thus can be considered the lowest quality article.

All studies but one [42] clearly reported their aims, which were all varied but made reference to some aspect of people's lived experience of MS. All used appropriate samples, however offered varying levels of detail about the justification for these samples. Interestingly, the majority of studies did not acknowledge the potential role of researcher bias/perspectives or comment on how this was managed. Two studies [42,47] did not sufficiently detail their analysis or how their themes were generated, despite including quotes to support these themes. Although all studies made clear statements in relation to their findings, two [42,47] did not discuss the credibility of these findings and so were deemed to have only partially met this criterion.

Table 4. Quality Appraisal of included articles using CASP [30]

Study Number	42	43	44	45	46	47	48	49	50	51	52	53	54	55	56	57	58
Quality Criteria																	
1. A clear statement of the aims of the research	N	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
2. Appropriate qualitative methodology	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
3. Research design appropriate to address aims	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
4. Recruitment strategy appropriate to aims	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
5. Data collection clear and in line with aims	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
6.Critical examination of researcher role and potential for bias	N	N	Y	N	N	Y	N	N	N	P	N	Y	Y	N	N	N	Y
7. Consideration of ethical issues	N	N	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
8. Rigorous and clear data analysis	N	P	Y	Y	P	N	P	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
9.Clear statement of findings	P	Y	Y	Y	Y	P	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
10. Discussion of contribution of research to existing knowledge	Y	Y	Y	Y	Y	P	N	N	Y	Y	Y	Y	Y	Y	Y	Y	Y

Synthesis and line of argument

Three main third-order and seven sub-themes emerged from the extracted data. The synthesis was led by the main author (LSP) with independent input from the other reviewers in the team. Forty-three extracted secondary-constructs were categorised thematically into 13 second-order summaries, from which the third-order themes were developed. Third-order themes are presented in table 5, and studies supporting and contributing to these themes are highlighted. Below, the third-order themes are presented sequentially to construct a *line of argument* [36]. The included papers captured different aspects of the notion of ‘invisibility’ that could be brought together to gain an overarching understanding of this phenomenon.

Table 5. Third-order themes and sub-themes developed through synthesis

Third-Order theme	Study Number																
Subtheme	42	43	44	45	46	47	48	49	50	51	52	53	54	55	56	57	58
The notion of 'invisibility'																	
Looking Healthy, feeling ill	*	*	*	*	*	*	*	*	*	*	*	*	*	*	*	*	*
The impact of invisibility																	
Issues of legitimacy	*			*	*				*	*	*		*		*		*
Hidden Needs							*				*						
Others don't understand	*		*	*	*	*		*		*		*	*	*	*	*	*
Emotional impact	*					*	*	*				*				*	
The burden of choice- a continuous conflict																	
Invisibility offers a choice	*	*			*							*					*
Choosing to stay invisible	*	*			*							*			*		
Struggling to be seen	*	*			*				*			*			*	*	

** represents first/ second order constructs of study contributed to theme*

The notion of 'invisibility': Looking healthy, feeling ill

In line with our inclusion criteria, participants in all included studies described the notion of 'invisibility' as being characterised by a discrepancy between their outward appearances (described as "normal", "robust" and "healthy"; [42, p.16]), and what they experienced internally as a result of their symptoms. Specific symptoms that were recognised and discussed in terms of their 'invisibility' were fatigue [42,44,48-52,55,56,57,58], pain [46,48,50,51], cognitive changes/impairments [45,48], dizziness [48,51], numbness [47], bladder problems [47], loss of vision [47], and temperature sensitivity [47]. 'Invisibility' in MS in a more general sense was also discussed in terms of lived experience of a cluster of symptoms for which there is no external, physical evidence and thus are 'unseen' by others [42,43,51,53,54].

People's perception that their MS symptoms are undetectable to others was highlighted as one of, if not the most distressing part of their MS:

> The most negative thing about MS [long pause]: that it is hidden. [47,p.16].
>
> I don't really look ill, and that's really the worst thing about this [illness]. [48,p.305].

The expressed wish of those with invisible symptoms to have their experiences fully conveyed is captured in the quote below:

> It doesn't show on the outside . . . still you feel really ill ... it is almost as if you want to shout ... can't you see how ill I feel ... [51,p.4].

The impact of 'invisibility'

Sixteen studies contributed to this theme. 'Invisibility' related to the symptoms of MS negatively impacted on individuals in a number of ways, captured in sub-themes of: issues of legitimacy, hidden needs, others don't understand, and emotional impact.

Due to the invisible nature of their symptoms, most people with MS experienced being disbelieved or dismissed by friends, family, colleagues and healthcare professionals when it came to their symptoms:

> You only have to break your arm or your leg and you've got a cast on it which people can see…and yet they say to me: 'What's wrong with you?' It's like the saying 'What you can't see, you don't sort of believe and for me to say: 'I really have had a dreadful day today' [people reply] 'Oh get out, you look all right to me.' [42,p.14].

The issue of feeling delegitimized by others ranged from people with MS feeling that others completely disbelieved them and did not accept the existence of their symptoms at all [42,45,46,50,51,54,56], to feeling that even though others acknowledged the reality of their symptoms, they would still not afford people with MS the allowances or understanding that they would to someone with visible symptoms of illness [42,44-47,49,51, 53-56].

Experiences with healthcare professionals and providers were described as difficult and invalidating [42,46,50,52,55,57]. People with MS recalled being told (or believing the professional was thinking) "you're exaggerating" and "it's imaginary" [50, p.247]. One participant commented "it's like you're making it up" [50, p.247].

The quote below demonstrates a recurring discussion in many of the included papers [44-46,49,56], where symptom-related behaviours were attributed to character flaws, choice, or 'making excuses' because of a lack of tangible evidence for the symptom:

> Not even my husband understands me. He thinks I'm just lazy [55,p.134].

A lack of understanding and flexibility from others in relation to invisible symptoms created strain in the relationships of some of the people with MS [45], and for some, social interaction decreased as a result of not feeling understood [49]. Some family members forgot that people with MS were living with difficult symptoms, due to the absence of visual reminders [56]. Participants expressed that even when others made attempts to empathise with their experiences, what could often result was the perceived minimisation of their symptoms, which compounded their sense of 'invisibility':

> And they sort of say, 'Oh yes, I know, I get so tired'. And I think, 'How annoying. You know, no you don't, YOU DON'T!' [56, p.86].

Embedded within issues of legitimacy was a fear of a delegitimized self, where people with MS perceived a likelihood that they would be disbelieved and invalidated by others, even if they had not directly experienced this [42,43,48,54,56,58]:

> maybe . . . this could be mixed up with laziness . . . it is not what this is about. . . but, maybe it may look like . . . now, she is sick, really and therefore she might blame everything on that [the illness], right? [48,p.306].

One participant expressed that she doubted her son's and co-worker's beliefs in her:

> I honestly think that they think I am just telling a story. I don't know. I just don't believe that some of them believe I have MS [42,p.14].

It was not only the actual experience of the negative reactions of others that was problematic for people with MS, but the expectation that people will respond to them (either publicly of privately) negatively due to the invisible nature of their symptoms. One participant described her experience on a crowded bus, and her perception that she couldn't ask others for help due to the invisibility of her symptoms, indicating a belief that she would not be believed or responded to:

> Suddenly I was surprised by fatigue but I couldn't ask someone to stood up for me because I looked completely normal [58, p.2242].

Many people with MS expressed doubts about their own legitimacy in response to the reactions they received from others [42,51,56]. This participant reflected on her experience of medical investigations into her symptoms:

> Once you've tried…to work out why you're feeling how you're feeling and it all comes back – 'you're a young, fit, healthy woman, there's nothing wrong with you, go and enjoy life' …you start thinking, 'Well if the doctors can't physically find anything, maybe I am going a bit mad'. [56, p.84].

People with MS discussed comparing themselves and their invisible symptoms to those who were more obviously disabled, believing that these people were treated with more sympathy and validation, and visual indications of illness were more legitimizing [42,46,51,52,54,56]. Some also viewed the experience of being discredited due to 'invisibility' as worse than the social stigma associated with visible symptoms [46]. Some communicated a wish to have a visible symptom in order to be perceived as legitimately ill:

> ...if I just could get it a little in one leg . . . you almost do not dare to think it…[51,p.4].

Entangled in this desire however, was a sense of guilt as they see the suffering of others with more visible symptoms:

> When I see these other two ladies reaching and walking really painfully, like they should be in a wheelchair, well you now, you think: 'You're not so badly off…Like you're a bit of a fraud… These people have so much more disability.'[42.p. 17].

The experience of living with MS and its 'invisibility' was understood by several participants in one study as living with an 'invisible employer', who hides their needs as a patient [48]. People with MS described having to 'speak out' about their needs in order to have them met, as the needs associated with their symptoms are undetectable, and "one who does not ask will not receive" [48,p.305]. Some participants also felt their needs were not as high a priority to healthcare providers as those with a visible illness [53].

The emotional impact of invisible symptoms was highlighted by a number of studies [42,47-49,53,57]. The experience of fear, dread, anxiety, humiliation, and low self-esteem were described as being entwined with invisible symptoms. People with MS recognised that that not only is their experience of 'invisibility' associated directly with the symptoms and their unseen nature, but also related to their emotional responses to them which were also hidden from others:

> you're the first person I've spoke to about the emotional things. I've never, ever spoke to anyone about that [53,p.363].

One participant spoke of an appointment with an 'occupational physician':

> She treated me well but it was not like she understood me. She didn't really listen to me, especially the psychological part, which is also quite invisible and therefore very difficult [57,p.2549].

This "lonely" [42,p.19] experience compounds people with MS' sense of 'invisibility', as they do not feel seen or heard in terms of their physical problems, but also their associated emotional turmoil which they feel they must cope with alone.

The burden of choice- a continuous conflict.

Whilst the impacts of the 'invisibility' inherent in their symptoms were very problematic for people with MS, 'invisibility' also afforded them a choice about the strategies they could use to navigate these problems. This choice was whether to disclose their symptoms to others (to be 'seen'), or to keep these hidden (to 'stay invisible')- an option unavailable to those with visible symptoms. While this choice offered people the opportunity to maintain a sense of control over their 'illness identities' and to preserve the image they present to others in their work and social contexts, this dilemma between staying hidden or struggling to be seen was also described as a burdensome and continuous conflict [42,43,44,50,53,56-58]. Participants took an active, purposeful role in managing their 'invisibility', making conscious decisions around 'coming out' as disabled every day.

This process involved weighing up the potential costs of telling others about their symptoms against keeping them private. Both had positive and negative consequences.

Some participants spoke about deciding they would seek to convince others of the disabling nature their symptoms, which resulted in a 'struggle to be seen'. One participant spoke of her disclosure during a job interview:

> … I said 'I think you should know. I have MS and there are times when I'll have to take a day off work to go to the MS Clinic or to a specialist or something.'... He said, 'I never would have guessed.' And I'm like, 'Well I don't wear a tattoo' [43,p 343].

Participants also described using mobility aids even when they were not needed in the struggle for their symptoms to become more visible to others:

> 1: I actually did an experiment with that with my doctors. I have a walking stick which I very, very rarely use and I went in one day [without the stick] and said 'Look, I'm having migraines, I need some Panadeine Forte,' and he gave me a really hard time to get a script. I went and saw him about 4 weeks later and I had my stick with me—wrote out the script no problems. 2: Sympathy stick they call it. 3: I've got one. [46,p.163].

Some participants highlighted that they felt a sense of responsibility to educate others about their invisible symptoms in order to receive adequate support and understanding:

> …that's what I have learned over the years; I have to talk… they don't know how tired I am or how tired I am in my head…If you do not show them what's really going on inside you, you cannot get the understanding of your colleagues [57,p.2548].

Even though they were distressed by the fact that others could not discern their symptoms, most participants chose to 'remain invisible' and not to speak about the existence of their symptoms. Some made concerted efforts to 'stay invisible' [42,43,46], and described minimising symptom-related behaviours as much as possible, shifting attention away from conversations that might lead to discussion of their symptoms, and continuing with daily activities despite exacerbation of symptoms:

> … I do the whole lot, if and when I am able… But I will do it to a point where I know I shouldn't be, you know…I shouldn't really be doing [42, p.18].

A group of participants discussed that even ‘every-day’ conversations with others resulted in conscious decisions to communicate that they were ‘fine’ even when not- “You usually say, ‘Yeah, I'm feeling great’” [46,p.164]. This allowed them to avoid feeling invalidated or disbelieved.

In remaining ‘invisible’, participants continued to tolerate some of the impacts of ‘invisibility’- coping with it alone and the emotional burden of this, and still not having understanding from others. For some participants, not acknowledging their symptoms appeared to be a way of denying the MS and its impact [53], and for others it was an act of defiance, as they felt it was not their responsibility to help others understand [56]. Some expressed fears that disclosure of symptoms in the workplace would affect their employment status and the way they are treated [43]. One participant explained:

> I don't trust how they [her colleagues] are going to behave if I say I have MS. I don't trust that I will have my job again next year. ... Like it feels better to me when people know. And yet I have a really hard time telling ... I mean most of my life I was the one who had a lot of energy, and I can still fake it ... [43,p.342].

This quote also demonstrates the conflict between the desire for others to know, and the fear of losing the perceived or projected image of the ‘self’.

A fear of a disabled identity was expressed by many participants [42,43,53]. For some, disclosing to others had led to the over-emphasis of their condition and pity, which felt unacceptable [53]. Participants chose to ‘stay invisible’ perhaps because the alternative to this is perceived to be risky in terms of being seen as disabled, or worse- a discredited liar [46]:

> …what I don't want to do is to... play the, the disabled cripple card, and get treated differently [53,p.360].

One participant commented about their decision to 'remain invisible', and how this allowed her to be treated as 'normal':

> … [But if your disability is hidden] people don't treat you any different. People treat you like nothing is wrong [42, p. 18].

Many participants perceived that attempts to make their 'invisible' symptoms more visible would result in a de-valued identity. This fear related to being pitied as disabled, judged as a fraud, or simply not being seen as themselves anymore. There was a strong consensus among participants in one study that 'staying invisible' was preferable to the risk of being discriminated against [53]. However, remaining 'invisible' keeps people trapped in an experience of hidden (and therefore unmet) needs, private suffering and poor understanding [53]. One participant expressed her desire to be validated by others, but also to have her sense of identity preserved:

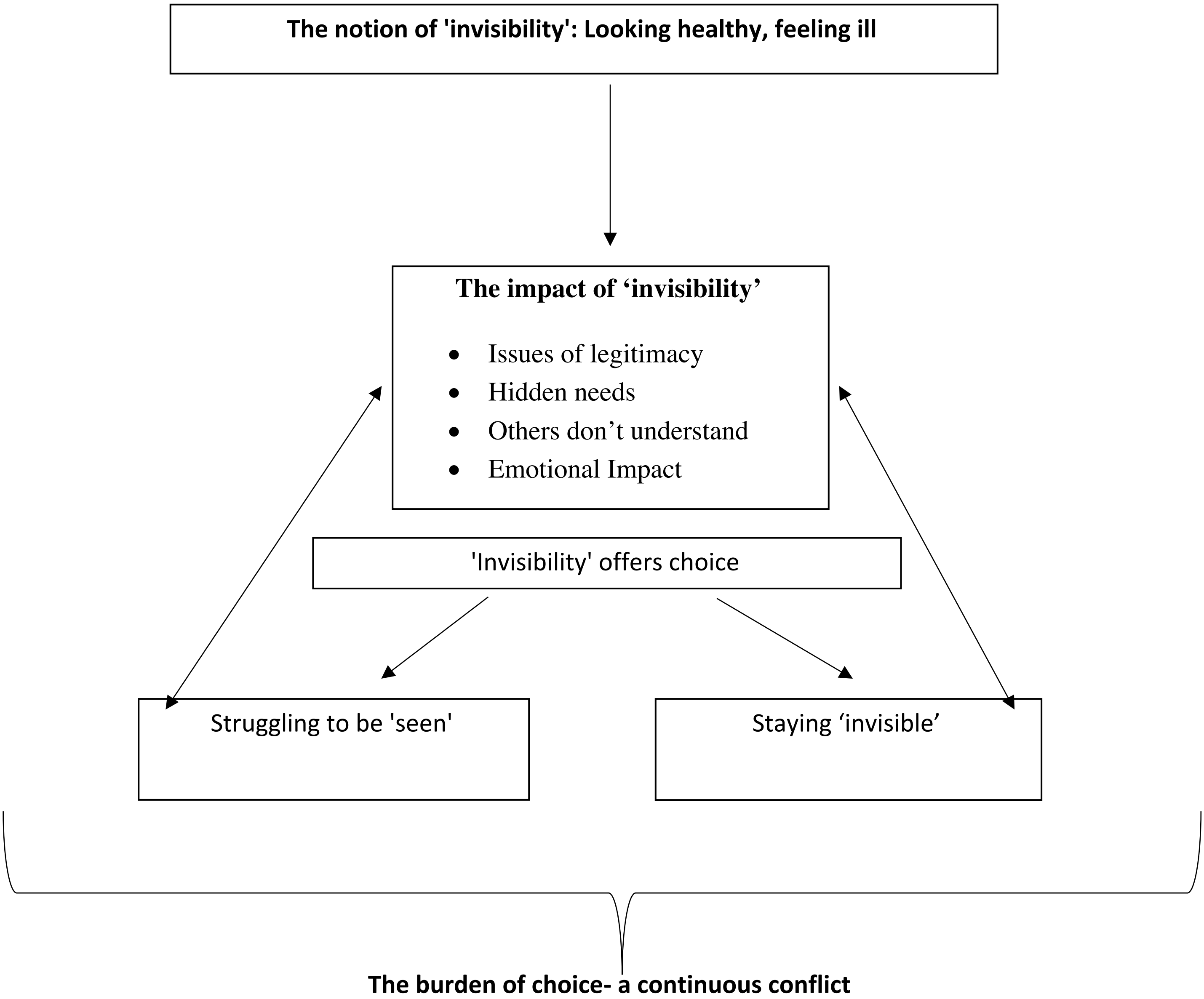
> I want them to acknowledge that there are some things that I can't do, but... I don't want that to change the way they look at me and treat me [53, p.361].

The notion of 'invisibility' in MS and the way participants managed it appeared to be experienced as a double-edged sword.

Line of argument summary

A summary of the line of argument is given in figure 2. We do not claim this to be a theoretical model (and arrows should be treated tentatively and as 'links'- they do not imply causation or a strict temporal sequence), but a conceptual organisation of our third-order themes and sub-themes that show how they can be understood together in relation to the notion of 'invisibility' in MS. The figure shows that 'invisibility' in relation to MS symptoms has a range of impacts that are distressing for people with MS. However, the ‘invisibility’ also offers people with MS choice in terms of the strategies they use to navigate their experiences in their individual contexts and environments. These choices have both positive and negative consequences and encompass an ongoing conflict between the struggle to be ‘seen’ and the desire to remain ‘invisible’.

Figure 2. Line of Argument



Discussion

This review explored the notion of 'invisibility' in relation to people's lived experience of MS symptoms, and unpicked not only how people with MS experience 'invisibility' and its impact, but how they manage this. The review findings elucidated the concept of 'invisibility' related to MS symptoms as the experience of feeling unwell and struggling internally with symptoms, however these experiences not being visible to the onlooker. This is consistent with the available definitions in the literature.

'Invisibility' in relation to people's lived experience of MS symptoms had a variety of impacts. People with MS spoke about feeling misunderstood by friends, family, co-workers and health professionals, leading to difficult and sometimes invalidating interactions. Not only did people feel their illness was delegitimized by those around them but they also doubted themselves and their right to a disabled identity. This echoes the reported experiences of people in patient-produced brochures published by the National MS Society [9]. Visible symptoms or indicators of illness were considered to be legitimizing, and a wish was expressed by those experiencing 'invisibility' to be understood and sympathised with in the way that someone with visible symptoms might be. The experience of 'invisibility' appears to keep people's needs hidden from view, and people with MS must explicitly ask from others in order to receive help. The emotional impact of the experience of 'invisibility' also remains hidden, compounding a lonely experience where people feel they must cope on their own.

The findings highlight a dilemma inherent in the notion of 'invisibility'. By its nature, experiencing invisible symptoms offers people with MS the choice to continue to stay silent about their symptoms, or to try to be 'seen'. This dilemma, whilst offering a sense of agency and control to people, can also be burdensome as they have to face these decisions constantly in their various contexts.

Ways in which people with MS might attempt to be ‘seen’ include having open discussions with others where they inform or try to convince them of the symptoms or adopting symptom-related behaviours or mobility aids deliberately, so that others might see these as visual reminders of their illness. The decision to attempt to be ‘seen’ appears largely motivated by people’s wish for others to understand and believe them- a search for legitimacy. However, there is also a perceived risk of discrimination associated with a disabled identity, and the struggle appears to be relentless.

Most people with MS chose to ‘remain invisible’, despite wishing that others could understand more about their experiences. This appeared to be motivated by a desire to protect and maintain their public image, and to avoid becoming associated with a sick and pitied disabled identity. This is supported by Goffman [59] and Charmaz [60] in their theorizing that people with disabilities experience a ‘spoiled identity’ and loss of self as a result of the stigma attached to being disabled. Goffman proposes two identities: the ‘stigmatized’ (those who bear the stigma and are viewed as discounted) and the ‘normals’ (who do not bear the stigma and appear as ‘normal’). This theory can be used to illustrate the struggle depicted in our findings for people with MS. We suggest that people who experience such ‘invisibility’ are torn between these identities, having the option to be perceived as one of the ‘normals’ but not have their needs met or sufficiently understood, or to become the ‘stigmatized’ in their search for ‘visibility’. The findings also suggest that people with MS expect others will not understand or will label them fraudulent if they attempt to convey their invisible symptoms, and so the ‘safest’ option to preserve a sense of self and sense of ‘normalcy’ in society is to stay silent and ‘invisible’. Parsons [61] suggested that people with conditions that are perceived to be under their control (consistent with experiences highlighted in this review) are more at risk of social rejection. Thus, the feared stigmatisation for people with MS may not just be about the assumption of a disabled identity, but a fear of a spoiled social identity where they are viewed as a liar.

As a result of ‘staying invisible’, some of the impacts of ‘invisibility’ are compounded- people’s needs remain hidden, and they still feel forced to cope alone. This choice to remain ‘invisible’ appears to create a self-fulfilling prophecy, perpetuating a continued cultural lack of understanding that keeps the individuals trapped in their ‘invisibility’.

Implications

Although the included studies provide data pertaining to ‘invisibility’, none took a primary focus exploring this. Therefore, future research should further explore more directly people with MS’ experiences of ‘invisibility’, and to develop a greater understanding of the issues that have been identified in this review, including the ways in which people manage their ‘invisibility’ on a day-to-day basis. A particularly distressing aspect of ‘invisibility’ in MS is related to a lack of awareness about the undetectable nature of many of its symptoms. Research can provide a vehicle to bridge the gap in both clinical and public understanding of this phenomenon. Raising awareness through research may help to shift the personal burden and dilemma of becoming ‘seen’ for people who experience ‘invisibility’, to wider society as being responsible for understanding it. This may allow people to feel more ‘seen’, without them having to make the perceived personal sacrifices of their identities or risking discrimination in their everyday decisions to ‘come out’ or not.

Contact with healthcare professionals and providers was largely difficult for people with MS, who experience not being believed, or being dismissed on the basis of the ‘invisibility’ of their symptoms. Healthcare professionals should be aware of the impact of ‘invisibility’ and be able to engage in sensitive discussions with patients around this.

Limitations and Strengths

Although quotes were presented from most of the studies to support the synthesis, there was variability in the number of quotes drawn from each paper. One paper [42] is more heavily represented than others, as it offered a greater number and longer quotes that were directly relevant to the review aims due to the narrative style in which it was written. This paper had the lowest quality however due to a lack of comments about methodological rigour, which highlights the need for future qualitative research in this area to be both methodologically rigorous, and rich in terms of direct supporting quotes. Although study [42] contributed to numerous themes, these themes were evident in many of the higher quality studies.

As the data in qualitative research is subject to the interpretation of the researcher, the research as a whole is likely to be influenced by the expectations, experiences and positions of the researchers. The reflexive process is considered essential in qualitative research for the researcher to remain aware of how the research process can construct the object of the research [62]. We considered this information in the synthesis process when making judgements about study quality; however, meta-ethnography studies are not as committed to the concept of quality appraisals as other approaches to meta-synthesis [63], and to exclude or disregard a study’s contributions based on quality alone could remove relevant and interesting information from the synthesis [27].

We attempted to find a balance between a broad scope review, and a search that would yield a manageable number of studies to synthesise [64]. The search strategy was limited to using keywords associated with specific common invisible symptoms (fatigue, cognitive impairment, pain, bowel and bladder dysfunction). Whilst this yielded a high number of results, and keywords to capture a general lived experience of MS were also included which allowed us to capture discussions about other invisible symptoms, the search may have been more inclusive (but potentially less manageable) by incorporating more invisible symptoms into the search strategy.

Due to the interpretive nature of meta-ethnography, we acknowledge that the author conducting the analysis (LSP) had their own expectations for the data based on their experience of speaking to people with MS informally about ‘invisibility’ related to their symptoms in patient involvement groups. Whilst measures were taken to maintain an awareness of LSP’s responses to the data (e.g. keeping a reflexive log of responses, coming back repeatedly to the questions when making decisions about the data and developing themes), we acknowledge that the synthesis will have been influenced to some degree by their expectations and position. Cross-verification of themes and regular discussions with the rest of the research team were used to manage expectations and potential biases so that their influence on the review findings were minimised.

Independent cross-verification of themes and third-order constructs by all members of the research team add credibility and reliability to the findings of this review [65], strengthening its quality. The screening processes for included papers and quality appraisals were conducted by at least two reviewers independently to improve the reliability of these processes.

Throughout the review we adhered to the principles of meta-ethnography [37] and conducted the review according to PRISMA guidelines [41] which outline preferred reporting standards for systematic reviews.

Conclusion

People with MS describe their experience of invisible symptoms (such as fatigue, cognitive problems, pain, and bowel and bladder dysfunction), where there is a discrepancy between their outward appearance and how they are feeling internally. The invisible nature of such symptoms is highlighted as distressing (and for some, the most distressing part of MS) in a number of ways. Many people with MS report experiencing a lack of understanding and sense of illegitimacy around their invisible symptoms, affecting their interactions with friends, family, colleagues and health professionals and compounding their sense of invisibility and loneliness. People with MS also feel that their emotional, physical and social needs are hidden and invisible, thus limiting the support they receive from others. People with MS who experience invisible symptoms are faced with a continuous dilemma of whether to disclose their symptoms to others and make their needs known, or to remain 'invisible' and maintain a sense of control over their illness identities. Health professionals should support people with MS to navigate these decisions.

It is important for both public and professional awareness of invisibility in MS to increase. Health professionals should seek to validate and instigate conversations around people with MS's experience of symptoms, giving people with MS the opportunity to speak about those which are unseen (and sometimes unspoken) and to receive appropriate support. These findings suggest also that health professionals should address and respond to invisible symptoms of MS with the same sense of significance as those which are visible. Encouraging more clinical and research acknowledgment and focus on what clearly appears to be a distressing element of living with MS, will increase our understanding of how people with MS navigate and manage their symptoms in everyday life and how to best support them.

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Declaration of Interest Statement

The authors report no conflicts of interest.

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Journal Paper

‘Seeing the Invisible: A Photovoice exploration of living with, and managing, the Invisible Symptoms of Multiple Sclerosis’

The following article is prepared for submission to the journal ‘Social Sciences & Medicine'[1][2].

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[1] Tables and figures have been included in the main text for ease of marking, which deviates from journal style

[2] Please see appendix C for author guidelines for ‘Social Sciences & Medicine’

Seeing the Invisible: A Photovoice exploration of living with, and managing the Invisible Symptoms of Multiple Sclerosis

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Abstract

People with Multiple Sclerosis (MS) typically experience a constellation of invisible symptoms that are not observable to others. Living with these symptoms is difficult because of their physical impact, and because they are hidden and present additional challenges for people with MS in navigating the invisibility of their experiences. This research aimed to explore how people with MS experience *living with* and *managing* invisible symptoms in daily life. We used a photovoice method, whereby 12 people with MS produced digital images over a two-week period to capture their experiences of living with and managing their invisible symptoms. Participants discussed their images in semi-structured interviews. The study was conducted in England, and data were collected between August 2019 and May 2020. We thematically analysed the interview data and developed three main themes and eight subthemes. The themes encompass the difficulties around conceptualising invisible symptoms not only due to their invisibility, but because people found it difficult to use language to accurately communicate their experiences. We outline the conflicts of legitimacy this presents for people with MS, where the reality of their invisible symptoms is invalidated by others and for themselves. Participants navigated these issues in dynamic ways, choosing to fit their symptoms to their lives or make space for their symptoms depending upon the context, often influenced by their desire for their symptoms to ‘stay invisible’ or to ‘be seen’. This study offers a unique exploration of experiences that cannot be captured through words alone, in which people with MS invited us to ‘see the invisible’ they navigate every day. We highlight clinical implications for supporting people with MS around the legitimacy conflicts they experience and how they negotiate living with invisible symptoms.

Keywords: multiple sclerosis; invisible symptoms; photovoice; visual methods

Introduction

Multiple Sclerosis (MS) is a multifaceted, chronic, and often progressive disease of the central nervous system, characterised by an abnormal immune system response and damage to nerve fibres and myelin sheaths[3]. The demyelination of axons causes cognitive, affective, sensory and motor impairments for those with MS, which significantly impacts their lives (Yorkston et al., 2001)[4]. These impairments are classified into symptoms which are 'visible' (e.g., easily discernible issues, such as mobility problems), and 'invisible' (Fenu et al., 2018).

Invisible symptoms of MS are largely defined in the research literature as symptoms that are difficult for others to notice as debilitating and appear 'hidden' to the onlooker (Stuke et al., 2009; Werfel & Trettin., 2020; White et al., 2008). Invisible symptoms commonly cited in the literature include cognitive impairment (e.g., memory problems), fatigue, pain, bladder and bowel dysfunction, sexual problems, and sensory dysfunction such as numbness, tingling, and issues with vision (Fenu et al, 2018; Kratz et al, 2016)[5].

Fatigue is the most common invisible symptom (experienced by up to 75% of people with MS) and is reported to have significant detrimental effects on daily

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[3] Please see extended paper section 1.1 for further information about MS characteristics, diagnosis and treatment.

[4] Please see extended paper section 1.2 for further information about the impacts of MS symptoms on people's lives

[5] Please see extended paper section 1.3 for further information about invisible symptoms of MS

functioning (Induruwa, et al., 2012; Kos et al., 2008). Fatigue is also the most commonly researched invisible symptom of MS (Krupp et al., 2005). Pain, neurogenic bowel dysfunction, and cognitive impairment are also among the most commonly reported, with the latter affecting up to 60% of people with MS (Amato et al., 2013; Norton & Chelvanayagam, 2010; Svendson et al., 2005).

Research papers, journalistic pieces and patient accounts highlight the various ways that invisible symptoms impact the lives of the people who experience them. Green and colleagues (2017) investigated the relationship between symptom severity and perceptions of health (physical, mental and social) in 1865 outpatients. Invisible symptoms of MS (specifically pain, fatigue and cognitive impairment) were predictive of negative health perception. Pain was the most predictive even when compared to 'visible' symptoms. The authors concluded that invisible symptoms of MS are as important to the individual's sense of wellbeing as visible symptoms, and that given the association between negative health perception and mortality rates in MS, the long-term impact of living with invisible symptoms should not be underestimated. Another study found that invisible symptoms were more predictive of health distress than visible symptoms, and the authors suggested that those with invisible symptoms required adequate support with these (White et al., 2008).

The UK MS Society (2017) published an edition of their periodical 'Research Matters' that focussed on invisible symptoms of MS and their distinct impacts that differed from the experience of visible symptoms. A patient-produced brochure entitled "But you look so good!" included a selection of quotes from people with

MS communicating the psychological distress of living with invisible symptoms (National MS Society, 2016). Both publications referred to patients' experiences of stigma related to their invisible symptoms, including perceptions of not having a 'real' illness, social isolation, reduced confidence in own perception of symptoms, and discouragement from help-seeking.

Qualitative studies have emphasised stigma in MS in general, with specific themes describing a lack of understanding and validation from others around invisible symptoms (Cadden et al., 2018; Grytten & Maseide, 2006; Turpin et al., 2018)[6]. In one study, participants reported purposefully concealing or disclosing their MS to influence judgement in their social encounters, although this was a study inclusive of but not specific to invisible symptoms (Grytten and Maseide, 2005).

Our meta-synthesis of 17 qualitative studies relating to lived experience of 'invisibility' in MS revealed numerous negative impacts (Parker et al., 2020). The invisible nature of people's symptoms was highlighted as distressing and, for some, the most challenging part of having MS. People with MS often did not feel understood or believed by others in relation to their invisible symptoms, and felt their social, physical and emotional needs were often invisible too as a result (Parker et al., 2020). The meta-synthesis showed that invisible symptoms, by nature, offered people a choice of strategies to navigate the invisibility, but confronted them with a burdensome dilemma of disclosing their symptoms to others and making their needs known, or remaining 'invisible' and maintaining a

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[6] Please see extended paper section 1.4 for an overview of research in relation to MS and stigma

sense of control over their illness identities (Parker et al., 2020). Whilst the meta-synthesis uncovered some understanding about the notion of 'invisibility' in MS, the studies included did not primarily focus on the experience of invisible symptoms. The meta-synthesis highlighted a need for focussed exploration of people's experiences of living with invisible symptoms and to better understand the ways in which people with MS manage these.

Studies suggest that many people living with invisible MS symptoms strive to find adaptive ways of managing them through developing strategies and utilising available resources (Norton & Chelvanayagam, 2010; Stuifbergen & Rogers, 1997). Many of these studies focus on practical management of symptoms, rather than elucidating the way in which people navigate the psychological and social impacts that are a prevalent part of living with invisible MS symptoms[7].

The majority of studies investigating the impact of invisible symptoms of MS have focussed on individual symptoms in isolation (Kratz et al., 2016; Olsson et al., 2005). There is a dearth of original empirical research offering an in-depth exploration of lived experience of a cluster of invisible MS symptoms (which often is the case, because people with MS experience multiple symptoms; Kratz et al., 2016). Furthermore, previous qualitative research in this area has solely used interview and focus group methods, with limited participant co-creation and interpretation of the data. There appears to be an absence of participatory visual approaches to MS research - an approach that has been reported to create a rich

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[7] Please see extended paper section 1.5 for further information about invisible MS symptom management

understanding of the experiences present in health populations (Topcu, 2015). No studies have used visual methodologies with people with MS, despite evidence to suggest the benefits of these approaches (Glaw et al., 2017).

Our main aim was to obtain an in-depth understanding of people's holistic experiences of living with invisible symptoms of MS, and gain insight into the ways people manage and navigate their lives in the context of their invisible symptoms, offering a rich account of this through visual methods. Additionally, we aimed to determine to what extent the data fit with Parker et al's (2020) conceptual framework around the notion of invisibility in MS.

Methods

Design[8]

We used a participatory approach using a visual method commonly referred to as 'Photovoice', whereby participants produced their own digital images to capture their experience of invisible symptoms of MS[9]. In photovoice, photographic images are created during the research process by the participants and used as a point of discussion in interviews to uncover corresponding and related narratives (Foster-Fishman et al., 2005). Photovoice offers the means for participants to capture and reflect on their experiences, and for key messages related to health issues to be communicated to healthcare providers, stakeholders and policy makers (Wang, 1999). Visual methods are considered complementary to participatory approaches, allowing researchers to become

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[8] Please see extended paper section 2.1.1 for information around the use of qualitative methods in health research

[9] Please see extended paper section 2.1.2 for further information about Photovoice as a methodology

immersed in the world of participants through the production of real-time, real-world data (Schwartz, 1989).

Photovoice has been successfully used in MS research to explore carers' experiences in relation to quality of life (Topcu et al., 2020), and as an exploratory tool to elicit experiences of invisibility in Parkinson's disease (Roger et al., 2018)[10].

Patient & Public Involvement & Engagement (PPIE)

We obtained feedback from members of a local PPIE group on the initial study protocol and incorporated this into the study design. A PPIE member became a part of the research team and was involved at every stage of the research process[11].

Epistemological Position

We adopt a critical realist position, which assumes that underlying unobservable processes cause real effects (Harvey, 1990). When applied to the present project, we assume that there is an objective and physical reality to invisible symptoms, but participants' experience of their symptoms is interpreted subjectively and is constructed by the participant in collaboration with the researcher[12].

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[10] Please see extended paper section 2.1.2 for further rationale for the use of Photovoice

[11] Please see extended paper section 2.2 for further information on PPIE

[12] Please see extended paper section 2.3 for further information on epistemological positioning

Participants & Recruitment [13]

Advertisements were placed in local MS Society newsletters and social media pages, in a UK National Health Service (NHS) MS outpatient service, and in a local MS PPIE newsletter. Opportunistic sampling was used with a maximum variation sampling frame in an attempt to increase diversity across the sample in relation to age, gender, ethnicity, and MS subtype[14].

We required participants to be aged 18 years and over; have a diagnosis of MS, and experience or have experienced one or more invisible symptoms of MS; have access to an electronic device with a camera function (e.g. smartphone, tablet) or digital camera; and be physically able to use this device to take an image.

Once eligibility to participate was confirmed, participants were informed of all aspects pertaining to participation, and were sent an 'information pack' prior to giving consent to participate[15]. Participants were made aware of their right to withdraw from the study.

Procedure & Data Collection[16]

Participants met with the first author for an orientation meeting and were provided with further guidance about the study processes. The information they received included a list of common invisible symptoms of MS, however, participants were advised that they could self-define invisible symptoms based on what this term

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[13] Please see extended paper section 2.4 for further information on recruitment (including inclusion and exclusion criteria)

[14] Please see extended paper section 2.4 for further information on sample size rationale and 'data sufficiency'

[15] Please see appendix for participant information sheet

[16] Please see extended paper section 2.5 for further information on study procedure

means to them individually. Participants were asked to use their own electronic devices to take a minimum of five images that they felt captured their experiences of living with and/or managing their invisible MS symptoms.

Participants were asked to send their images to the first author by their choice of Multimedia Message (e.g., text message), email, or secure encrypted 'WhatsApp' message as soon as they could after taking the pictures. They were also requested to send a short narrative about what the image captured or represented. Participants were asked to send all images within two weeks.

One week from the orientation meeting, the first author telephoned each participant to offer support if needed, and to identify any issues with participation. Two weeks after the orientation meeting, participants were invited to a semi-structured interview with the first author and to select five images to discuss in their interview.

The semi-structured interview included inductive questions to elicit discussion about each image and its meaning for the participant[17]. Once the five selected images had been discussed, all participants were given the opportunity to talk about any additional images they had taken. Interviews were audio-recorded and transcribed verbatim by the first author[18]. The participants received a gift voucher with a well-known retailer for the value of £10 as an expression of gratitude towards their contributions.

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[17] Please see extended paper section 2.5.2 for further information on data collection, the use of semi-structured interviews and the development of the interview schedule for this study

[18] Please see extended paper section 2.5.2 for further information about the use of transcription in the research process

Ethical Considerations

Ethical approval was obtained from the first author's academic institution, and the National Health Service (NHS) Research Ethics Committee for East Midlands-Derby (19/EM/0196)[19]. Some participants opted to be referred to by their real names in the study, and others chose their own pseudonyms[20].

Data Analysis

Data (audio recordings, written narratives and transcriptions) from semi-structured interviews were analysed by the first author using an Inductive-Deductive Thematic Analysis (TA)[21,22]. Guidelines provided by Braun and Clarke (2006) were used for the TA, outlining six phases of the process including: familiarisation with the data, generation of initial codes, drawing out salient features and patterns as themes, exploring connections between themes, and checking and defining themes. Data were firstly approached inductively (data driven and open to discovery of new knowledge), and secondarily using a deductive (theoretically driven) framework. The deductive framework (see Appendix A) was developed by drawing on the key themes and conceptual framework highlighted by Parker et al. (2020) in their systematic review of experiences of invisibility in MS[23].

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[19] Please see appendix for ethical approval

[20] Please see extended paper section 2.6 for further ethical considerations

[21] Please see extended paper section 2.7 for further information on Thematic Analysis and consideration of other analytic approaches

[22] Please see extended paper section 2.7 for further information on inductive vs. deductive analysis

[23] Please see extended paper section 2.7 for further information on the development of the deductive frame

We applied Yardley's (2000) guidance for good qualitative research to this study. To improve rigour and credibility of the analysis, a third of the transcripts were cross-checked by other members of the research team for consistency of coding. In addition, the themes generated by the first author were checked by all other authors in terms of the plausibility of all interpretations made. We discussed and resolved any discrepancies or queries in regular meetings[24]. To ensure quality and transparency of study reporting, we used the Critical Appraisal Skills Programme (CASP) for qualitative research[25].

Reflexivity[26]

We maintained an active process of critical reflection throughout this research. The first author used a reflective diary to record and manage their subjective expectations, assumptions and personal stances in relation to the research. This diary was shared with other members of the research team for further reflection and discussion of potential biases[27].

Results

In total, 14 adults with MS volunteered and consented to participate, however two of these withdrew in the early stages, leaving 12 participants. Participants were

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[24] Please see extended paper section 2.7 for further detail on the analysis process (including second checking)

[25] Please see extended paper section 2.8 for further information on evaluating quality of qualitative research and how the Evaluative Criteria and CASP were considered

[26] Please see extended paper section 2.8 for further information on reflexivity

[27] Please see extended paper section 2.8 for further detail on how reflexivity was practised (including reflexive statement by first author)

asked for their preferred pronouns which are used throughout this publication. Demographic details for the participants are presented in Table 6[28].

Table 6. Participant characteristics

Participant Name/Chosen Pseudonym	Age (years)	Gender	Ethnicity	MS Subtype	Time since diagnosis (years)
Fay	51	Woman	White-British	Secondary progressive	15
Laura	30	Woman	White-British	Relapsing Remitting	4
Claire	34	Woman	White-British	Relapsing Remitting	10
Jacqueline	57	Woman	White-British/ Swiss	Secondary progressive	10
Stuart	36	Man	White-British	Relapsing remitting	6
Kate	57	Woman	White-British	Relapsing remitting	4.5
Wendy	43	Woman	White-British	Relapsing remitting	5
Anniemac	56	Woman	White-British	Secondary progressive	20
Daniel	41	Man	White-British	Secondary progressive	10.5
Ann	51	Woman	White-British	Relapsing remitting	16
Jennie	43	Woman	White-British	Relapsing remitting	12
Esther	36	Woman	White-British	Relapsing remitting	7

A total of 73 images were taken by the participants, however these were not used as data per se, but to elicit discussion during the interviews[29]. The inductive-deductive analysis led to the development of three main themes and eight subthemes. Inductive codes contributed to the construction of all themes as did deductive coding and were synthesised together. The resulting themes provided substantial support for our deductive framework and allowed us to uncover richer

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[28] Please see extended paper 3.1 for additional results and demographic information

[29] Please see extended paper for more images and corresponding narratives

detail and explicate the processes that exist within the experience of living with and managing invisible symptoms[30]. The themes outlined below are those which developed the framework further. The data were an almost perfect fit with the deductive frame, and one inconsistency is highlighted in this section of the paper.

The challenges of conceptualising the invisibility of symptoms

All participants described experiencing multiple symptoms of varying severity that they defined as being invisible in nature (outlined in Table 7) and the challenges they are faced with when conceptualising and communicating about their symptoms because of their invisibility[31].

Others' blindness to my internal struggles

Invisible MS symptoms were conceptualised by all participants as a discrepancy between what they experience internally and what is observable externally. All described experiencing symptoms and internal struggles related to their symptoms that cannot be seen by others:

> *It's the invisible-ness of MS. Because most of it is internal… I can feel it, I know it's there, but to anybody else, they can't see it. (Fay)*

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[30] Please see extended paper section 3.5 for details on the use and fit of the deductive framework

[31] Please see extended paper section 3.2 for additional comments on this theme and its related sub-themes

Table 7. All Symptoms Identified by Participants as being Invisible

	Participant											
Symptom defined by participant as being invisible *(Italicised when sub-type specified)*	Fay	Laura	Claire	Jacqueline	Stuart	Kate	Wendy	Anniemac	Daniel	Ann	Jennie	Esther
Fatigue	*	*	*	*	*	*	*				*	*
Depression					*		*	*				*
Anxiety												*
Pain	*		*	*	*	*	*		*	*		*
Trigeminal Neuralgia	*			*								
Other caused by nerve damage							*					
Headaches										*		*
Joint pain												
Temperature regulation problems/sensitivity	*	*							*		*	
Balance problems						*			*	*		
Vertigo										*		
Optic Neuritis		*	*	*						*	*	*
Blurred/Double Vision				*						*	*	*
Blindness		*								*	*	*
Colour Blindness				*							*	
'MS Hug' (incl. pain and spasms)[32]		*									*	*
Cramping/ Muscle Spasms									*		*	
Muscle stiffness/ Spasticity	*	*							*			
Muscle Weakness	*					*						
Difficulty Swallowing										*		
Cognitive Difficulties (memory/word-finding/organising thoughts/concentration		*	*	*		*		*	*		*	*
Sensory Difficulties (e.g. numbness/tingling/ itching/Lhermitte's Sign-electrical sensation down spine)	*	*	*	*		*		*	*	*	*	*
Bladder issues (urgency/weakness)	*				*				*	*		
Bowel Issues										*		

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[32] We have listed the 'MS Hug' separately from pain and spasms as it is often recognised in the literature as a separate symptom, and our participants described it as a unique experience that is distinct from other experiences of spasms and pain.

For some, invisible symptoms were felt to be the most challenging aspect of their MS, even for those who experienced visible symptoms:

> *…the invisible symptoms that people just don't know about… They're the ones that really get you…I struggle, as you can see with mobility…but it's the invisible that I really struggle with. (Jacqueline)*

Can't be seen, can't be spoken about

Participants described their symptoms as "*strange*", "*odd*", "*weird*", "*bizarre*" and "*abstract*" experiences and sensations which are difficult to describe. Ten participants spoke about not having the language with which to communicate their invisible symptoms or explain them to others, despite a desire to. This has a compounding effect on their sense of invisibility and the discrepancy between what is felt internally and what can be said about it:

> *It's trying to find ways to explain to people how you feel. That can be quite challenging. (Laura)*
>
> *Trying to put it into words sometimes can be a bit awkward… Trying to explain to people, you just can't. (Daniel)*

Participants relied on a range of linguistic methods including similes, metaphors, use of word emphasis and sound effects to attempt to communicate their experiences, yet maintained a sense of being unable to translate their symptoms accurately and in a form accessible to others. Some images taken by participants represented their symptoms in abstract and metaphorical ways. For example, Kate demonstrated the heaviness and stiffness in her legs in her image (Figure 3):

> *I often feel like I'm* <u>*wading*</u> *through treacle… your legs are really heavy. I feel like I've got big boots on and it's real hard work. (Kate)*

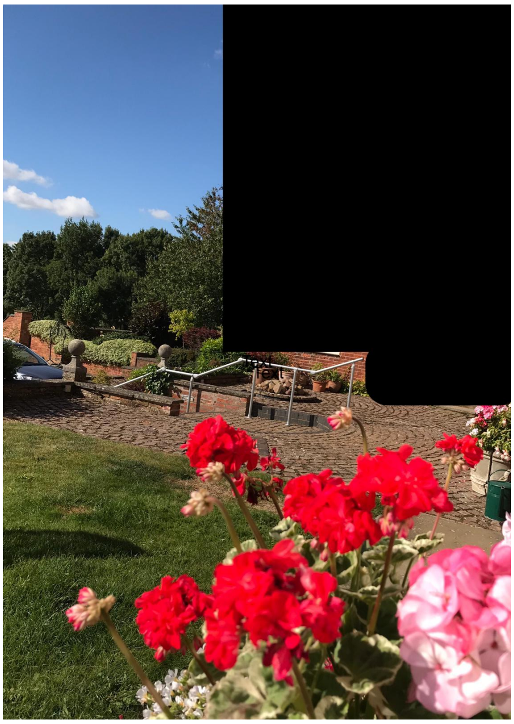
Figure 3. *Treacle* taken by Kate

Jacqueline's experience of Trigeminal Neuralgia pain was "indescribable", so she made use of colour in her image (Figure 4) to communicate this:

> *the invisible is visible because that red…my goodness me…I just* <u>*cannot*</u> *describe the pain of neuralgia, I really can't. It is so* <u>*excruciating*</u>*…like having electric shocks down your face…And then *explosion noise* it really hits your face…Zzzz just going down, and it's a* <u>*raw red*</u> *(Jacqueline)*

Participants' accounts indicate that their invisible symptoms are not only experiences that cannot be 'seen', but ones that cannot be spoken about.

Figure 4. *Red Geraniums* taken by Jacqueline



[redacted]

Conflicts of legitimacy

All participants discussed issues of conflict in relation to the perceived legitimacy of their invisible symptoms, grounded in the fact that they are not observable[33]. We have understood these issues of legitimacy conceptually as *epistemic* and *experiential*, where participants struggle to validate the 'realness' of their invisible symptoms to both themselves and to others and feel invalidated as a result.

<u>An invisible reality (epistemic issues of conflict)</u>

Participants highlighted epistemic issues created by the fact that their invisible symptoms cannot not be clearly seen or communicated, and the difficulty of validating the 'realness' of their symptoms without external evidence of them, to

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[33] Please see extended paper section 3.3 for additional comments on this theme and its related sub-themes

both themselves and others. Some participants spoke of the importance of their Magnetic Resonance Imaging (MRI) scans in validating their symptoms by providing ‘objective’ evidence of their reality. Esther felt it was important for clinicians to help people with MS to understand their MRI scans to validate their invisible experiences, and believed the absence of MRI evidence could lead her to question the reality of her symptoms:

> *It might help the patient to feel less like they’re going mad… ‘here it is, your MRI scan shows that yes you do have these symptoms’... Because otherwise it’s just my word against anyone else’s...whilst I’m not making it up, it can feel like because it’s all hidden that it’s all just a figment of your imagination. (Esther)*

Some participants described longing for a physical indicator of their symptoms to verify their invisible reality and resolve the legitimacy conflict:

> *‘why couldn’t I have just lost a leg or something?’ Because then if somebody asks what’s wrong with you, you just go ‘that’s what’s wrong with me’ and it’s something you can see. (Stuart)*

Laura discussed the difficulties of quantifying her invisible symptoms to others, which had an impact on deciding whether to ‘call in sick’ for work or not. She worried that others would question the reality of her symptoms at work because she could not provide evidence of it as she would with “a snotty nose”:

> *it’s so hard to measure [invisible symptoms]…You can’t quantify how tired you are…I just couldn’t ring up and say, ‘I can’t come into work today*

> *because I'm fatigued or exhausted'. I would just make up an excuse and say I'd been [physically] sick. (Laura)*

<u>*External invalidation (experiential issues of conflict)*</u>

Experiences of feeling dismissed, misunderstood and invalidated by others in relation to their invisible symptoms were described by 11 participants, which brought the reality of their symptoms into question and worsened the legitimacy conflict. Esther spoke about people telling her that she did not look unwell, capturing this in an image of a seemingly 'fine day' (Figure 5):

> *People say to me 'oh you look fine though, even though you've got MS', 'oh, but you look so well!'…people actually forget that I have MS…it can make people feel misunderstood and that you may even be fabricating the whole thing (Esther)*

Figure 5. *Fine* taken by Esther

Some participants believed invalidating reactions from others most often occurred in the absence of physical evidence for their symptoms. Claire spoke about her tram commute to work, and her experience of others assuming that she does not need to use a disabled seat, despite her internal struggle with fatigue. She denoted this with an image (Figure 6) of the disabled seat she often arrives deliberately early at her tram stop to ensure she can sit on:

> *anyone getting on the tram when it's packed doesn't see me as needing to sit in those seats…I sit in a disabled spot, but I don't look disabled…you just hear people talking…That picture's showing someone with a stick needing a stick…whereas people need to sit on there that don't have sticks. (Claire)*

Figure 6. *Public transport* taken by Claire

Claire observed that the 'disability' image includes a representation of an 'aid' which clearly marks a disability, serving to further invalidate her hidden disability.

Participants discussed feeling validated about their invisible symptoms when speaking to others with MS, who have an understanding that those without MS cannot evoke. When Wendy's friendships became strained due to their lack of understanding around her symptoms, she created new friendships with those she felt understood by (see Figure 7 to see how Wendy conceptualises her friends as a supportive 'herd'):

> *I set out to make new friends through the MS society…you have quite a lot in common with them and can talk to them. It's quite reassuring. (Wendy)*

Figure 7. *Cows* taken by Wendy

Some participants commented that their closest loved ones have tried to understand and have made conscious efforts to validate their experiences. Often, they have more of an understanding than the general public and at times even notice when the participants are struggling with symptoms. Whilst this does not fit with the deductive framework, these accounts were also accompanied by the commentary that despite the efforts of loved ones, participants still do not feel understood due to the invisibility of their symptoms. We therefore considered this to be inconsistent with the deductive framework to a degree, but not refutational.

Participants questioned their own symptom legitimacy in response to their experiences with others, such as wondering whether their symptoms are "*psychosomatic*" and "*mind over matter*". Participants described self-questioning whether they were "*lazy*" (when experiencing fatigue and depression), "*dim*" (cognitive issues), "*fabricating*" their symptoms, and "*making excuses*" for not doing things because of their symptoms.

The negative emotional impact of invalidation was highlighted, which could exacerbate the symptoms themselves. Laura captured an image (Figure 8) on a particularly challenging day at work, where she felt dismissed by her colleagues whilst struggling with invisible symptoms. She went to the staff toilets to cry when she took the image:

> *I just found it really overwhelming…that lack of understanding from other people...when you're trying your best and you're trying to manage a condition, if someone says something quite out of turn and not very kind it has such a big impact (Laura)*

Stuart captured an image of his alarm clock (Figure 9) during a night where he lay awake worrying about his ordeal of being disbelieved by benefits assessors to grant him Personal Independence Payments[34]. Others highlighted similar

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[34] Personal Independence Payment is a welfare benefit in the United Kingdom that is intended to help adults with the extra costs of living with a long-term health condition or a disability. Each person is individually assessed by a benefits assessor.

difficulties, compounded by the invisibility of their symptoms and pressure to 'prove' the symptoms' existence:

> *they [benefit assessor] assessed me and said 'that's fine, we believe you'...And then they reassess and go 'oh there's nothing wrong with you'...they go 'nah, there's nothing wrong with you. Go back to work' (Stuart)*

Figure 8. *Overwhelmed and isolated* taken by Laura

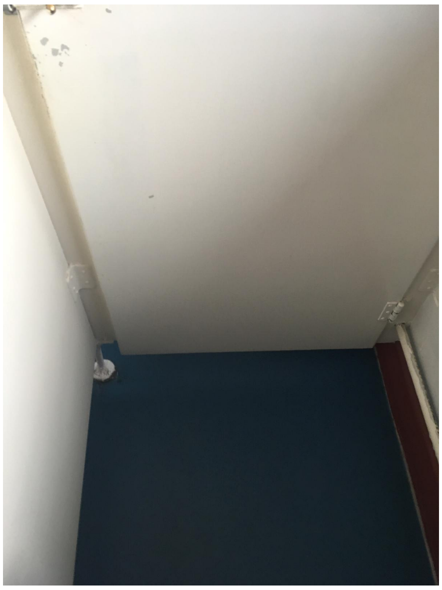

Figure 9. *Clock* taken by Stuart

Navigating life with Invisible Symptoms[35]

We found that all participants negotiate their lives with invisible symptoms in dynamic ways dependent upon their contexts and needs. We identified that participants' styles in doing so can differ and may be characterised by 'pushing through' their invisible symptoms and trying to make the symptoms fit around their perceived lives, roles and responsibilities. We also uncovered narratives around 'making space' for and 'accepting' their invisible symptoms, with participants organising their lives around their symptoms. Not only do the approaches of 'pushing through' and 'making space' help participants to manage living with their invisible symptoms more generally, but also have implications for the invisibility of their symptoms and the control the participants have over whether their invisible symptoms are made known to others ('being seen'), or kept hidden ('staying invisible'). Participants described performing a 'cost-benefit' analysis across different situations to inform their navigation style, taking into account various factors, including whether they wish to be 'seen' or 'remain invisible'. We conceptualised this as striving to find a balance on a double-edged sword.

Making symptoms fit to life

Eleven participants described trying to make their invisible symptoms 'fit' to their lives, often characterized by 'pushing through' and attending to daily tasks despite feeling unwell. Symptom invisibility often allowed participants to 'push through' daily life undetected by others. Fay talked about needing to put her invisible symptoms "on the back burner" when she has responsibilities to consider:

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[35] Please see extended paper section 3.4 for additional comments on this theme and its related sub-themes

> *There are days where you have to forget the MS and really fight through things to get them done and be as normal as you can… times where being a mum has to take priority over how you feel physically…you have to do things that put you in discomfort because they are really important. (Fay)*

Daniel is responsible for his team of employees, and spoke about the "*brave face*" he puts on when he is struggling with invisible symptoms to maintain his managerial persona and avoid worrying others:

> *I almost have to sort of put a brave face on…even when I am struggling I do have to come across as being my normal self, almost as though there's nothing wrong…I've got to project to my staff that there's nothing to worry about...You don't want people to see that you're hurting. (Daniel)*

For some, pushing the invisible symptoms from their minds was a way of managing the symptoms in order to get on with life:

> *I try and push it away most of the time because I don't want it to be there. It's an annoyance, it's an irritation, so why would I think about it more than I have to? (Jennie)*

Making space for symptoms in life

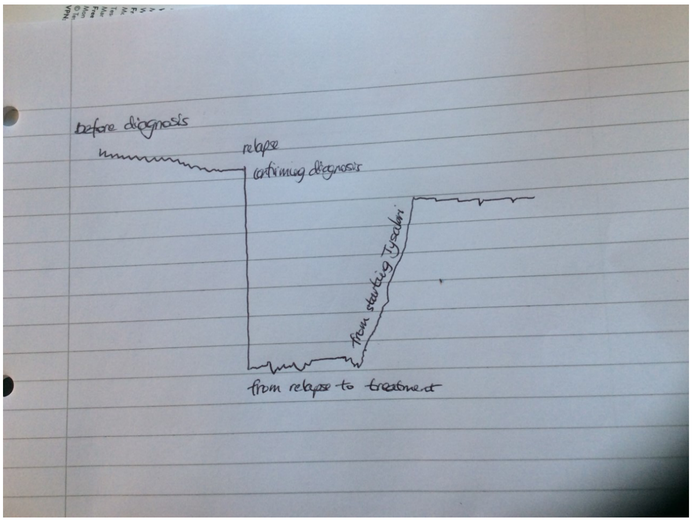
All participant accounts supported this subtheme, describing how they have 'made space' and adjustments in their lives for their invisible symptoms. Wendy spoke about being a "career girl" before being diagnosed with MS, and having to adjust her lifestyle to accommodate her invisible symptoms:

> *I figured I had to leave my old life behind and start a new one, because I couldn't possibly have continued at the rate, the pace that I was living my life...I had to put my life into perspective and think about how I was going to manage (Wendy)*

Ann talked about how she changed her working patterns, stepped away from unhelpful relationships, changed her medication, and made conscious decisions to reduce the impact of various stressors on her invisible symptoms. She illustrated this in a graph (Figure 10) depicting her MS journey and how this steadily improved after making changes:

> *I did various things to change the way I lived my life, and that really helped (Ann)*

Figure 10. *How my MS has progressed* taken by Ann



Jennie described feeling as though "the sun was going down" on her life when she was diagnosed with MS (depicted in Figure 11), however she has made and continues to make changes to her life to manage her invisible symptoms in a way that offers her a sense of agency. She reflected that this has helped her to manage the impact of her symptoms.

> *I kind of pre-empted and got into a slightly different role…that was the main change I made. I wanted it to be my choice, and not something that was pushed on me or decided by the illness…that was my way of exerting control over it…you need to know how to plan around it. (Jennie)*

As well as making larger life changes, participants also described a plethora of practical strategies they use to manage daily living with invisible symptoms[36]. Anniemac spoke about the technology she uses to support her with invisible symptoms, including using an Alexa Dot device (depicted in Figure 12) to help manage cognitive difficulties:

> *Alexa makes life so much easier. My MS brain struggles to organise! (Anniemac)*

Figure 11. *Sunset* taken by Jennie

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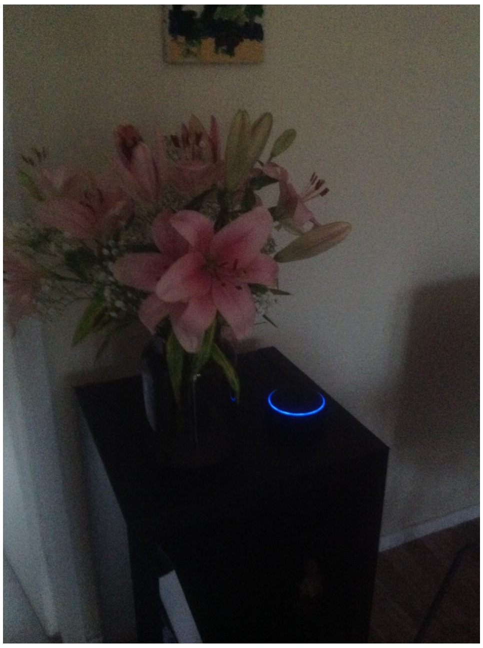
[36] Please see extended paper section 3.6 for a summary of management strategies shared by participants

Figure 12. *Alexa* taken by Anniemac



There was evidence for participants arranging their day-to-day lives around their invisible symptoms, and also of making mental space for the symptoms and accepting their impacts:

> *It's much better to embrace it…there's no point ignoring it because it's not gonna go away…let it be part of you but not the whole thing… it doesn't define you….it doesn't consume me like it used to. It's part of me, I've accepted that. (Stuart)*

<u>*Implications of navigation styles on symptom invisibility*</u>

This subtheme was supported by all participants. We found that the choice to either make space for invisible symptoms or fit them into life often had consequences for the invisibility of participants' symptoms, in terms of whether these remain hidden to others and 'stay invisible' or are made known and result in 'being seen'. These consequences were sometimes intentional, and at times unintentional for the participants. Claire described her reluctance to stay home from work when she struggles with fatigue, instead choosing to push through undetected by others. This sometimes results in the exacerbation of her symptoms, then her struggle becomes apparent to her colleagues:

> *I wasn't letting my team leader know that I was struggling…I was trying and trying and seeing how far I could go…I've been known to fall asleep at work… and the woman I usually sit next to, she's like 'I know when you're ill'. And I'm like 'oh dear' (Claire)*

Making space for the symptoms by participants making their needs known sometimes had the outcome of helping their invisible symptoms to be acknowledged and understood to a degree by others. However, sometimes despite participants' efforts to make others aware of their symptoms, the legitimacy conflict keeps them feeling invisible and unvalidated.

Finding a balance on a double-edged sword

This subtheme explicates how participants choose the way they navigate their invisible symptoms by taking account of their context, needs, wishes, potential consequences for the invisibility of their symptoms (i.e., 'being seen' or 'staying invisible') and the advantages and disadvantages of these outcomes. Participants described striving to strike a balance in life between pushing through symptoms and making space for them. Fay depicted this 'balancing act' in Figure 13 by capturing a set of scales. The ability to be flexible in her approach across different situations helped with this balance.

> *Everything is a balancing act…knowing what you can and can't do…on some days the MS is gonna make the scales drop really low…and there's gonna be days where you have to forget the MS and really fight through things to get them done and be as normal as you can (Fay)*

For many participants, their choices were influenced by the desire to maintain a sense of identity and not be defined by their MS and trying to balance this against their other needs and the impacts of their invisible symptoms. Stuart said his desire for others to be aware of his symptoms varied according to the situation:

> *Sometimes you don't want people to see but sometimes you just want to go 'look, that's what it is'…depending on the situation…with my dealings with the DWP [UK Department of Work and Pensions] I'd like to be able to say 'that's what's wrong with me'…But not necessarily day-to-day for meeting [other people] (Stuart)*

Figure 13. *Balancing Act* taken by Fay

Whilst being able to choose a navigation style offered a sense of control for people over their invisible symptoms and how they present to others, many participants experienced this choice as a double-edged sword where each choice has both costs and benefits. Jennie discussed this choice and the conflicts she experiences in daily life. By ‘pushing through’ she keeps her symptoms hidden from others, however this puts her in a very “*lonely place*”. However, making space for the symptoms and making them known can lead to unwanted input and worry from those around her:

> *I don’t know that I’d want everybody to see. Because you don’t want that feeling sorry for you… But it can be quite a lonely place to be. Sometimes you want people to know what’s going on inside but other times you think ‘actually no I don’t’… that’s a bit of a frustration, not knowing where you want people to be in the situation. What do you want from them, what you want from yourself...It’s sometimes really hard to know what you want…I don’t want people to judge me by what I’ve got, not what I am. (Jennie)*

Participants were often caught in a conflict of their wish for the reality of their invisible symptoms to be validated, and a desire to be treated as “normal” and no different to others.

Discussion

This photovoice study allowed us to explore and uncover people’s experiences of living with the invisible symptoms of MS, and the ways they manage and navigate their daily lives in relation to these invisible symptoms. To our knowledge, this is the first study to use visual methodology to unpack the lived experience of invisible MS symptoms. Our findings were consistent with those of our meta-synthesis (Parker et al., 2020), and provided us with rich information that allowed us to further elucidate and understand the implications of living with invisible symptoms and how people negotiate this. We have integrated our original framework (see Appendix B) with the findings from the present study and have outlined this as a conceptual framework in Figure 14. This is not intended as a theoretical model, but an arrangement of the themes to better understand them in relation to our research aims. The dashed lines in the framework represent links rather than linearity or causation[37].

Invisible symptoms were conceptualised as a discrepancy between the struggle people with MS experience internally, and what is visible externally to both onlookers and to the sufferer themselves. This is consistent with the available definitions in the literature and what has been expressed previously by those who experience these symptoms (Parker et al., 2020; Werfel & Trettin., 2020). Our findings expand on this and show the complexities of fully conceptualising the invisible nature of people's MS symptoms. This is due to the notion that not only can invisible symptoms not be outwardly seen by the person with MS or others, but the language with which to communicate these symptoms is not available. Participants described sensory experiences without tangible, observable evidence, which feel so abstract that they cannot explain these experiences accurately with words. This introduces a legitimacy conflict around the perceived 'realness' of people's invisible symptoms, where people cannot validate the symptoms for themselves or communicate this to others, and the invalidating

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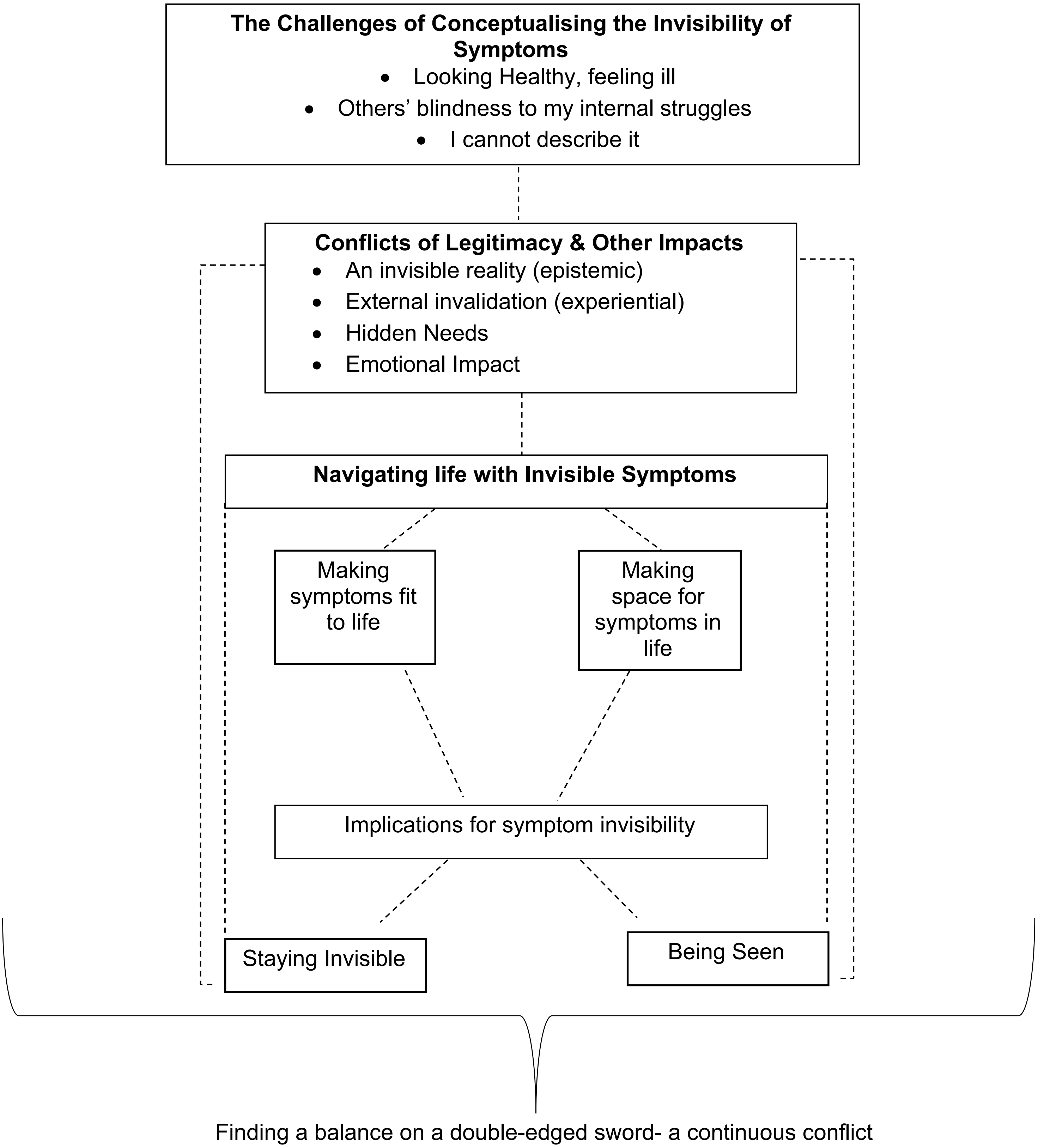
[37] Please see extended paper section 4.1 for further commentary on the integrated conceptual framework

responses of others lead the person with MS to doubt their own reality and experience of their symptoms. The lack of understanding from others compounded the invisibility of people with MS' needs and the emotional impacts of their symptoms, worsening this legitimacy conflict. Our findings here support the existing evidence whereby people with invisible MS symptoms feel ignored and dismissed, questioning the validity of their lived realities (Cadden et al., 2018; Parker et al., 2020).

The way people with invisible symptoms navigated daily life had two key functions, one was the management of the physical experience of the symptoms themselves and being able to complete daily tasks more generally, and the other was navigating symptom invisibility and the legitimacy conflict this presents. The invisibility of people's symptoms afforded them a choice as to how they navigated their lives, not accessible for visible disabilities. We uncovered two key navigation styles whereby 1) people with MS tried to make their invisible symptoms fit into their lives and 'push through' almost as if to ignore their invisible symptoms, and 2) people with MS consciously made space in their lives for their invisible symptoms and reorganised their lives around symptom-related needs. This was not only true of the way they overtly behaved, but also how they negotiated living with symptoms psychologically.

Figure 14. Conceptual framework to represent the relationship between themes identified from the analysis, and the deductive framework from Parker et al. (2020).



We learned that although some participants were more inclined to particular styles of navigating their lives, there was a general sense of striving to find a balance between ‘pushing through’ and ‘making space’ for their invisible symptoms, and participants often oscillated between these styles depending upon the context and their evaluation of needs in a given situation. Participants weighed up the advantages and disadvantages of navigation styles by considering their perceived roles and responsibilities, personal values, and having a sense of control over the way in which they present themselves to and are perceived by others.

Participants expressed an awareness that their navigation styles in a given situation inherently influenced the degree of invisibility of their symptoms. Many participants described making active choices to directly influence their symptom invisibility and to ‘be seen’ by making their invisible symptoms known to others, or to ‘stay invisible’ and continue to let their symptoms remain undetected. At other times, participants indirectly influenced their symptom invisibility as a result of ‘pushing through’ or ‘making space’. We learned that the impact of people’s chosen navigation styles at times had both intentional and unintentional consequences on whether their symptoms were ‘seen’ or remained invisible. For example, sometimes the approach of pushing through invisible symptoms proved effective in concealing an illness identity, however, sometimes led to participants being unwittingly ‘seen’ when the impact of their symptoms became apparent to others.

Many participants described trying to ‘stay invisible’ and ‘push through’ their invisible symptoms, particularly in work-place settings, which appeared to be motivated by a desire to protect the image they portray to the world, and to avoid becoming associated with a pitied illness identity. Goffman (1963) and Charmaz (1983) theorized that people with disabilities inherit a stigmatized ‘spoiled identity’ and loss of self which is shaped by other people’s (and general societal) responses to them. Two societal identities are suggested by Goffman (1963): the ‘stigmatized’, who are dismissed and discounted, and the ‘normals’ who appear not-disabled and are valued. Our findings are consistent with the conflict people with invisible symptoms of MS find themselves in, torn between being perceived as one of the ‘normals’ and being insufficiently supported or understood, or being one of ‘the stigmatized’. This conflict informs their choices as to how they navigate their invisible symptoms in daily life and within this negotiate their identities[38].

The choice to try to ‘be seen’ or ‘stay invisible’, and to ‘make symptoms fit to life’ or ‘make space for symptoms’, whilst offering a sense of control and agency for people with invisible symptoms of MS, was a source of conflict where people strove to find a balance on a ‘double edged sword’ as they navigated their lives. This is consistent with our previous findings and those of chronic invisible illnesses (Parker et al., 2020; Vickers, 1997), where choosing to ‘be seen’ in an attempt to be validated also involves the risk of stigmatization, but ‘staying invisible’ can harm people’s psychological wellbeing through keeping their needs

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[38] Please see extended paper section 4.2 for further discussion of findings related to existing literature and theory

hidden- both compounding the legitimacy conflict that people with MS seek to resolve.

Implications

Our study provides insight into the challenges faced by people living with invisible MS symptoms, and the ways they negotiate day-to-day living and their illness legitimacy and identities. It is important that clinicians and healthcare professionals are aware of the conflicts created by symptom invisibility so that they can offer appropriate support to people with MS. It may also be helpful, given that many participants reported doubting the reality of their own symptoms, to acknowledge this and discuss the neurological evidence for the symptoms with patients to help them resolve the epistemic issues highlighted in our study.

Hayes et al (2004) suggest that mental wellbeing and effectiveness in navigating life and its challenges centres around the ability to demonstrate psychological flexibility, which includes being able to contact the present moment, accept and make space for one’s experience and step away from avoidance of this, and to act in accordance with one’s values. These are skills that can be developed in order to flexibly respond to challenging contexts (such as living with invisible symptoms) in a way that is consistent with what is important and meaningful to the individual. Acceptance and Commitment Therapy (Hayes et al., 2006) focusses on supporting people to increase their psychological flexibility and has yielded some promising results in terms of improving psychological flexibility and reducing symptom distress for people with chronic, long-term health conditions (Graham et al, 2016). More specifically, ACT has been shown in some studies to be effective in improving quality of life and reducing psychological distress for people with MS (Barooti et al., 2019; Brown et al., 2016; Nordin & Rorsman, 2012). Although it is theoretically intuitive to suggest that improved psychological flexibility (and therefore ACT) could help people to navigate the invisible symptoms of MS, our study was not designed to explore this. To our knowledge, no research exists exploring the benefits of improving psychological flexibility through ACT with people who experience invisible symptoms of MS. ACT has been shown to reduce perceived stigma around pain as a symptom more generally (Whitney et al., 2019), however further research into how improved psychological flexibility could potentially support people with MS to navigate their invisible symptoms and the legitimacy conflict they present could have important indications for supporting people to live on the 'double edged sword'[39]. This could be investigated first by way of a single case experimental design (SCED) and then if psychological flexibility is found to lead to better management of living with invisible symptoms, the effectiveness of a specific ACT intervention for invisible MS symptoms could be tested using a Randomised Controlled Trial (RCT).

We found that photography facilitated the communication of invisible symptoms for people with MS and allowed people to voice these experiences which are often difficult to describe. Using photography in the therapeutic context could possibly support people with MS to express their experiences and make meaning of these. Phototherapy has been linked with increased self-awareness and positive therapy outcomes (Stevens & Spears, 2009; Saita & Tramontano, 2018). Future research could explore the potential of using photo-production in therapy

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[39] Please see extended paper section 4.3 for further discussion of implications of our findings

with people who have MS, and a SCED could be used to examine the effectiveness of this technique in terms of therapeutic outcome.

Limitations and Strengths[40]

Although a range of ages between 30-57 years and a range of years living with MS were captured, the study has limitations in terms of demographic diversity of the sample, with there being no ethnic diversity amongst participants. This allows for limited representation of the population group's perspective and excludes the voices of those from other cultures and ethnic groups for whom the experience of living with and managing invisible symptoms of MS may differ or offer new insights.

Data in qualitative research are subject to the researchers' interpretations which are likely to be influenced by their expectations, experiences and positions. We acknowledge that the first author who conducted the analysis had their own expectations based around previous research they have conducted in the area of MS and invisible symptoms. Whilst we used a rigorous reflexive process, the interpretation of the data cannot be completely separated from the expectations and position of the researchers. We acknowledge the potential for confirmatory bias in our results as a possible limitation, given that there was considerable overlap and fit with our deductive frame. Independent cross-verification of coding and themes, and transparent discussions within the research team throughout analysis helped to mitigate potential biases and improved the credibility and reliability of our findings. We evaluated the study against quality criteria to ensure

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[40] Please see extended paper section 4.4 or further critique of this study

that our research met the standards of good qualitative research and reporting (e.g., the CASP and Yardley's guidance; 2000).

Conclusions

Photovoice was shown to be a useful method in exploring people's lived experience of their invisible symptoms of MS and how they manage these. We found that people with MS navigate their lives with invisible symptoms not only by managing the symptoms generally, but also their invisibility. Evidence suggested that people with MS make often burdensome choices across a range of situations as to how they manage their symptoms, taking into account their needs and whether they wish for their symptoms to be known to others or not. Participants experienced conflict around these choices and valued the ability to respond flexibly to each context. Our research highlights the potential importance of supporting people with MS to validate their invisible symptoms and respond flexibly to the issues they present.

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Declaration of Interest Statement

Conflicts of interest: none.

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[41] References have been formatted according to journal style and therefore deviate from APA style

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Extended Paper

1. Extended Background

1.1. Further information about Multiple Sclerosis

1.1.1 Characteristics & Diagnosis

Multiple Sclerosis (MS) is a neurological condition and inflammatory disease of the central nervous system (and other select areas, such as optic nerves), and is characterised by axonal degeneration and demyelination. This occurs when the body's immune system incorrectly recognises healthy nerve tissue as foreign, attacking healthy cells. The loss of myelin sheaths disrupts nerve function, causing a range of debilitating symptoms, including but not limited to pain, cognitive issues, motor difficulties, sensory deficits and fatigue (Werfel & Trettin, 2020).

The characteristics of MS and its related symptoms vary widely between people. For some, the condition is characterised by periods of relapse and remission, and for others a progressive pattern occurs. Four courses or 'subtypes' of MS are recognised: clinically isolated syndrome (CIS), relapsing-remitting (RRMS), primary progressive (PPMS), and secondary progressive (SPMS) (Lublin et al.,2014). Regardless of MS subtype, the clinical presentation is usually characterised by uncertainty and unpredictability of symptoms and progression, and the possibility of neurological disability despite the use of treatments (Cocco et al., 2015; Melis et al., 2014; Werfel & Trettin, 2020). Diagnosis occurs through the identification of lesions on the brain and spinal cord, which can be accessed through Magnetic Resonance Imaging (MRI) procedures, as well as a range of other assessment considerations (Birnbaum, 2013; Calabresi, 2004).

CIS is diagnosed when a first episode of inflammatory demyelination is apparent, and if further clinical relapses are observed, the person is diagnosed with MS. RRMS is the most common subtype, affecting 85% of people with MS, and is characterised by unpredictably symptom flare-ups and relapses interspersed with periods of recovery and stability (Bogosian et al., 2017; International MS Federation, 2020). During the periods of stability, partial or complete remission of symptoms can occur, and RRMS is defined by an absence of illness progression

(Werfel & Trettin, 2020). If disease progression occurs gradually and without a relapse-remitting pattern, PPMS is diagnosed. SPMS is a diagnosis usually made following a worsening of symptoms and accumulation of disability after an initial period of RRMS and acknowledges the progressiveness and lack of recovery from the disease (Lublin et al., 2014). In around half of cases, progression from RRMS to SPMS usually happens within 15 years (Koch et al., 2010; Scalfari et al., 2014).

1.1.2 Epidemiology

MS is estimated to affect 2.8 million people worldwide and 127,000 in the United Kingdom (Mackenzie et al., 2014; MS International Federation, 2020). Global data shows that there are at least twice as many women (69%) with MS as there are men (31%), increasing to more than three times as many women than men in Western Pacific and South-East Asia regions (MS International Federation, 2020). Research shows that MS incidence and prevalence are significantly higher for people of White European ancestry than for people of other ethnicities (Albor et al, 2017; Cree et al., 2004; Langer-Gould et al., 2013; Wallin et al., 2012). The clinical manifestation and characteristics of MS are also thought to be influenced by a person’s ethnicity. For example, Black and Hispanic people are more likely to experience severe disease aggression and greater disability progression (Cree et al., 2004; Kaufman et al., 2003; Koffman et al., 2013; Rivas-Rodriguez & Amezcua, 2018).

People are usually diagnosed between ages 20 and 50, and the average age of diagnosis globally is 32 years, making MS the most common cause of disability for young adults (International MS Federation, 2020; National MS Society, 2020).

1.1.3 Treatment

There is no recognised cure for MS, however, inflammatory activity, relapse rate and disability progression can be slowed through the use of Disease Modifying Therapies (Birnbaum, 2013; Moss et al., 2017). A range of other pharmacological interventions are recommended by the National Institute for Health and care Excellence (NICE, 2019) for the management of individual symptoms (e.g. amantadine for fatigue, gabapentin for muscle spasticity). There is also growing support recommendations for the use of psychological interventions such as Cognitive Behavioural Therapy (CBT) for fatigue, and lifestyle strategies such as exercise and nutrition (Moss et al., 2017; NICE, 2019).

1.2 The Impacts of MS

Research has demonstrated a range of physical, psychological, social, emotional and cognitive impacts of MS on those who have it. As mentioned, MS is a progressive condition and involves a gradual worsening of symptoms and decrease in physical functioning, resulting in permanent physical disability. Within 15 years after onset, approximately 20% of people with MS become bedridden, and a further 20% require mobility aids (Rolak, 2003). It is approximated that within 30 years of diagnosis, 80% of people with MS require the use of a unilateral cane to mobilise effectively (Compston & Coles, 2002). Symptoms can also include a deterioration in cognitive functioning (discussed in section 1.3) as a result of lesions and disrupted transmissions to and from the brain.

In addition to living with the effects of the symptoms themselves and the physical disabilities they create, the symptoms caused by the MS often have reciprocal and complex inter-relationships with other difficulties that present secondarily, and in turn have an exacerbating effect on the symptoms themselves (Werfel & Trettin, 2020) For example, pain, muscle spasticity and bladder issues can lead to difficulties sleeping, and then insomnia can worsen the experience of the primary symptoms.

The emotional toll that having MS takes on the individual is acknowledged in the research, and common emotional responses include grief, anger, shame, resentment, confusion and overwhelm (Reitman & Kalb, 2012). Stress can be defined as “a process that involves an environmental triggering event (stressor), which overwhelms the individual’s ability to cope or adapt, and results in a psychological or biological consequence" (Heesen et al., 2007, p.144). The role of stress as both a consequence of experiencing the symptoms of MS and something that impacts the symptoms themselves has been highlighted. Meta-analysis has supported the existence of a relationship between stress levels and clinical exacerbation of symptoms (Mohr et al., 2004; Lovera & Reza, 2013). Those with MS therefore find themselves in a double bind, where living with MS is a stressful experience, and stress can make the symptoms worse (Werfel et al., 2016).

Living with MS has been shown to affect people's mental health and wellbeing. Depression is more common in people with MS than those with other chronic illness and neurological disorders (Lobentanz et al., 2004). Although there is some heterogeneity in prevalence rates for anxiety and depression in people with MS, rates are significantly higher than in the general population (Beiske et al., 2008; da Silva et al., 2011; Siegert & Abernethy, 2005) and the associated burden is higher (Marrie et al., 2015). In the largest systematic review to date, pooled mean prevalence of depression was 30.5%, and 22.1% for anxiety (Boeschoten et al., 2017). Epidemiological data suggest that suicide rates for people with MS are approximately twice that of the general population (Feinstein & Pavisian, 2017). Anxiety problems in people with MS have been found to be related to decreased social support and psychosocial stressors in response to the MS (Korostil & Feinstein, 2007). Mental health difficulties in MS are associated with reduced quality of life and treatment adherence, and increased symptom severity, disability progression, negative health perception and mortality rates (Fiest et al., 2015; Lobentanz et al., 2004; Marrie et al., 2015; McKay et al., 2018; Moss et al., 2017). Given the links between emotional and psychological distress in MS and negative health outcomes, and evidence that these difficulties often worsen and persist if adequate support is not offered (Burnfield & Burnfield, 1978), it is important for clinicians to understand and support people with MS around difficulties related to the condition and its symptoms.

MS has also been shown to have negative impacts on an individual's social functioning and networks (Mohr et al., 1999; Halper, 2007). Functional changes as a result of symptoms and relapses can affect ability to participate in activities of interest, leisure and recreation, including social activities (Krupp et al., 1988; Krupp, 2000; Werfel & Trettin, 2020). People may also retreat from social activities for reasons related to the emotional impact of their MS (Halper, 2007). The literature highlights the strains that MS places on close interpersonal relationships, and the impact of societal stigma around disability on people with MS' experiences (Bennett, 2002; Marinelli & Dell Orto, 1977; Yamamoto, 1977). Maintaining employment is also enormously impacted by MS, threatening not only the individual's economic status, but their sense of role in society (Rumrill, 2004). People with MS are often faced with the challenge of adjusting their self-identity to accommodate living with a chronic illness, bringing with it a range of psychosocial impacts (Werfel et al., 2016).

1.3 Invisible Symptoms of MS

The journal paper highlights commonly cited 'invisible' symptoms of MS, so-called because they are not readily apparent to others. Here, some further information is shared about the nature, prevalence, and impacts of these symptoms.

As stated in the journal paper, fatigue is considered to be the most common, chronic and debilitating symptom of MS, and is invisible in nature (Giovannoni, 2006). A lack of consensus exists around the definition of fatigue; however, it has been conceptualised as an overwhelming sense of mental and physical tiredness that is out of proportion to a 'normal' tired feeling (Krupp, 2003). MS fatigue is understood to be distinctly different from fatigue associated with other conditions or healthy individuals, and has been shown to carry significant psychological and physical burdens in the completion of everyday tasks and the disproportionate amount of effort needed to do them (Barak & Achiron, 2006; Krupp et al., 2010; Leocani et al., 2008). MS fatigue has been found to affect the sufferer's quality of life, employment, self-esteem and sense of identity (Aronson, 1997; Janardhan & Bakshi, 2002).

Cognitive difficulties are recognised as debilitating, can occur at any stage of MS, and refer to dysfunction in the areas of memory, attention, information processing, executive function such as problem solving, verbal fluency and word-finding, and visuospatial analysis (Benedict et al., 2006; Bobholz & Rao, 2003; Chiaravalloti & DeLuca, 2008; Deloire et al., 2011; Sumowski et al., 2018). Even people with mild MS-related cognitive deficits experience greater difficulties in daily activities, socialising and work (Rao et al., 1991).

Pain is another prevalent and often severe invisible symptom of MS, (Foley et al., 2013). More than 88% of people with MS report pain in more than one area of their body, and the average number of bodily pain locations is six (Ehde et al., 2006). Many people with MS experience Trigeminal Neuralgia, a neuropathic pain affecting the face and head (Texakalidis et al., 2020). Although pain can be acute, it is usually experienced as chronic, and has been shown to negatively affect physical and emotional functioning, as well as overall quality of life (Kerns, 2000; Kerns et al., 2002; Namerow, 2011). Greater pain severity has been found to be associated with severity of depression and anxiety symptoms for those with MS (Kalia & O'Connor, 2005).

In addition to pain, many people with MS experience sensory symptoms such as paraesthesia (tingling), numbness, dysesthesia (burning or prickling sensations), Lhermitte's phenomenon (electrical shock feeling down the neck and spine) and temperature sensitivity (Afshari et al., 2016; Beckmann et al., 2015; Christogianni et al., 2018; Indaco et al., 1994; Rae-Grant et al., 1999; Sanders & Arts, 1986). Sensory deficits and abnormalities have been reported to be present for around 80% of people with MS and have a detrimental and significant impact on quality of life (Heijenbrok et al., 1992; Sanders & Arts, 1986; Svendsen et al., 2005). Sensitivity to heat and cold occurs when neurological symptoms are exacerbated temporarily by a change in environmental or internal temperature (Christogianni et al., 2018; Simmons et al., 2004). The sensitivities can make it difficult for people with MS to exercise, complete daily activities, or expose themselves to particularly warm or cold environments (Simmons et al., 2004; White & Dressendorfer, 2004).

Where the inflammation or demyelination in MS affects the optic nerve, this can cause a range of visual deficits for people with MS which are unobservable.

These deficits include eye pain and most commonly, optic neuritis (Lotan et al., 2018). Optic neuritis in MS can affect vision in a number of ways, ranging from blind spots or poor vision in one eye to complete loss of sight in both eyes. Colour vision can also be affected, and double vision and phosphenes (‘flashes’ of light when moving the eyes) can occur (UK MS Society, 2020). Although functioning tends to be restored a few weeks after the onset on an episode of optic neuritis, the extent of the restoration is uncertain, and it some cases does not occur (UK MS Society, 2020).

Bladder and/or bowel dysfunctions are often considered to be the most distressing symptoms of MS and are cited in the literature as being invisible in their nature in that the associated problems are not usually apparent to onlookers (DasGupta & Fowler, 2003). Approximately 75% of people with MS develop bladder dysfunction (Betts, 1999; Marrie et al., 2007). The most common neurogenic bladder problems are urinary incontinence, bladder urgency (even when the bladder is not full), and incomplete emptying of the bladder (DasGupta & Fowler, 2003). Bladder problems are extremely disabling, negatively impacting on quality of life, social functioning and ability to adhere to daily routines (DasGupta & Fowler, 2003; Khan et al., 2009). A study by Hemmett & Colleagues (2004) found that 70% of people with MS classified the impacts of their bladder symptoms on their lives as ‘moderate’ or ‘high’. The most common bowel dysfunctions in MS are that of faecal incontinence and constipation, which often co-exist and occur in approximately 50-80% of people with MS, however this number varies depending upon the severity of the condition (McClurg et al., 2017). These issues have been reported to be understudied, perhaps due to the sensitive and personal nature of the symptoms and their impacts (DasGupta & Fowler, 2003).

Sexual dysfunction is a common invisible problem for people with MS, with between 40-80% of women and 50-90% of men reporting this (Krupp, 2000; Schairer et al., 2014). Symptoms of this nature include erectile dysfunction, diminished libido, genital numbing and vaginal dryness. Other invisible symptoms such as fatigue and pain can also interfere with a person’s ability to be intimate with another. Sexual dysfunction can be present even in the early stages of MS where severe disability is not present (Demirkiran et al., 2006), and evidence demonstrates a detrimental impact of these symptoms on mood, relationships and quality of life (Domingo et al., 2018; Schairer et al., 2014). It is acknowledged in the literature that these symptoms can cause discord in the intimate relationships of people with MS, where partners may misinterpret the changes in sexuality as related to themselves and the relationship rather than the symptoms (Foley & Werner, 2000). It is uncertain as to whether these misinterpretations are linked to the often-invisible nature of these symptoms.

Depression and anxiety are recognised in the literature as being primary invisible symptoms of MS and related to brain lesions which affect the ability to regulate affect, and are also believed to develop as a secondary response to the impacts of the other primary symptoms and their influence on mental wellbeing (Bakshi et al., 2000; Butler et al., 2016; Feinstein et al., 2004; Feinstein, 2011; Gold et al., 2010; Kiy et al., 2011; Pujol et al., 1997). It is widely accepted that complex relations exist between biological and psychosocial factors in the development of depression and anxiety in people with MS, and that these are difficult to separate and understand in isolation (Butler et al., 2016; Shelton et al., 1991). Therefore, they can be understood in the context of this study as invisible symptoms.

In addition to the impacts of the invisible symptoms outlined in this section, the journal paper highlights research and literature to support the notion that people with MS encounter additional and qualitatively different difficulties brought about by invisible symptoms, related to their invisible and unobservable nature. It is therefore deemed important to understand not only people's objective neurological disabilities related to MS, but of their subjective sense of health beyond what can be 'seen'' (White et al., 2008). The majority of studies investigating the impact of invisible symptoms of MS on a physical, psychological, emotional, social, and cognitive level have focussed on individual symptoms in isolation (Kratz et al., 2016; Olsson et al., 2005). However, it is widely known that each person with MS will experience a unique combination of symptoms, many of which are invisible (Christogianni et al., 2018). It is therefore important to understand the impacts of a constellation of invisible symptoms as opposed to exploring these in isolation.

1.4 Stigma and MS

As discussed in the journal paper, stigma and social impact are deemed a significant part of people's experience with MS, and issues specific to invisible symptoms are described. Social stigma is described as a psychosocial process by which an individual is 'marked' (because of a particular characteristic such as having a chronic illness) as having a spoiled identity that is of lesser value than other people who do not bear the 'mark' and are therefore regarded as 'normal', and being rejected, excluded and discriminated against as a result (Broersma et al., 2018; Dovidio et al., 2000; Goffman, 1963).

In health research with people who live with chronic illness, stigma is recognised as a social determinant of health. Those who are stigmatized experience discrimination, stress, social isolation, and loss of status which can limit their access to important resources (e.g. employment and medical care), consequentially impacting their health (Hatzenbuehler et al., 2013; Link and Phelan, 2006; Mak et al., 2007). The psychological stress from the experience of stigma can also negatively impact the immune system and lead to worsened health outcomes (Cohen et al., 2007).

In addition to social or 'enacted' stigma, people can experience self-stigma in relation to their illness, described as the internalisation of the emotional, behavioural and cognitive impacts of other people's negative and stigmatising attitudes (Broersma et al., 2018; Rao et al., 2009). In their model of internalised stigma, Corrigan, Watson and Barr (2006) outline a process whereby a person experiences stigma from another individual. The stigmatised person then becomes aware of the negative attitudes related to their illness and develops a sense of agreement and alignment with these views, internalising them. Internalised stigma has been shown to create psychological distress in the sufferer, and is linked to increased depression, anxiety and poorer quality of life in those with chronic illness (Earnshaw & Quinn, 2012; Earnshaw et al., 2013).

It has been found that people with MS who experience stigma report a lower overall quality of life and were more likely to report productivity losses at work (Anagnostouli et al., 2016; Hategeka et al., 2017). In terms of the psychological consequences for people with MS, experienced stigma has been found to be a predictor of depression (Cadden et al., 2018) and to affect resilience (Silverman et al., 2017). Stigma in MS is also linked with low self-esteem and low self-efficacy in both people with RRMS and SPMS (Demircia & Yilmaz, 2019).

Cook and colleagues (2016) found that most people with MS reported experiencing anticipated stigma (experiences of or concerns about biased treatment from others) and isolation stigma (a sense of being socially isolated as a result of MS stigma). Anticipated stigma was found to predict people's efforts at concealing their MS (Cook et al., 2016). This study used a small sample, however, lends support to a wider evidence base demonstrating that social stigma is a significant concern for people with MS (Rivera-Navarro et al., 2007). One study with 14 people with MS found that people strategically made decisions around concealment and disclosure of their MS in response to perceived social pressures, particularly in the workplace (Grytten & Maseide, 2005). It was also found that the embodied perception of illegitimacy was experienced as a dimension of stigma (Grytten & Maseide, 2005). Other qualitative research has highlighted people with MS' concerns about being stigmatised and stereotyped (Vickers, 2010). Theoretical understandings of stigma indicate that people adopt various strategies to mitigate future stigma experiences and use evaluation of the successes (or otherwise) of these strategies to inform their future responses (Lazarus & Folkman, 1984; Miller & Kaiser, 2001). This suggests that people are likely to improve the way in which they manage stigma experiences over time, which has been supported in a study of people with MS where perceptions of stigma decreased over time (Spencer et al., 2019). Although stigma has been highlighted as an important issue in MS more generally, research into stigma and invisible symptoms is very limited.

Joachim & Acorn (1999) combined the results from a meta-study of qualitative research with a review of quantitative literature in relation to stigma and chronic illness. They proposed a theoretical framework to describe the way that people with chronic illness may cope with stigma by either disclosing (and risk suffering further stigma) or concealing their condition (and pass for 'normal'), and how these decisions are often based upon whether the condition is visible to others or invisible. Those who have visible conditions are acknowledged as having less choice about the way they manage stigma than those with invisible or mixed visible/invisible conditions. The framework is grounded in Goffman's (1963) ideas that stigma is experienced in two ways whereby an individual is either discredited or discreditable. A discredited person is described as showing visible signs of being different (or in this case, having visible signs of disability), leading them to be immediately stigmatised by others before they have the opportunity to show their other qualities beyond their assumed differences (Goffman, 1963). The disclosure of their disability is often automatic and out of their control. Some people may try to manage the stress of interpersonal issues with a visible disability by 'covering', where they attempt to minimise or 'downplay' the effects of the condition (Joachim & Acorn, 1999). Covering can take the form of joking about one's own disability in an attempt to make others feel more comfortable and to cultivate a sense of belonging with this group (Goffman, 1963). People may also manage being discredited by isolating themselves into subgroups with similar characteristics, challenging and resisting the status quo, and disregarding the painful occurrences associated with the stigma (Joachim & Acorn, 1999).

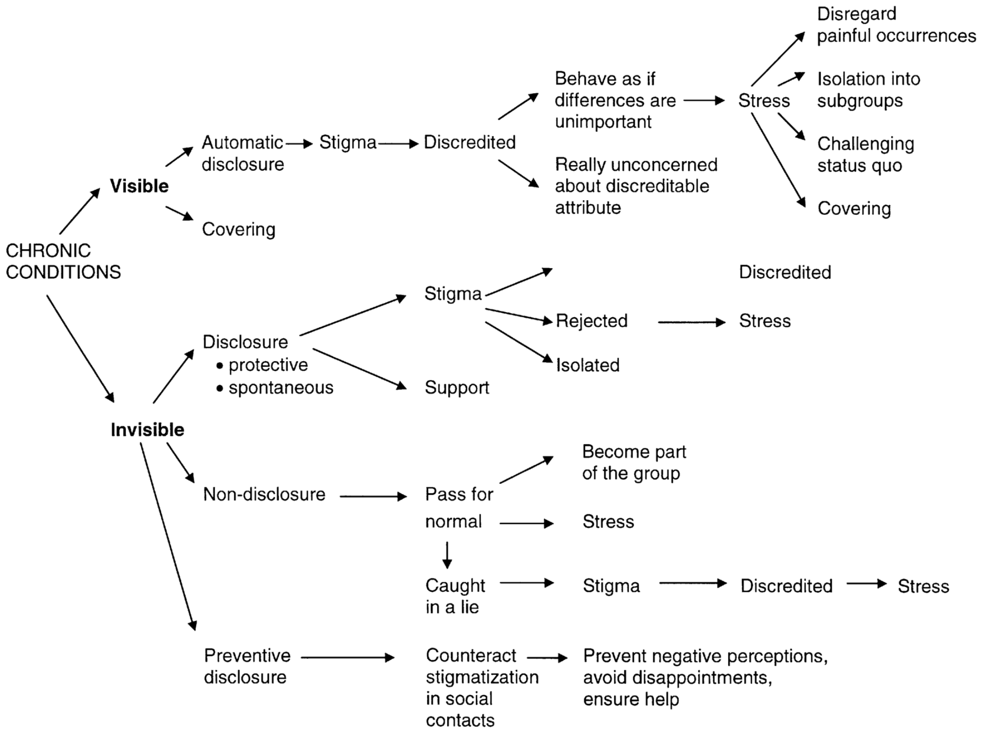
Goffman proposes that if a person's differences (or, in this case, their illness or symptoms) are invisible, they are discreditable but not yet discredited. The person must therefore decide whether to disclose or to conceal their condition. If the individual discloses, they risk being stigmatised for their illness, however with select individuals, disclosure may lead to increased support and understanding (Joachim & Acorn, 1999). People with invisible conditions also have the option not to disclose and attempt to 'pass for normal', deliberately concealing their illness in what is described as a stressful act (Goffman, 1963). If a person is successful in concealing their condition, they are likely to become a part of the 'normal' group, however they risk being 'outed' if they cannot conceal their differences fully and thus be discredited and stigmatised.

A person who discloses their condition may do so in a number of ways (Charmaz, 1991). Protective disclosure is planned in such a way that the individual controls what and how they disclose about themselves, and who to. Spontaneous disclosure is more emotionally led and unplanned. Preventative disclosure (or 'informing') is where the decision to disclose is based upon the risk of others finding out about the condition and the expected social consequences of this (Troster, 1997). This method of disclosure is thought to promote greater control over the response of others and sets expectations around help the individual might need (Joachim & Acorn, 1999). Figure 15 summarises Joachim & Acorn's (1999) model.

Little is known about how people decide whether and when to disclose and when to conceal their illness. Many of the sources on which the above framework was based were from over 20 years prior to its publication, and so it's relevance in the present day may be questioned. Our meta-synthesis (Parker et al., 2020) using more recent studies focussed on the notion of invisibility in MS, and found that symptom invisibility impacted whether people with MS chose to 'stay invisible' and not disclose their symptoms, or to 'be seen' and to disclose this to others. Lending support to Joachim & Acorn's model (1999), we found that invisibility offered people with MS a choice as to how they navigate life with their symptoms in choosing whether to disclose or not. The meta-synthesis found that people could chose to disclose or not disclose specific symptoms across a range of situations, and that navigating these choices can be burdensome and stressful (Parker et al., 2020). Unlike Joachim & Acorn's model (1999), different types of disclosure were not identified. Joachim & Acorn (1999) proposed that non-disclosure resulted in positive outcomes when successful concealment occurred, however that the underlying stress of being 'found out' remained. Our meta-synthesis uncovered additional stressors whereby as a result of 'staying invisible,' people's needs remain hidden, and they feel forced to cope alone. This choice to remain 'invisible' appeared to create a self-fulfilling prophecy, perpetuating a continued cultural lack of understanding that keeps the individuals trapped in their 'invisibility', and the stigma surrounding MS that contributes to the felt need to 'stay invisible' remains unchallenged and unchanged.

Figure 15

Model of stigma and factors that influence disclosure or non-disclosure in chronic illness (taken from Joachim & Acorn,1999).



1.5 Symptom Management in MS

People with chronic conditions seek find ways to self-manage their symptoms (Bodenheimer et al., 2002). As mentioned in the journal paper, research suggests that people with MS find practical ways of managing their invisible symptoms. Knaster and colleagues (2011) conducted focus groups with 12 people with MS. They found that participants were motivated to self-manage their symptoms out of necessity when they could not function in the same ways any longer, and often sought out information about MS to inform their management strategies. People with MS described a range of management strategies they used in addition to pharmaceutical intervention: including: ‘pacing’ and taking naps throughout the day, making adjustments to working schedule and hours, regularly exercising, enhancing their support system and in particular seeking our peer support from others with MS (Knaster et al., 2011). Another qualitative study found that pacing and planning activities, and developing supportive social network were among helpful management strategies for people with MS (Frost et al., 2017). Although these studies did not focus specifically on the management of invisible symptoms (and the latter study focussed only on people with progressive MS), the strategies described were applied to invisible symptoms including fatigue and depression.

Some studies have focussed on individual invisible symptoms rather than managing a constellation of symptoms. Norton and Chelvanayagam (2010) conducted a survey with 155 who have MS and detailed the variety of practical strategies people used to manage their neurogenic bowel problems. Strategies included increasing nutritional intake in their diet and exercising more. Those with constipation described adjusting their position when seated on the toilet to aid their bowel movements, and those with incontinence reported using continence pads to provide added security when leaving their homes (Norton & Chelvanayagam, 2010). Another study exploring the experience of fatigue for people with MS found that people consciously reduced their energy consumption by limiting their activities and exertion (including accepting help from others to do energy consuming tasks), and also that they used planned opportunities for rest and recharge (Stuifbergen & Rogers, 1997). The study also found that participants sourced their management strategies from MS publications, advice from family and friends, and through self-discovery. Practical management strategies are largely indicated and understood, however the ways that people manage the impacts of the invisibility of their invisible symptoms is less understood.

2. Extended Methods

2.1 Design

We adopted a qualitative design and ‘Photovoice’ methodology, which is a visual and participatory approach to research.

2.1.1 Rationale for Qualitative Research

Qualitative research concerns itself with how people make sense of and manage their world and experiences (Willig, 2013). Qualitative approaches to research allow the opportunity to explore, in depth, different phenomena and how they are experienced and perceived by a group of people (McGrath et al., 2019). Such methods are widely used in health psychology research to bring new insights to our understanding of health and illness (Yardley, 2000). Qualitative methods are recommended when researchers wish to explore a person’s experience of an illness, as these methods have the potential to give voice to marginalised groups in society (McGrath et al., 2019). It was therefore deemed the most appropriate method to address our research aims.

Qualitative and quantitative research differ in terms of their assumptions about the nature of reality and whether it can be directly measured (Newman et al., 1998). Qualitative research is grounded in the assumption that we cannot directly access and measure complex realities or find cause-and-effect patterns (in contrast to quantitative research), but that we can understand these phenomena by actively constructing them through investigating people’s accounts of these realities (Silverman, 2016). Qualitative method, therefore, was also consistent with our epistemological stance appropriate to our research aims[42].

2.1.2 Photovoice

Community-based participatory research has emerged as a paradigm that helps to ‘bridge the gap’ between science and practice, by collaboratively engaging people from particular communities in the research process in order to address healthcare inequities (Wallerstein & Duran, 2010). These collaborations can lead

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[42] Please see section 2.3 for further information about epistemology and our stance

to deeper understandings of the health needs of a population and the changes needed to address these (Cabassa et al., 2013).

Photovoice is a participatory visual research method, whereby people are asked to produce their own photographs or images in order to capture their experiences and perspectives on a particular aspect of health and illness (Lorenz & Kolb, 2009; Topcu, 2015). These people can then be asked to write narratives about the images they produce or asked to discuss them in either group or individual interview settings to help uncover meaning (Topcu, 2015). The method was originally developed by Wang and Burris (1994) as a health promotion tool to support participants to better understand their own health needs and to create change in the health policies that impacted on them. Photovoice is grounded in three theoretical frameworks together with health promotion principles: feminist theory (Reinhartz, 1992), documentary photography (Rosler, 1989), and education for critical consciousness (Freire,1970; Wallerstein & Bernstein, 1988). Photovoice has been used increasingly in health research, and is regarded as a powerful tool for advancing health equity by promoting empowerment and giving voice to marginalised groups, shining a light on the issues that impact them and communicating these to healthcare providers, policymakers and members of the public (Evans-Agnew & Rosemberg, 2016; Minkler and Wallerstein, 2008; Wang, 1999).

2.1.2.1 Rationale for using Photovoice

Visual methods are a growing field in psychology research (Balmer et al., 2015) and have been used in numerous studies to generate rich data that may be difficult to access using other approaches (Frith et al., 2005; Pain et al., 2012; Rose, 2007). Researchers argue that visual methods give detailed understanding and insight into the lives of the target population, allowing access to this through the participants' own 'lens' (Del Busso, 2011; Silver, 2009). This comes from the notion that people do not communicate through language alone, and photographs can give access to difficult to articulate, abstract experiences (Frith & Harcourt, 2007). Harper (2002) goes further, arguing that because visual processing brain regions are older than those used for verbal processing, visual methods do not just elicit more information, but yield a different kind of data. Visual data can be analysed in its own right or used to elicit verbal data which is then analysed (Silver, 2009). Given that the literature pertaining to the invisible symptoms of MS highlight struggles that cannot be clearly seen by others (Parker et al., 2020), we felt that visual methods would help us to uncover participants' internal experiences through a medium not used before with this population.

Visual methods have been found to serve as a memory prompt for participants in interviews, leading to accounts that would otherwise have not been available to them (Radley & Taylor, 2003). Given that memory problems are a common invisible symptom of MS, such an opportunity to capture 'in-the-moment' experiences rather than relying on recall at a later date, and the visuals serving as recall prompts during the interview stage, made this a particularly attractive method for this population. Indeed, our approach allowed participants the option to send images to us as they went through their daily lives, capturing experiences as they happened. Collier (1957) found that interviews where photographs were discussed and used to prompt discussion demonstrated increased participant engagement, rapport and memory recall, and decreased fatigue (also an important consideration for this population) compared to interviews that did not use this approach. It therefore was deemed a sensible choice to adopt visual methods with our study population given the types of symptoms our participants would likely experience during the research process. Visual methods that involve participants producing their own images for the purpose of the research has been argued to empower participants by giving them agency and control of the research process (Del Busso, 2011). This was also taken in account for this study, as it was important for participants to have an active role in the research process and for their 'voices' to be heard.

Photo-elicitation studies use already existing images in research interviews to elicit discussion, whereas photo-production involves use images that are created during the research process (Silver, 2009). Photovoice, however, involves the participants producing these images and speaking about the way in which they apply to their own lives and experiences, and are a method of promoting social change for marginalized groups (Silver, 2009). Of all the visual methods available to us, we used Photovoice because it is the most well established and well-defined visual method which encompasses participatory principles and aims to ‘give voice’ to participants and potentially change the way their public presence is defined, particularly when their needs are overlooked or ignored (Lal et al., 2012; Wang et al., 2000). This was important for people with MS who often report feeling stigmatised and dismissed (Grytten & Maseide, 2006; Parker et al., 2020;), and also because the lack of objective evidence for their invisible symptoms limits the way in which their public presence is defined, and photovoice can offer a way to do this visually. Lal and colleagues (2012) found in their literature review that Photovoice can be successfully used to address a wide range of research topics, including exploring people’s lived experience of illness and examining the interaction of individuals with their environments. As these topics fit with our own research aims, this provided further evidence of Photovoice as an appropriate method.

Finally, Photovoice is said to offer an alternative to positivist ways of ‘knowing’ about health issues by listening to and learning from people’s own perspectives of their health issues (Wang, 1999). Using this method, we are able to combine objective images with subjective accounts of their meaning to gain insight into the realities of people living with illness (Wang & Burris, 1994). This is consistent with our epistemological stance, discussed later in section 2.3.

2.2 Patient and Public Involvement and Engagement (PPIE)

The valuable and important role that Patient and Public Involvement and Engagement (PPIE) has in qualitative research is widely acknowledged and has been shown to enhance research quality and its relevance to the population being studied (Brett et al., 2014; Gillard et al., 2012; Jennings et al., 2018). PPIE is widely encouraged in health care research internationally, and in the United Kingdom it is a requirement in most publicly funded research (Department of Health, 2006; Mosavel et al., 2005; Schneider et al., 2004). PPIE has been defined as patients, carers and the public taking an active role in the research process, including involvement and collaboration in study design, delivery and dissemination (Ashcroft et al., 2016). A systematic literature review found multiple advantages to having PPIE in health and social care research, including ensuring the appropriateness and relevance of the study objectives, design and materials, and improving overall study quality (Brett et al., 2014). Previous research has also highlighted the benefits of PPIE in qualitative data analysis, and input from PPIE members can correct misinterpretations made by the researcher and shape the way in which the findings are communicated (Brett et al., 2010; Staley, 2009).

The importance of PPIE being valued by the researchers and purposeful as opposed to tokenistic in nature has been emphasised (Goodare & Lockwood, 1999; Lemonsky, 2015). Muller et al. (2019) state that meaningful PPIE occurs when PPIE contributors are active members of the research team and are involved in all aspects of the research process.

In the design phase of our study, the first author (LSP) presented the initial research idea to a local PPIE group for MS. The group consisted of people who have a diagnosis of MS across a range of subtypes, carers, and MS researchers and clinicians. The presentation included an opportunity for PPIE members to ask questions, share thoughts and raise concerns with the first author (LSP). We gathered anonymous feedback from 9 PPIE members (5 people with MS and 4 carers) using a questionnaire that enquired about views on our study proposal in five areas (the study purpose, proposed recruitment, method, intended outcomes, and service user involvement). The questionnaire used a five-point Likert scale and asked PPIE members to provide a rating for each area (1 was very poor and 5 was very good)[43]. Overall, the feedback was very positive, and the average scores are presented in Table 8 below. Members commented on the importance for them of increasing public and professional understanding of invisible symptoms in MS. Some members used the additional comments section to express their interest in participating and one member commented that they felt it was a “very interesting study”.

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[43] Please see appendix D for a copy of the questionnaire given to PPIE members

Table 8

PPIE member feedback on initial study proposal

Question	Average Score (1-5 scale)
Do you think the purpose of the research is clear?	4.4
How likely are we going to get people to sign up for this research?	4.4
Do you think the way we plan to conduct this research is appropriate?	4.4
Do you think the outcomes (things we hope to change) are appropriate?	4.3
How well do you feel we have planned to involve people with MS and carers during the development of the intervention?	4.5

The first author (LSP) also spent time at an MS Clinic in a general hospital on an approved 'insight visit' and had the opportunity to discuss the project idea with several MS patients who provided positive feedback and interest. One person in particular expressed their appreciation for a study which attempts to gain insight into symptoms they felt were poorly understood by people.

Following the presentation with the PPIE group, a member expressed interest in contributing to the project as a member of the research team. They have contributed to the study design, recruitment and other participant-facing materials, eligibility screening, documents for ethics submission, analysis and write-up. They will also be involved in supporting the wider dissemination of our findings.

2.3 Epistemological Position

The differences in research methodologies across studies are rooted in and guided by the philosophical and theoretical views that are assumed (Al-Ababneh, 2020). Ontology relates to the nature of reality and whether this is assumed to be constructed by human interpretations and understanding, or whether reality exists independent of human consciousness (Al-Ababneh, 2020; Braun & Clarke, 2013). Epistemology looks at the nature of knowledge and how we access valid and acceptable evidence of knowledge (Braun & Clarke, 2013; Hamlyn, 1995; Saunders et al., 2009). It is a lens through which we can look at the world and make sense of it. Researchers adopt a particular lens out of multiple positions when seeking to answer a research question, to help make sense of the relationship between themselves and the phenomenon they seek to investigate (Guba & Lincoln, 1994). Crotty (1998) outlines the way in which epistemology informs research methodology, which in turn determines which methods are chosen to explore the research question. Epistemology guides what can be said about the data and how meaning is theorised.

We have adopted a position of critical realism in the present study. This stance assumes that reality exists independently of human knowledge and beliefs, however our access to this reality is socio-culturally mediated and therefore never directly obtainable (Al-Ababneh, 2020). Both a reality that is external to humans and a reality constructed by humans is acknowledged here (Oliver, 2011). Critical realism indicates that the language people use can provide access to their personal interpretation of reality, and the research then creates interpretations of these accounts (Braun & Clarke, 2013). Therefore, we cannot identify or understand the reality that exists with any certainty, and we can only interpret this through the discourses of others and through empirical domains which investigate more linear and less complex, more accessible realities (Roberts, 2014; Sayer, 2004).

In relation to this study, we assume that there is an objective and physical reality to invisible symptoms, however that participants' experience of their symptoms is interpreted subjectively and is constructed by the participant in collaboration with the researcher. This approach requires that concepts such as invisible symptoms are examined in the context from which they emerge, in order to remain mindful of biases and social forces that shape subsequent understandings (Pilgrim & Bentall, 1999).

2.4 Study Sample and Participant Recruitment

2.4.1 Sample size

The recommended number of participants for photovoice studies is 7-10, based upon prior research and similar studies (Wang,1999). For qualitative research using Thematic Analysis (TA), 10-20 interviews are recommended in order for themes and patterns to be identified within the data (Braun & Clarke, 2013). We considered using the concept of ‘data saturation’ to determine sample size. A widely used principle, data saturation refers to meeting the point where introducing new data into the analysis would not generate any new knowledge (Glaser & Strauss, 1967; Sandelowski, 1995). This approach, however, is grounded in the idea that data collection is theory-driven and that there is an absolute truth to uncover and a ‘knowing’ when this has been reached (Vasileiou et al., 2018). As this position was not completely consistent with our own epistemological stance, we did not focus on data-saturation. We adopted an alternative approach of data ‘sufficiency’ as opposed to data ‘saturation’ (Dey, 1999), where sample size is based upon how well the data are able to create an understanding sufficient enough to answer the research question.

Vasileiou and colleagues (2018) encourage researchers to consider their study sample size in relation to the context of their study and the methods used, as opposed to applying numerical guidelines. Due to the reported high risk of attrition using Photovoice (Topcu, 2015; Baker & Wang, 2006) and with unpredictable periods of illness and symptom relapses in MS, we felt it appropriate to aim to recruit up to 16 participants to allow for study drop-out, however we treated this sample size flexibly. Data sufficiency was monitored during analysis with the intention to cease recruitment earlier if the data was adequate, and to extend recruitment if it was not. We confirmed data sufficiency when we considered the analysis process to have produced themes with considerable coherence and when relationships between themes could be delineated.

2.4.2 Recruitment

We used multiple avenues for recruitment to maximise opportunities for sample diversity. Participants were recruited through an NHS Trust Department of Neurology MS outpatient clinic, three local branches of the MS Society UK, and a local MS PPIE group. Williams, Sheffield and Knibb (2014) recommend face-to-face contact with participants in studies using a photovoice method to reduce attrition rates, which informed our decision to recruit from local services and groups. The clinicians in the MS outpatient service also told their patients opportunistically about the study during their routine clinic appointments, and a study advertisement[44] was displayed in the service's waiting room. The MS society local branches printed the study advertisement in their newsletters and shared the advertisement on their respective 'Facebook' groups. The study advertisement was also sent to the PPIE group members and the study was presented at one of their meetings. Those recruited from the MS Society and PPIE groups contacted the first author (LSP) to express interest using the information provided on the advertisement. For those recruited via the MS clinic, there was an additional option for the potential participant's clinician to pass their details to the research team with written authorisation from the potential participant[45]. They were then contacted by the first author (LSP) by telephone.

During the initial telephone conversation between LSP and the potential participant, eligibility to take part was assessed according to the inclusion/exclusion criteria (outlined in the following sections)[46]. If the inclusion criteria were met and the person deemed eligible to take part, demographic information was collected over the phone, and a pre-paid returnable consent form[47] was sent to the potential participant along with an information pack[48]. Potential participants were asked to return the consent forms to the researcher within two weeks of receipt if they wished to take part. When informed consent

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[44] Please see appendix E for a copy of the study advertisement
[45] Please see appendix F for authorisation form to share potential participants' contact details
[46] Please see appendix G for eligibility screening tool
[47] Please see appendix H for a copy of the participant consent form
[48] Please see appendix I for a copy of the participant information pack

was received and participants were given ample opportunity to discuss the study and ask questions, study participation commenced.

2.4.2.1 Inclusion criteria

Our inclusion criteria were intentionally broad to allow different types of people with different experiences to participate.

Participants were required to:

- Have a diagnosis of MS and self-report experiencing/ having experienced one or more invisible symptoms of MS.
- Have the capacity and be willing to give informed consent.
- Be aged 18 or over.
- Be able to communicate in verbal and written English (translator services were not used due to the large amount of verbal and written exchange between participant and researcher, and limited resources to fund this. We also felt that using an interpreter would likely lead to the loss of nuanced information).
- Have access to a smart phone, electronic tablet (with camera function) or digital camera and be physically able to use one of these devices to take an image. Approximately 76% of adults owned a smartphone in 2017 (Ofcom, 2017), and in 2012 78% owned a digital camera (Bridger, 2013). It was therefore expected that this study would not exclude a large number of people from participating on the basis of this requirement.
- Have the physical ability to use and access to email, multimedia messaging services (MMS; on mobile phone or tablet), or ‘WhatsApp’ (mobile application software).

2.4.2.2 Exclusion criteria

Individuals who were not able to independently use either a smart phone, electronic tablets or digital camera to take images were not be eligible to participate in the study. This is because an important part of the study was for the individual to capture their own experiences. Training could be provided by the first author for those who were physically able but are not confident with using the technology.

2.5 Study Procedure

2.5.1 General Process

Figure 16 outlines the study process for each participant, however here I describe the procedure in more detail.

2.5.1.1 Orientation Meeting

Meeting in person took place at the participant's home, at the University of Nottingham, or via video link (i.e. Skype) if the former options were not possible for the participant. The meetings were arranged to last for up to one hour. The first author (LSP) discussed the following with the participant:

- The overall study procedure, including procedures for sending images to the researcher. Participants were encouraged to caption images with a short title if they wished to or offer a brief written narrative to aid recollection in later interviews.
- If the participant was not confident in using their chosen device for taking images, training was provided at this point by the researcher.
- Examples of images that might be taken in a photovoice study. Whilst there may have been a risk of priming or influencing the images taken, it was important that the participant was clear about the methodology and expectations. The example images and corresponding narratives captured LSP's experience of working towards a doctorate in clinical psychology[49]. It was made explicit that the exemplars offer a variety of ways to capture experiences, but that the research was interested in participant's own subjective experiences of the invisible symptoms of MS.

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[49] See appendix J for example images shown to participants

- Ethical considerations for taking photographs/digital images[50].
- Copyright regarding images taken by participants. Participants were informed that they would have legal ownership of the image and the image would only be used for the purpose of the research and dissemination. Participants were informed that they could decide on an individual basis what acknowledgment (if any) they would like for their photos, but that in using their full or first name anonymity would be compromised. Alternatively, they had the option to use a pseudonym.
- Safeguarding issues. The participants were made aware of the potential limits of confidentiality should the content of their images or interviews indicate that they themselves or someone else are at risk of harm. In such a situation LSP would first seek support and advice from the Chief Investigator (RdN) and academic supervisors.

Although much of the above was included in the information sheets provided to participants at the stage of consent, it was verbally communicated to offer further clarity for the participant. At the end of the meeting the participant was asked to spend the next two weeks (approximately) taking a minimum of five images that capture their experience of living with and managing invisible symptoms of MS, using their personal phones or digital cameras. Participants were told that it did not matter if they took more images related to living with symptoms than managing them, or vice versa, and they could choose to take pictures that captured both aspects of their experience. We asked participants to send their images to the researcher via email, MMS message, or ‘WhatsApp’ (an encrypted mobile application software) message (the latter was recommended, and participants were made aware of potential data protection issues with the first two options).

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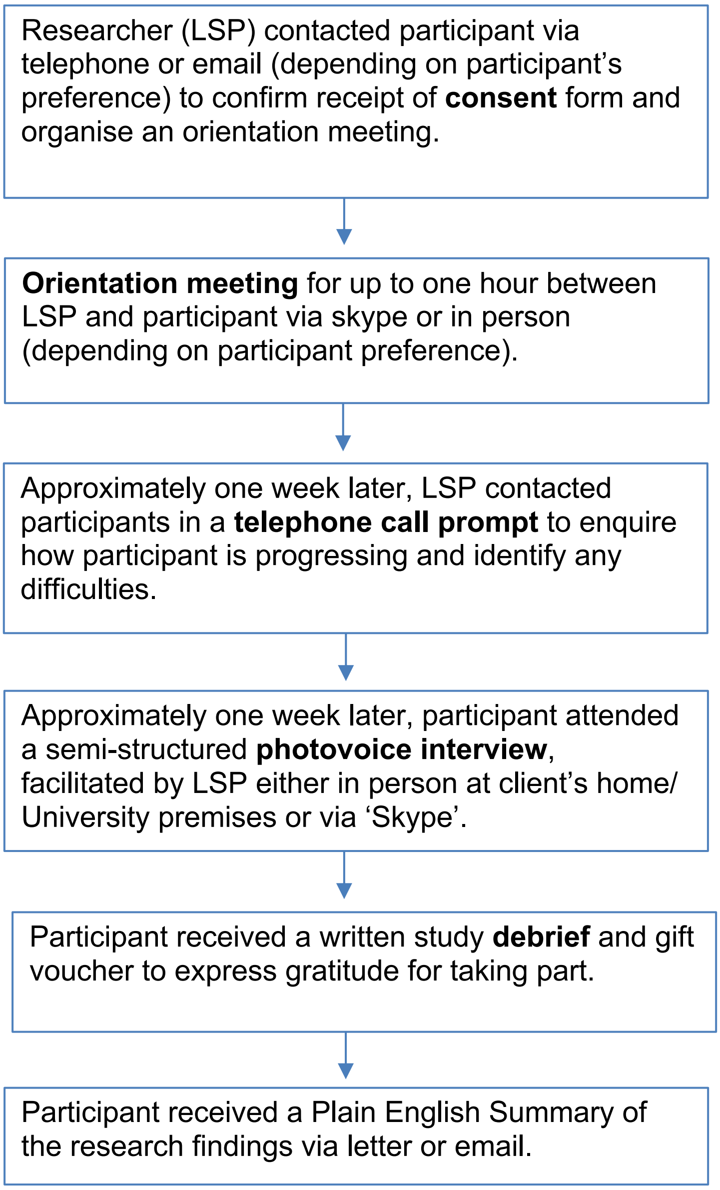
[50] Please see appendix K for ethical considerations sheet shared with clients

Figure 16

Study process for each participant.



Researcher (LSP) contacted participant via telephone or email (depending on participant's preference) to confirm receipt of **consent** form and organise an orientation meeting.

Orientation meeting for up to one hour between LSP and participant via skype or in person (depending on participant preference).

Approximately one week later, LSP contacted participants in a **telephone call prompt** to enquire how participant is progressing and identify any difficulties.

Approximately one week later, participant attended a semi-structured **photovoice interview**, facilitated by LSP either in person at client's home/ University premises or via 'Skype'.

Participant received a written study **debrief** and gift voucher to express gratitude for taking part.

Participant received a Plain English Summary of the research findings via letter or email.

LSP had a phone for the purpose of the research. We asked participants to send us their pictures as soon as they took them if possible. We asked those with 'free data' to use their mobile phone to send images, so as to not add to their costs. For those with no free data, we requested they send the images by email or MMS messages. Participants were reimbursed the cost of up to 12 photo MMS messages if they had used MMS messages and did not have free data.

2.5.1.2 Telephone Call

The first author (LSP) contacted participants via telephone and asked about their experience of participating in the study so far. This gave opportunity to identify any barriers to participation and check-in on participant wellbeing. At the end of the call, LSP arranged an interview date and time with the participant, approximately one week in the future (as was convenient for the participant), and the participant was asked to continue taking and sending images for the duration leading up to this.

2.5.1.3 Photovoice Interview

The participant was sent a message prompt via their chosen method of communication with the researcher (email, text or 'Whatsapp') prior to the interview, requesting that they select the five images that most accurately capture their experience of invisible symptoms of MS, and to inform LSP a day before the interview. These images were presented at the interview by LSP and were used to elicit discussion about each image and their meaning for the individual. We felt that asking for five images as a minimum reduced participant burden and also would allow interview duration to be sufficiently long enough to elicit rich data, but that this would not be a long and burdensome process for the participants.

2.5.2 Data Collection

2.5.2.1 Demographic Data

We collected demographic information on age, ethnicity, gender and preferred pronouns, MS sub-type, and years since MS diagnosis. This allowed us to determine the diversity of our sample and to adequately consider our findings in line with our participants' contexts (as advised by Brain & Clarke, 2013).

2.5.2.2 Semi-Structured Interviews

Interviews are a widely used method of data collection in qualitative research, given the focus on the exploration of human experience and phenomena (Côté & Turgeon, 2005; Rubin & Rubin, 2011). Researchers have emphasised the importance of regarding interview method as data collection tools to be given great consideration when answering a research question, and not as informal discussions with participants (Brinkmann & Kvale, 2005). Structured interviews usually involve the researcher asking all participants a set of predetermined closed questions in the exact same manner, with no deviation from the interview schedule (McGrath et al., 2019). Semi-structured interviews, however, treat the predetermined questions as a guide for discussion, and the researcher can probe the participant for more detail through follow-up questions and prompts (Lingard and Kennedy, 2010), allowing for greater flexibility in the exploration of the topics, whilst retaining a sense of structure and focus on the research question that can be lost when the process is unstructured and produces large quantities of data (Alsaawi, 2014). Structured interviews tend to be used to generate quantitative data (Whiting, 2008), and as we wanted to elicit rich data and allow opportunity for co-construction of meaning between participant and researcher, we felt that a semi-structured approach would enable our research aims to be met.

Although focus groups have been used in previous photovoice research (Cooper & Yarborough, 2010, Peña-Purcell et al., 2018), given the personal and often emotive nature (Parker et al., 2020) of some of the common invisible symptoms of MS (e.g. bladder/bowel/sexual dysfunction), we felt that this method may have hampered discussion of these issues due to participant discomfort. We opted instead for one-to-one interviews between the participant and one researcher (LSP).

The semi-structured interview was also consistent with our epistemological position of critical realism in the study. Within the questions, invisible symptoms are acknowledged as real experiences, but it is maintained that the experience of these symptoms is constructed in part by the participants' individual beliefs and perceptions (Maxwell, 2011). We also regarded the interviewer as active in the co-creation of data and meaning with the participant (Lingard & Kennedy, 2010).

While face-to-face interviews are the recommended method for gathering data in photovoice research and are considered ‘gold standard’ in qualitative research more generally (Novick, 2008; Williams et al., 2014), we also provided the option for participants to be interviewed via video-conferencing software (‘Skype’). This was so that people who had difficulty meeting in person at the University (e.g. due to mobility problems) and did not wish to be seen in their home could still participate, helping to maximise diversity within the sample. ‘Skype’ has been found to be a cost-effective and viable alternative to face-to-face interviewing (Cater, 2011). The Covid-19 global pandemic began during the time we were collecting data, While prior to this, all participants opted for face-to face interviews, after the pandemic began we were no longer able to interview participants face-to-face (in accordance with governmental guidance and participant safety), and all subsequent interviews were conducted via ‘Skype’.

2.5.2.3 Interview Schedule and Process

McGrath & colleagues (2019) highlight the importance of planning for participant interviews carefully, with great consideration to the research question, and they developed guidance which we used to inform some of our decision-making when developing and facilitating the interviews. We designed the questions for the semi-structured interviews to be open-ended, inductive, non-leading and free of jargon (McGrath et al., 2019). We developed them to be relevant to our study aims and to elicit discussion specifically about the images, inviting participants to explore what the image relates to or captures with regards to their experience of living with and/or managing the invisible symptoms of MS[51]. The first question asked the participant what symptoms of MS they either experience or have experienced that they consider to be invisible. This was not only to orientate the participant to the topic and help them feel comfortable with a question they could easily answer (McGrath et al., 2019), but also allowed us to identify which symptoms mentioned in the interview were conceptualised as being invisible by

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[51] Please see appendix L for a copy of the interview schedule used in the semi-structured interviews

the participant. Interview style and building rapport with the participant is a crucial part of a successful interview process, however this can be difficult due to time limitations (DiCicco-Bloom & Crabtree, 2006; McGrath et al., 2019). The interviewer (first author LSP) built rapport early in the experiment through the orientation meeting, and then further in the interview through demonstrating active listening, curiosity and openness (Bell, 2014; Giger, 2017; Seidman, 2013).

The images taken by participants were not used as a primary data source, and were used to enhance discussion and exploration during the interviews of living with and managing invisible symptoms of MS. The interviewer followed guidance in the literature around setting up interviews, including explaining the purpose and format of the interview (Rose, 1994). During the interview, participants could take as much time as needed to answer questions and to speak about the images they had taken and could say as little or as much as they wished. The interviewer also had the freedom to explore other topics that were mentioned by the participant in the interview and ask the participant to expand on meaning (e.g. ‘could you tell me more about X?’). If the interview was in person, the images were displayed on the researcher’s laptop screen, and if over Skype, the participant ensured that they could view the images on their own device for the interview. The participants’ images were discussed one at a time, and the participant decided which order they would be discussed in. When the participant had finished speaking about their image, the researcher offered their reflections on the image, which at times generated further reflections and responses from the participant. The researcher could also point to parts of the image they were interested in, asking the participant about that particular feature. Five images were decided in advance to discuss as a guide (except with one participant who took four images), however once these had been discussed, all participants were given the option to talk about any additional images they had taken if they wished to. The researcher could also turn the discussion to additional images after others had been discussed.

Breaks were negotiated prior to the interview; however, participants were also free to indicate during interview if they required additional breaks. Participants were informed that they can stop the interview should they become distressed. In such instances the interviewer planned to use their clinical skills as a trainee Clinical Psychologist to support the participant. Participants were also made aware that they could decline to answer questions.

After the interview, the participants indicated via a publication consent form[52] which of their images could be used for various dissemination of the research. Time was allocated for debriefing after the interview[53]. The participants received a gift voucher with ‘Amazon’ for the value of £10 as an expression of gratitude towards their valuable contributions (Anderson, 1991)[54].

2.5.2.4 Recording equipment

All 12 interviews were recorded on an Olympus DM-720 digital audio recorder and stored onto an encrypted memory stick, backed up onto a University-approved secure server.

2.5.2.5 Transcription

Although it is acknowledged that the transcription process inherently will transform the data and cannot replicate the audio data exactly (Willig, 2013), verbatim transcription is widely recognised as a method to increase accuracy and rigour in research (Loubere, 2017). The first author chose to transcribe all 12 interviews verbatim. This allowed them to become immersed in the data, begin the analytical process early, and practice reflexivity throughout the process (Potter, 2003). Listening to the interview audio recording before transcribing improves accuracy in capturing the content (Whiting, 2008). The first author listened to each recording from beginning to end twice before listening again during the transcription process. After transcription was complete, they listened a final time in full whilst reading the transcript to ensure accuracy.

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[52] Please see appendix M for a copy of the image publication consent form.

[53] Please see appendix N for participant debrief form

[54] Please see appendix O for copy of form given to participants to confirm receipt of gift voucher

2.6 Ethical Considerations & Approvals

Research must adhere to ethical standards in order to uphold the rights of participant and improve study credibility (Resnik, 2020). The study was granted ethical approval by the East Midlands-Derby Research Ethics committee and the Research & Innovation Departments of the participating NHS trust, which enabled us to recruit participants from the NHS Neurology outpatient clinic. The study was also granted ethical approval by the Division of Psychiatry & Applied Psychology at the University of Nottingham, to allow us to recruit via MS Society newsletters and relevant 'Facebook' groups[55]. We also had adequate insurance and indemnity to cover liabilities which could have arisen in relation to the study.

The study was conducted in accordance with the ethical principles outlined in the UK Policy Framework for Health and Social Care Research (The Health Research Authority, 2017), and The British Psychological Society's Code of Research Ethics (2018). The ethical implications of the study were considered throughout the research process, and key acknowledgments are highlighted below.

2.6.1 Informed Consent

At the recruitment stage, potential participants who were informed of the study by their regular clinician at their Neurology outpatient appointments and wished to be contacted by the researcher, completed a consent form for the clinician to share their contact details with the researcher[56].

All participants provided written informed consent to take part in the study. After confirming eligibility, participants were sent a Participant Information Sheet about the study procedure[57]. They were also sent a consent form with a pre-paid envelope to return this to the researcher[58]. The researcher's contact details were provided to give potential participants the opportunity to ask questions before

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[55] Please see appendix P for all ethical permissions paperwork
[56] Please see appendix F for the consent to contact form
[57] Please see appendix I for Participant Information Sheet
[58] Please see appendix H for participant consent form

giving consent and were given ample time of two weeks to consider their participation and return the consent form if they wished.

Participants were also asked for verbal consent prior to audio recording their semi-structured interviews.

2.6.2 Confidentiality and Data Protection

It was important that we protected the confidentiality of the study participants, but also that participants retained the right to choose whether their real names, quotes and digital images were shared in the write-up, dissemination and publication of the research (as rigidly adhering to anonymity can silence people's individual voices; Braun & Clarke, 2013).

For the purpose of data storage, the study was conducted in accordance with the Data Protection Act (2018) and the approving Research Ethics Committees. Access to the data was limited to the researcher and research supervisors. The administrative staff for DClinPsy controlled access to the locked filing cabinets in which the hard-copy data (e.g. consent forms, expenses confirmations) were stored.

Given that participants were not given the option until after their semi-structured interviews to choose their own pseudonym or to consent to their real name being used, they were assigned a code for the purpose of data storage. Codes were assigned numerically (e.g 'P1', 'P2', 'P3' etc.) according to the order that participants were recruited in. All electronic data was stored according to the relevant participant code throughout to ensure confidentiality, and individual documents were all password protected. All interview recordings, transcripts and digital images produced by participants were stored on a password protected, encrypted data stick and backed up on a secure University of Nottingham server. All data will be stored for seven years in accordance with University policy, following which it will be destroyed. This information was made clear to participants on the information sheet and consent form.

Each participant was informed of the limits of confidentiality, and that this may be compromised if issues pertinent to safeguarding were raised, either through the content of the interviews or the images they take.

2.6.3 Withdrawal

Participants were made aware that they could withdraw from the study at any time without giving a reason. Their participation was entirely voluntary, and participants were aware that they would incur no penalties for withdrawing. Participants were made aware (via the information sheet and consent form) that should they withdraw, the data collected up to that point might not be able to be erased and may still be used in the final analysis. Withdrawn participants were not replaced as the over-recruitment strategy allowed for study attrition.

2.6.4 Participant Safety and Wellbeing

As we asked participants to produce visual material relating to potentially difficult emotional experiences, their wellbeing and distress levels related to the photovoice process were monitored and reviewed at the orientation meeting, telephone check-in, and semi-structured interview. Participants were also given the number for the researcher's work phone and email address so that they could make contact for support if they experienced distress as a result of their participation. No participants contacted the researcher to express distress during the study process, and no notable issues arose at the telephone check in. One participant became tearful in their semi-structured interview in response to a topic they had initiated (no distress occurred in response to a direct interview question from the researcher), and LSP used their clinical skills as a Trainee Clinical Psychologist to support them. They chose to continue with the interview and the topic of conversation was changed. The participant reported no negative effects at the point of study debrief. All participants were offered a debrief [59] at the end of their semi-structured interview. Some literature indicates that it is not uncommon for participants to report the benefits of producing these types of images for research (Guillemin & Drew, 2010). Participants in Oliffe & Bottorff's

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[59] Please see appendix N for debrief document given to participants

(2007) study of health and illness reported finding the act of talking about their photos to be therapeutic. The comments from participants who provided feedback about the process were positive, and no adverse events were reported.

2.6.5 Ethical Considerations in Photovoice Research

The photovoice methodology introduced specific ethical issues which were addressed fully in the study and approved by the aforementioned committees. One consideration relates to ownership of any digital images used in the study. As the creators of the image, participants are deemed to be the legal owners (British Sociological Association, 2017). Therefore, their consent must be obtained for their digital images to be used in the study itself as a basis for interview, and for any subsequent dissemination or publication. This information was made explicit on the consent form. Due to the images being used exclusively for research purposes, participants were not asked to enter into any formal copyright agreements. Participants were given the opportunity at the end of their interviews to specify any images that they did not wish to be used as outputs of this study and completed a consent form to specify the use of their images in dissemination and publication[60].

It is important in visual research that the images used do not breach ethical guidelines or the rights of others. All participants were educated about the legal implications, etiquette and exercises of ethical decision-making when taking their images and were asked to implement these practises throughout participation. We developed a list of ethical considerations based on previous studies that have used visual methods (Reavey & Johnson, 2008; Wiles et al., 2008;) and best practice guidelines for taking photos in public (Techradar, 2012; Amateur Photographer, 2016) to share with participants[61].

Participatory studies such as photovoice create insights into personal contexts not usually open to public scrutiny. Unless explicitly stated by the participant in a documented format that they wish to compromise their anonymity in this way, any quotes or images from which the participant could be identified were not included

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[60] See appendix M for image publication consent form

[61] See appendix K for ethical guidance given to participants

in the study write-up. Images involving third parties on private property were not included in the write-up as the third parties could not give consent for this. On some images, minor identifiable information has been blurred or removed to preserve the image and its meaning for the participant (participants were made aware in these situations what these changes would be and agreed to them). Some participants chose to waiver their anonymity and be credited for their images and quotes by their real name during the publication and dissemination of the research, and additional written consent was obtained for this. Pseudonyms chosen by the participant were used in all other instances.

2.7 Analysis

2.7.1 Rationale for Thematic Analysis

To gain access to participant's subjective experiences, we used inductive-deductive Thematic Analysis (TA). As a theoretically flexible method that can be applied across a variety of approaches and epistemologies, it could be used within a critical realist framework and in conjunction with visual methods to meet the research objectives (Braun & Clarke, 2013; Willig, 2013). The TA process allows patterns (also known as themes) in the data to be identified, described and understood in relation to the phenomenon under investigation, and the researcher assumes an active role in the development of the themes (Braun & Clarke, 2006). Identifying themes across the data allows for rich accounts of a phenomenon to be understood and explored in depth, and thus was fitting for our research aims (Braun & Clarke, 2013). High quality TA also has strengths in terms of the transparency of the analysis process (Joffe, 2012).

We considered using Interpretative Phenomenological Analysis (IPA) to help us to explore people's experiences of living with and managing invisible symptoms of MS. IPA is a methodology which provides in-depth guidance throughout the research process and is concerned with understanding the inner worlds of participants (Braun & Clarke, 2013). Whilst this approach may have enabled us to address the research question to an extent, we felt that IPA's double hermeneutic stance would take us further away from the co-construction of meaning that we wished to have with our participants (Biggerstaff & Thompson, 2008). In IPA, the researcher does not draw on prior assumptions and theory, and analysis is a purely inductive process (Reid et al., 2005). Given that there has been previous literature around invisibility in MS and its impacts, it was important for us to use an analytic approach that allowed us to adopt an inductive-deductive lens based on participants' accounts and previous research. We also chose TA as an approach because it is consistent with the first author's epistemological stance and maps onto the assumptions the study makes about the information that we can access through photovoice.

2.7.2 Analysis Process

2.7.2.1 Inductive vs. deductive analysis

Themes can be developed in two main ways in TA: through an inductive or 'bottom-up' process, or in a deductive or 'top-down' way (Braun & Clarke, 2006). Inductive analysis is data-driven in that the data is coded and themes are developed from the data itself without drawing on any pre-existing theories or preconceptions (Patton, 1990). Deductive analysis, on the other hand, is theory-driven in that the coding is led by existing theory and how this applies to the data, thus providing a detailed analysis of a particular aspect of the data in order to answer a research question (Braun & Clarke, 2006).

Whilst it was important for us to gain new insights into the experience of living with and managing a cluster of invisible symptoms for people with MS, we also wished to recognise the previous research and meta-synthesis into this area (e.g. Parker et al., 2020) which may be relevant. A hybrid approach of inductive-deductive coding is permitted in TA (Fereday & Muir-Cochrane, 2006), and fits with the critical realist position we adopted.

We therefore decided *a priori* to employ a deductive-inductive TA, whereby the data was initially coded using an inductive method, and then a deductive sweep of the data was performed. With the deductive sweep, rather than coding anything relevant to the research question, the data is coded selectively and is systematically searched for ways in which it fits with existing research or ideas.

In this case, the existing research (i.e. Parker et al's 2020 meta-synthesis) was developed into a deductive coding framework which was used to analyse the data. During the deductive sweep, the first author asked a series of questions of the data in line with the deductive framework and coding where there were consistencies, also noting when the data did not fit with the frame[62]. The results of the deductive sweep will be further commented on in the next chapter ('Extended Results').

2.7.2.2 The approach used

The TA process has previously been criticised for an absence of clear guidance for how to conduct this (Antaki et al., 2002). In response, Braun and Clarke (2006) developed and outlined six stages of TA that captured the process but also maintain some flexibility within each phase. We decided *a priori* to follow Braun and Clarke's (2006; 2013) six phase method of TA. Although we present this below as a linear process, we maintain that the analysis was an iterative and reflexive process. To ensure the quality of the TA, we referred to Braun and Clarke's (2006) '15-point checklist for a good TA', which covers transcription, analysis and write-up of the findings. A reflexive stream of consciousness was maintained by the researcher throughout the analysis process.

2.7.2.3 Phase 1- Familiarisation with the data

We have already detailed the ways in which the transcription process helped the first author to become immersed in the data in the early stages of analysis by re-listening to the audio recordings of interviews and re-reading transcripts. It is also deemed useful to make initial notes on early impressions of the data (in a 'stream of consciousness' fashion), which the first author did (Maguire & Delahunt, 2017).

Braun & Clarke (2006; 2012) highlight the importance of reading the data actively, critically and analytically, thinking about what the data might mean. The first author asked questions such as 'how is the participant making sense of their experiences?', 'what assumptions do they make?' and 'what lens are they using

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[62] Please see appendix A for deductive coding framework

the make sense of their world?', and made brief written notes whilst listening to the audio recordings, and then reading the transcripts.

2.7.2.4 Phase 2- Generating initial codes

At this stage, the data starts to be organised in meaningful and systematic ways through reducing the data into small chunks of meaning and labelling them (i.e. coding). Codes identify a feature of the data and represent the most basic unit of the raw data that can be assessed in a meaningful way (Boyatzis, 1998). When coding, the researcher works through the dataset in a systematic way, identifying aspects of the data which are interesting and may indicate repeated patterns across the dataset (Braun & Clarke, 2006).

The transcripts were transferred to a template table to begin the coding process[63]. When generating codes through inductive method, the first author coded the data line by line, identifying anything that may be relevant to the research question and writing a short phrase (a code) that summarised the point of interest (Braun & Clarke, 2013). After a whole transcript was coded inductively, the deductive sweep was conducted. When a code was identified, it was given a 'marker' so that it could be linked easily to the text[64]. Data could be given more than one code. Inductive and deductive codes were given different font colours so that the first author could identify how the code had been developed. Similar codes were collated together into separate files. Data were analysed at a semantic-latent level, which involves coding what participants have explicitly said (semantic) and interpreting what is said to develop meaning beyond what is explicit in the data (latent) (Braun & Clarke, 2006; Clarke et al., 2015). This was deemed appropriate so that the individual voices of the participants were reflected in the codes, as well as the researcher's interpretations.

The first author's coding technique was checked by two of the other authors, and the codes generated were also second-checked by two other authors to ensure that codes were coherent and had been subjected to a range of perspectives and interpretations. Coding was discussed in supervision and new codes were

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[63] Please see appendix Q for table used for coding

[64] Please see appendix R for an anonymised example of coding the data

sometimes generated and existing codes modified as appropriate as a result of these discussions.

2.7.2.5 Phase 3- Searching for themes

When all of the data have been coded and collated, this phase begins whereby the researcher/ analyst sorts the different codes into potential themes by grouping together codes which have similarity and overlap and can be combined to create a coherent and overarching meaning. Themes are proposed to capture something important about the data which helps to answer the research question, and to represent a pattern or meaning in the data (Braun & Clarke, 2006). Subthemes can be created which are subcomponents of a theme. Whilst there are no rigid rules about what constitutes a theme, Braun and Clarke (2006; 2012) highlight that a theme should be significant in terms of answering the research question, that themes should be distinctive from one another but also work together in telling an overall 'story' about the data.

In this study, the first author organised the codes into broader features that represented patterns of meaning across the data. For example, the codes 'others dismiss my symptoms' and 'people assume my symptoms aren't there' were grouped under the title 'invalidating response from others'. To do this, the first author printed out each code, colour coded them for each participant, and organised them into piles according to similarities, overlaps and salient features. They then gave these piles of codes a 'title' and a descriptive summary [65]. These clusters of codes were then further grouped conceptually into areas of overlap, eventually developing themes.

By the end of this step, the codes had been organised into broader related 'candidate' themes which told us something about the ways people experience and live with invisible symptoms of MS.

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[65] Please see appendix S for a photo of the code grouping process

2.7.2.6 Phase 4- Reviewing themes

In this phase, the analyst begins the recursive process of reviewing and defining the candidate themes in relation to the coded data and entire dataset (Braun & Clarke, 2012). The first step involves checking themes against the relevant data extracts to assess whether the them works in relation to the data (Braun & Clarke, 2006; 2012). The first author read the data associated with each theme to ensure that the theme was supported by what was in the data- the key position held in mind throughout was the notion that ‘any interpretation made must be grounded somewhere in the data’. The second step involves checking whether the themes reflect the meaning held within the entire dataset (i.e. within and across all interviews).

The first author created visual representations of the candidate themes as thematic maps, in line with Braun and Clarke’s (2006) suggestions. This allowed the research team as a whole to view and discuss the potential themes and how they related to one another and to the research question. The process involved scrutinising the themes and coming to agreements in relation to plausibility and the extent to which the themes were representative of the data and answered the research question.

Key questions asked during this phase are highlighted below, and were adapted from Braun and Clarke (2006), and Maguire and Delahunt (2017):

- Do the themes make sense? What does it include and exclude?
- Do the themes tell us something useful about the data?
- Does the data support the themes? Are there enough meaningful data to support them?
- Am I trying to fit too much into a theme?
- Are the themes distinct? If any themes overlap, are they really separate themes?
- Are there themes within themes (subthemes)?
- Are there other themes within the data and the research question?

Research supervisors agreed with the final themes constructed and felt that they captured the most important and relevant elements of the data in relation to the research question.

2.7.2.7 Phase 5- Defining and naming themes

At this stage, the purpose is to identify the 'essence' of each theme independently and in relation to one another, in terms of what aspects of the data the themes capture (Braun & Clarke, 2006). The themes are analysed in terms of the 'story' they tell individually, but also to check that they fit with the broader overall 'story' being told in relation to the research focus. The analyst also must identify whether a theme contains any sub-themes, as they can be useful to give structure to complex themes (Braun & Clarke, 2006).

Braun and Clarke (2012) suggest that a way to test that refinement at this stage has been achieved is to task the analyst with summing up each theme in a few sentences. This was tested in research supervision verbally and then in written form. Each theme and sub-theme were given a definition to ensure clarity. Through discussions as a research team, we felt that developing sub-themes within the themes helped to 'tell the story' of each theme and the collection of themes overall more clearly.

2.7.2.8 Phase 6- Producing the report

In this final stage, the analysis is 'written-up' in a way that provides the reader with a compelling and coherent story about the data, and clearly meets the research aims (Braun & Clarke, 2012). The analysis process continued throughout write-up, in that the first author made decisions about what data (or participant quotes) were included to demonstrate each theme/sub-theme and add richness to their descriptions. All authors provided input into the first author's write-up of the analysis by providing feedback and commentary.

2.8 Ensuring the Quality of the Research

2.8.1 Quality in qualitative research

Evaluating the quality of research involves judging how well the study was designed and conducted, whether research aims were met and how useful and relevant the findings were (Yardley, 2008). In contrast to quantitative research, the standards against which to evaluate the quality of qualitative research are more difficult to define, and there is not a widely agreed method to assess this (Yardley, 2000). It has been recommended that research is evaluated by assessing the validity and reliability of the study (Silverman, 2006). However, Yardley (2000) proposes that because of the multiple and varying approaches, methodologies and epistemologies that can steer qualitative research, it is not appropriate to develop a stringent and universal set of standards against which to measure qualitative study quality. She suggests instead that research quality should be evaluated in flexible ways, beyond the scope of what has traditionally been considered 'valid' and 'reliable' (particularly as the notion of validity lends itself to the idea that there is an absolute 'truth' to be found which is largely inconsistent with qualitative ideas; Yardley, 2000).

It is proposed that there are five key criteria that can be used to appraise the trustworthiness and quality of qualitative research: credibility, dependability, confirmability, transferability and reflexivity (the latter of which will be discussed in a later section; Korstjens & Moser, 2018; Lincoln & Guba, 1985; Stenfors et al., 2020).

2.8.1.1 Credibility

The concept of credibility refers to the research having good plausibility and being trustworthy (Guba, 1981). There should be a cohesiveness between the existing literature on the topic, the formulation of the research question, the study design and methodology, data analysis and resulting discussion, and these should be justified at each stage of the study (Stenfors et al., 2020). Peer scrutiny, reflective commentary and assessment of data sufficiency are commonly used to indicate credibility (Shenton, 2004, Stenfors et al., 2020).

This project came under peer scrutiny regularly in the form of presentation at academic panels on the DClinPsy course and PPIE meeting, research supervision and academic submission of the research proposal. Feedback gained from this scrutiny was incorporated into the design of the study to further improve credibility. Decisions made throughout the process were tracked back in supervision in terms of coherency. A reflective diary was also kept by the first author to acknowledge and monitored the ways in which subjective responses could influence the research process (this is addressed further in a later section). Credibility was also strengthened and triangulated by having members of the research team (including the PPIE member) involved in the analysis process and second checking of both coding and themes (Korstjens & Moser, 2018).

2.8.1.2 Dependability

Dependability refers to the extent to which a researcher could replicate the study and requires that enough information is provided in the write-up about the methods and procedures used (Shenton, 2004; Stenfors et al., 2020). We have provided sufficient detail in our extended paper for the study process to be replicated (albeit the findings may well be different).

2.8.1.3 Confirmability

For a study to demonstrate confirmability, a clear link must be evidenced between the data and the findings and the researcher should include detailed descriptions and data (e.g. quotes) to do so. Our results include rich and contextualized quotes from the participants, including the ways in which they link to the theme or finding being presented.

2.8.1.4 Transferability

This relates to the degree with which the study results can be transferred to other settings, contexts or groups (Korstjens & Moser, 2018; Stenfors et al., 2020). In qualitative research, it is not the task of the researcher to determine transferability, but for those who wish the ‘transfer’ the results to make that judgment (Lincoln & Guba, 1985). The researcher, however, can facilitate this assessment by providing detail of the research context and central assumptions of the research (referred to as providing a ‘thick description’; Lincoln & Guba, 1985). The descriptions have been captured in our findings, where we have taken time to understand participants’ contexts.

2.8.1.5 Evaluating Quality of the Research

Several checklists are available to facilitate the processes of evaluating the quality of qualitative research. Stenfors and colleagues (2020) propose that making use of these checklists can be beneficial as they contain helpful markers that indicate quality, however that such checklists should be applied whilst taking into consideration the wider context of the research and employing critical thought.

The Critical Appraisal Skills Programme (CASP, 2018) encompasses 10 quality criteria against which research can be assessed. We decided to use this as a tool to evaluate our study, given that it can be applied generally across a range of methodologies and is unspecific in this regard. We asked ourselves of the research, the following questions taken directly from the CASP (2018) in order to assess quality:

- Was there a clear statement of the aims of the research?
- Is a qualitative methodology appropriate?
- Was the research design appropriate to address the aims of the research?
- Was the recruitment strategy appropriate to the aims of the research?
- Was the data collected in a way that addressed the research issue?
- Has the relationship between researcher and participant been adequately considered?
- Have ethical issues been taken into consideration?
- Was the data analysis sufficiently rigorous?
- Is there a clear statement of findings?
- How valuable is the research?

These criteria are addressed in the content of this extended paper and the journal paper. Yardley (2000) developed guidance around ensuring quality in health psychology research. These principles were intended to be interpreted as open-ended and flexible rather than rigid rules, so that researchers can think critically about how the criteria can be met considering their study's methodology, methods and epistemological stance. The flexible principles are 'sensitivity to context', 'commitment and rigour', 'transparency and coherence', and 'impact and importance'. In addition to the CASP (2018) we used Yardley's evaluative principles as this encouraged critical thought in the way that these apply to our study. We asked of the research the following questions, adapted from Yardley's (2000) guiding principles:

Sensitivity to context

- Does the researcher have an awareness of the relevant literature and previous related empirical work?
- Is there a grounding in the philosophy and epistemological position of the approach that has been adopted?
- Does the researcher explore and account for findings that are not consistent with existing literature or their expectations?
- Does the researcher acknowledge the sociocultural setting of the study?
- Does the research acknowledge the social context of the relationship between researcher and participant?

Commitment and rigour

- Is prolonged engagement with the research topic demonstrated?
- Did the researcher demonstrate competency and skill in the methods used?
- Did the researcher become immersed in the theoretical/empirical data?
- Did the study demonstrate sufficient rigour with regards to data collection and analysis?

Transparency and coherence

- Were all aspects of data collection and analysis detailed?
- Did the researcher exercise reflexivity?

- Was there coherence between the research question, philosophical perspective adopted, methods and analysis?

Impact and importance

- Does the research enrich our understanding of the phenomena under investigation?
- Does the research have practical or socio-cultural implications?

2.8.2 Reflexivity

Reflexivity refers to an exploration and awareness of the ways in which the researcher influences the research at a personal and epistemological level (Willig, 2013). This process is considered essential in qualitative research for the researcher to maintain an awareness of the ways in which the research process can construct the object of the research (Bolam et al., 2003). It involves the researcher making sense of their own influence (through their assumptions, values, prejudice and experiences) over the research process and findings, and their relationship to the participants and the wider social context of the phenomena being studied (D'Cruz et al., 2007; Jootun et al., 2009; Koch & Harrington, 1998). Researchers are encouraged to attend to their ‘insider status’ (when a group identity is shared with the participant such as gender or age) and their ‘outsider status’ (when a group identity is not shared with the participant) throughout the study, identifying and understanding the ways this can influence each stage of the research (Gallais, 2008). Reflexivity in qualitative research is argued to improve rigour (Hand, 2003; Mills et al., 2010), however, there is little information or consensus about how reflexivity should look in practice and what methods should be used for this (Mauthner & Doucet, 2003). It is argued that due to the complex nature of reflexive processes, they are difficult to unfold and make explicit (Finlay, 2002).

A commitment to reflexivity is a recognised feature of research which adopts a position of critical realism (Price & Martin, 2018). In line with this position, the researchers (in particular the first author) acknowledged themselves as research instruments to represent the views of the participants, and therefore it was important to be mindful of and explicit about our relationship to the research topic, and the way in which our expectations, values, analytical styles and assumptions could impact the research (Elliot et al., 1999; Madill et al., 2000).

2.8.2.1 Consideration of the researcher role

It is recommended that researchers make a declaration about their stance so that this can be considered alongside the study (Mills et al., 2010). This has been acknowledged briefly in the journal paper, however more detail will be provided here. The first author is a 30-year-old White-British woman who is employed as a Trainee Clinical Psychologist. They do not have a diagnosed disability or chronic illness. Their previous experience clinically involved at times working therapeutically with people who have long-term chronic physical health problems and disability, some of whom described their symptoms as being invisible. Therefore, they approached the study with prior assumptions about the potential impacts of invisible symptoms more generally (for example, people not being believed by others and the debilitating impact on daily living). The first author had not worked clinically with a person who has MS and held no specific assumptions about people who suffer with this particular condition, however it would be naïve to assume that expectations from previous work would not influence their interpretations, and reflexivity was important in monitoring and responding to this. The first author had also spoken to people with MS (at the PPIE meetings) and gathered anecdotal evidence around the impact of invisible symptoms, and also had expectations in terms of the output from their meta-synthesis around invisibility and MS (Parker et al., 2020). The first author's main expectations were around the types of symptoms that would be deemed 'invisible' by participants, and that they would report negative impacts associated with their invisibility. They strove to attend to these assumptions throughout the process to minimise bias in the way the data was treated and interpreted.

The use of a research diary, where the researcher makes notes throughout the research process, can create a space in which to practice reflexivity (Lincoln & Guba, 1985; Nadin & Cassell, 2008). The first author kept a reflexive diary throughout the course of the research and noted down preconceptions and personal thoughts and reactions that may have influenced the study. Where deemed important, the reflections were discussed further in research supervision to discuss the potential impact on the study process.

2.8.2.2 Reflexivity during data collection

The importance of reflexivity throughout the data collection and interview process, and awareness of how the researcher can influence the discussions with the participant and hence the interview output has been highlighted (Hand, 2003; McGrath et al., 2019). Attending to researcher-participant interactions through keeping diaries, listening to audio recordings of the interview and reading the transcripts whilst noting reflexive observations is termed ‘interpersonal reflexivity’ (Walsh, 2003). It has been suggested that the use of a reflective diary can aid interpersonal reflexivity, as well as the noting down thoughts and feelings before, during and after the interview (Chesney, 2000; Clarke, 2006).

As the participants sent their images to the first author prior to interview, the first author noted down their initial thoughts, feelings and assumptions in relation to viewing it, and then returned to the photo one week later and did the same, exploring alternative perspectives. This was a way of attending to the researcher’s biases. Sometimes, this process provided a foundation for sharing these reflections with the participant at the end of the interview or asking about parts of the image that captured the researcher’s attention. However, these were always only shared at the end of the interview so as not to unduly influence or lead the participant’s responses. The first author also made notes before, during and after the interviews, and during the transcription process, which not only served as a reflexive tool to manage their perceptions, but to identify strengths and weaknesses in their interview style and skills. In the diary, the first author recorded their responses to the participant and the interview content, and their observations of themselves as a researcher and what the experienced had uncovered in terms of their own assumptions and how these could impact on the research.

Early transcripts were checked by the third author to ensure that interview style was of good quality and that the questions did not unduly lead participants’ responses. A transcript of the first interview was reviewed by a research supervisor and feedback given on strengths and weakness of the interview style (the weaknesses were not such that the participant’s responses were unduly influenced). The first author incorporated this feedback into the next interview which was checked again. The first author was particularly mindful of the tension between their clinical work which involved making sense of client difficulties (formulating) throughout discussions and where paraphrasing is considered a skill, and their researcher role where paraphrasing can be problematic (McNair et al., 2008) and it is important to allow the participant to share their views with minimal interruption or influence from the researcher.

2.8.2.3 Reflexivity during analysis

The reflexive diary was maintained by the first author throughout the analysis process. During analysis, it has been suggested that reflexivity can be described as interpreting one’s interpretations of the data (Alvesson & Skoldberg, 2000), and the first author embedded this within each stage of the Thematic Analysis. Nadin & Cassell (2006) propose that merely keeping a diary is meaningless if the researcher is not committed to reflexive thought and a desire to integrate this into the process. The first author recognised that it was important for them not only to note down their reflections but highlight ‘action points’ and ‘things to be mindful of’ to ensure that these reflections fed back into the way the research was approached.

3. Extended Results

3.1 Additional Results

3.1.1 Study retention

All individuals who expressed an interest in the study were deemed eligible to take part. Two participants decided to withdraw from the study after the orientation meeting. One of these participants became physically unwell (MS-related) and required hospital admission so could not continue, and the other participant withdrew due to time constraints. Only demographic data had been collected for these participants at the time of their withdrawal. The remaining 12 participants completed the study in full.

3.1.2 Study output

Emails were used by four participants to send their images, and the remaining 8 opted to use ‘Whatsapp’. All except one participant took a minimum of five images. The minimum number taken by one participant was four, and maximum was eight. Eight participants sent short narratives with their images. Semi-structured interviews lasted between 35 and 80 minutes.

3.1.3 Additional demographics

Five participants regularly used mobility aids and described themselves as having visible mobility difficulties in addition to invisible symptoms. Seven participants were in paid full-time or part-time employment, and two more worked in voluntary roles on a flexible basis.

3.1.4 Participant feedback of interview process

Many participants reported during their debriefs that they had found the interview to be a positive experience and that they had felt listened to by the first author (LSP). One participant sent a letter to LSP after the interview, which included the following: “It’s been a pleasure being part of this study. You are a very easy person to talk to, which made being a part of it very easy and not too stressful.”.

Others commented that the process had been “thought-provoking” and have facilitated reflections and insights about their invisible symptoms that they had not considered before.

3.2 Extended comments on theme ‘The challenges of conceptualising the invisibility of symptoms’ and subthemes

All participants reported experiencing clusters of invisible symptoms, and none were reported to be experienced in isolation. Table 7 in our journal paper shows the symptoms that the participants defined as being invisible. Participants’ definitions of specific symptoms differed at times and were subjective to the individual. For example, some participants identified bladder dysfunction as being an invisible symptom because it generally cannot be seen by other people. Some participants, however, conceptualised this as a visible symptom because there can sometimes be physical evidence for this, and there have been times when their bladder problems have been very much visible to others (e.g. when they have lost control of their bladder in public). Some people defined their balance problems as invisible, because although sometimes others could see that they were struggling to walk or stand, they cannot see the underlying cause. Others, however, did not define balance problems in this way, as for them the fact that any aspect of their symptom was visible excluded it from being defined as invisible.

3.2.1 Extended comments on subtheme ‘Others’ blindness to my internal struggles’

Some participants explained that even in situations where symptom-related behaviour is evident, the reason and symptom behind the behaviour is hidden:

> *it’s something nobody else sees …I think they can see I’m cold, but they wouldn’t know what the reason was as to why I was so cold (Fay, speaking about wearing a thick coat to manage temperature sensitivity)*

The majority of participants' understanding of their symptom invisibility extended beyond the direct experience of the symptoms themselves, and to other areas of their lives impacted by the symptoms which they described as also being invisible to the observer, such as their emotional responses to their symptoms, and worries about the future which are hidden and internal:

> *So, I may look well but you don't know my troubles…by looking at me you don't know that I have to go to hospital…that I might have to think about 'well will I have mobility issues in the future?'... 'will I be employed much longer? Am I going to struggle cognitively?' (Esther)*

> *someone can look like they're having the best time on the outside but actually under all that there can be a lot of stress and anxiety and pains that people can't see. (Laura)*

Several participants spoke about the invisible experience of having to take medication to manage the symptoms, something that others don't see and are unaware of. Ann Depicted this in Figure 17:

> *…people don't see it, because I don't sit there and take my tablets in front of people…it's just invisible. (Ann)*

Some participants whose images were of themselves reflected on the fact that any person looking at the image would not have any way of knowing about their MS or invisible symptoms:

> *If you looked at that picture now, you wouldn't know that I had MS. (Claire)*

> *I know I look like I'm probably able and active here, but I can't do anything really. (Jacqueline)*

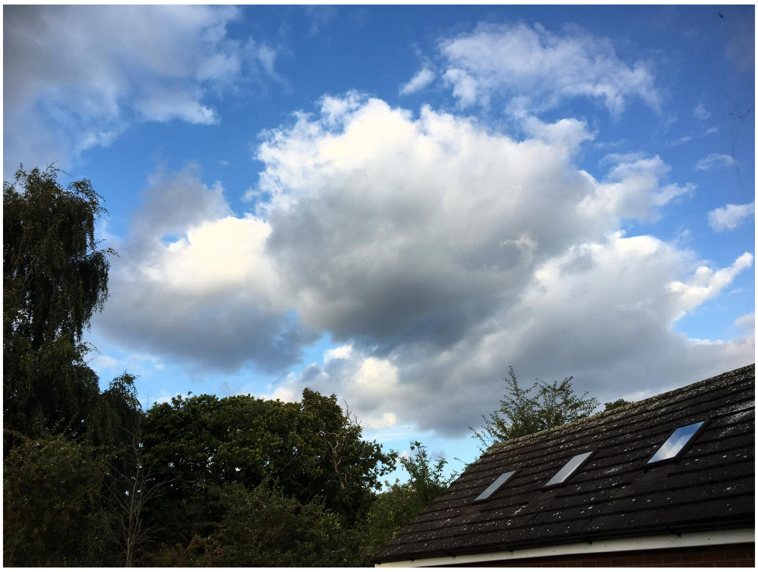
The images were reflective of their daily lives in this sense, where they looked "normal" but were suffering with symptoms. Stuart captured the discrepancy between how he feels internally as a result of his invisible symptoms, and what people actually see in Figure 18.

Figure 17

'Behind the scenes' taken by Ann

Figure 18

'Cloud' taken by Stuart



> *The bright ‘top’ with the darker cloud underneath made me think about invisible symptoms of MS; pain, fatigue, and depression, You can’t see them on the surface… someone looks down so you see the fluffy white bit on top, but if you dig deeper you see the darker bits underneath… that’s a good representation of invisible symptoms, it’s the dark underbelly you can’t see on the surface. (Stuart)*

3.2.2 Extended comments on subtheme ‘Can’t be seen, can’t be spoken about’

We write in the journal paper about the range of ways that participants attempted to communicate their invisible symptoms. Participants also invited others to think about commonly experienced phenomena, highlighting the ways in which this mirrored or was different from their symptoms, as a way of explaining what their invisible symptoms are like:

> *If I’m talking about being tired then I try to relate it to a hangover or being jetlagged. And my boyfriend came back from a work trip a while ago and there was a bit of jet lag and he had loads of delays and problems getting home and he was really tired. And I said to him ‘that’s what it’s like when I’m really tired is how you’re feeling now. (Laura, speaking about trying to communicate what MS fatigue is like to her boyfriend)*

Some participants invited the researcher during their interviews to draw on their experiences to start to make sense of what invisible symptoms feel like:

> *Sometimes it will feel like… you know when you burn your tongue on a cup of tea or something? It often feels like that, but then sometimes it will feel quite a lot worse than that. (Ann, trying to communicate the sensory symptoms she experiences in her mouth)*
>
> *… I think my neurologist called it ‘the hug’. It’s like someone’s got their arms around my sides and is literally crushing... It’s like you need to take your bra off but you haven’t got a bra on. You know when you kind of…like if you’ve had a really big dinner and you ‘oh! I’ve got to let this go’. (Jennie, trying to explain the sensation of the invisible ‘MS hug’.*

Daniel captured his sensory symptoms by comparing an aspect of this to the tangible experience of holding a hairbrush on the skin:

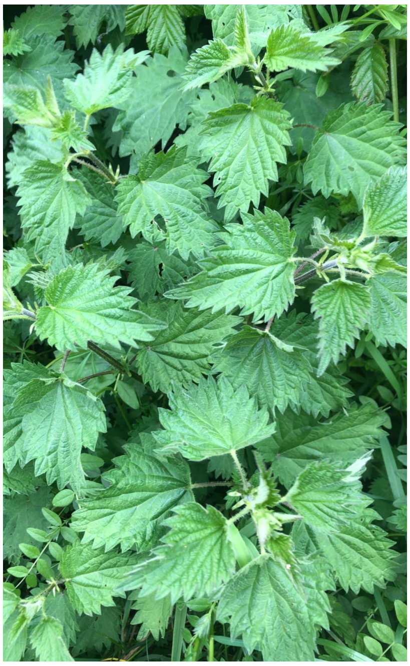
> *I suddenly start itching. It could be on my arm, it could be on my chest, it could be on my stomach, it could be on my back. For no reason whatsoever. And I will itch and itch and itch and itch and I can make it really quite sore…when it happens you can scratch it really really raw…And what you feel from that picture is how it feels. It's a visual representation of how it feels. What they actually are is sparks, it goes hot as well. There's lots of little…. Instead of there being one area, one solid area, it's lots of little points. Almost like if you got a hairbrush and you touched your skin with the hairbrush, and you've got lots of little spikes. (Daniel)*

By comparing their symptoms to something tangible for others, some participants felt able to communicate what their symptoms are like to a degree. However, there was still a prevalent issue that the language devices used by participants did not adequately allow them to explain their invisible symptoms. Esther acknowledged that whilst she tried to capture something that others could relate to in her image (Figure 19), she still felt that this did not fully communicate her sensory symptoms:

> *So, whilst the strange sensations I have I can't…it's really hard to explain them to someone who hasn't had them. It's not like a nettle sting as such, but I was trying to symbolise something that is a sensation that people can sort of relate to. (Esther)*

Figure 19

'Sting' taken by Esther



3.3 Extended comments on theme 'Conflicts of legitimacy' and subthemes

Symptom invisibility generated a clear conflict around legitimacy for the majority of participants where they not only had their realities questioned by others but could doubt their own judgements. This presented them participants with challenges in terms of navigating this conflict.

3.3.1 Extended comments on subtheme 'An invisible reality (epistemic issues of conflict)'

Invisible symptoms created a conflict between the participants' felt realities and the fact that these experiences cannot be seen, quantified, or communicated, generating issues around their legitimacy and a need for external validation of their realness.

Many participants spoke about how the reality of their experiences can feel illegitimate in the context of their social interactions with others because of the invisibility of their symptoms. Jacqueline spoke of her attempts to communicate the legitimacy and reality of her symptoms, and the notion that they could be perceived as not being ‘real’ to others:

> *I say always to people I’m not faking being ill… I’m actually faking being well. (Jacqueline)*

Participants spoke about the difficulties the invisibility of their symptoms bring when there are no physical indicators of their existence that can be seen by others, particularly when others tend to understand disability to be something that can be seen (e.g. a wheelchair):

> *unless we all walk round with ‘I have MS’ tattooed on your forehead, people don’t know. And it’s very frustrating…most people with MS don’t look different than anybody else. You know, we’re not…we’re not an amputee, we’ve not got…a lot of us aren’t permanent wheelchair users. (Fay)*

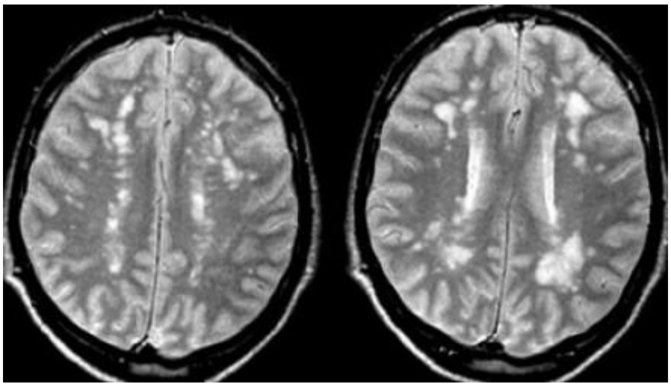
> *wheelchairs make people understand that it’s a disability...for most people…a chair, that means they’re disabled. (Ann)*

Not only were MRI scans deemed by some participants to verify the realness of their invisible symptoms to others, but also to themselves. Fay spoke about her MRI scans and how this helped to normalise and validate the realness of her invisible symptoms as something she could make sense of (captured in Figure 20). She spoke of her invisible symptoms ‘being seen’ by the MRI scanner, confirming their reality to her.

> *[The MRI scanner] can actually see and make a picture like that… Of inside me that there’s no way of anybody else seeing…and see where lesions are, and explain why certain bits of me don’t work properly…without the MRI, I wouldn’t know I’d got MS. I’d just have these weird symptoms and weird things happening to me…that MRI made all these weird symptoms I was having make sense…it normalised me…without that you can’t see what’s causing it. …this makes it visible to the doctors to know what is wrong with me. (Fay)*

Figure 20

‘Diagnosis and Progression’ taken by Fay



3.3.2 Extended comments of subtheme ‘External invalidation (experiential issues of conflict)’

Many participants spoke about other people commenting on how healthy they looked, and how this invalidated their internal invisible struggles:

> *‘Oh aren’t you doing well’ or ‘you’re doing really well’ but they don’t see that actually you’re not, you’re absolutely shattered. (Fay)*
>
> *people say ‘well you look well’…But it’s all the stuff that’s going on inside, it’s like a duck paddling away furiously under the water. (Jaqueline)*
>
> *‘well you know, you look normal’, ‘you look more normal than you ever did’ but actually it’s not…there’s other stuff in the background. It’s like the stuff that goes on in the wings rather than on the stage. (Ann)*

The issues of being delegitimized by other people’s responses ranged from people with MS feeling that others did not accept the existence of their symptoms and would attribute symptom-related behaviour to character flaws or unusual behaviour, to feeling that although others acknowledged the realness of their symptoms and even at times made attempts to understand, they still did not fully appreciate the symptoms and could overlook the very hidden needs of people with MS.

Jennie spoke about the annoyances of other people attempting to empathise with her experience of fatigue:

> *it's one of the most annoying symptoms I think because people just don't get it. They say, 'oh yeah, I'm tired as well' and it's like you actually have no idea of how* <u>*this*</u> *tired feels. (Jennie)*

Other participants spoke of similar experiences, where efforts from others to draw comparisons and empathise with their experiences actually resulted in the minimisation and dismissal of their invisible symptoms.

> *you say 'Oh, I'm tired' 'I didn't sleep well last night'. But they don't see that that's not the same as them going 'Oooh yeah, I'm tired, I went out last night and did such and such and…' It is a different type of tired, and it's an* <u>*invisible*</u> *type of tired. (Fay, speaking about her fatigue)*
>
> *there's other people where I guess you come into work and you say "oh, I'm really tired today" and they're like "Oh yeah, me too" but* <u>*their*</u> *tired and* <u>*my*</u> *tired are just not on the same par. (Laura, speaking about her fatigue)*

Many participants expressed feeling that others do not understand their invisible symptoms, which felt invalidating. However, participants also showed a lot of understanding towards others as to why their symptoms are often misunderstood:

> *And I find a lot of people don't understand to be honest with you. They might be sympathetic but they don't really understand that much about it (Wendy)*
>
> *People don't get it, and you can't blame them…If you've not felt it, you really won't understand it…if you haven't felt it, you're just not gonna get it. And that's fair enough, why should you understand something you've never experienced?...You can't expect people to know what they don't know. (Jennie)*

Laura suggests that instead of trying to fully understand or assume understanding of invisible symptoms, it is important for others to accept and acknowledge that they can never truly know what MS invisible symptoms are like:

> *You can be the most understanding person in the world but unless you actually know what that feeling is like…I think that's kind of important for other people to… they might say to you 'oh I understand how you're feeling' but accepting that they don't fully know what it's like, or that they can sympathise or empathise with you but they don't quite [understand] (Laura)*

Jennie felt that wider public acknowledgement of invisible symptoms has improved over the years and felt that this was linked to the increase in social media campaigns that promote awareness. However, she felt that much is still left to understand:

> *A lot more people understand it a bit better now…it's like 'oh yeah I've heard about that', but they won't know what it feels like still. They know it exists, they don't know how it exists, shall we say. (Jennie)*

Some participants commented that their closest loved ones have tried to understand and have made conscious efforts to validate their experiences. Often, they have more of an understanding than the general public and at times even notice when the participants are struggling with symptoms. However, these accounts were also accompanied by the commentary that despite the efforts of loved ones, participants still do not feel understood due to the invisibility of their symptoms:

> *My partner's really good, he seems to notice sort of triggers now, but people who don't see me all the time, they don't. They just don't understand. It's just totally, totally invisible…Family is a little bit different because they sort of understand more, but not always…However much people try to understand, they don't know because they're not you. They're not the ones that's feeling the exhaustion and the frustrations. (Fay)*

> *my friends and my family and my boyfriend are very understanding people and they do understand, but without them actually living with it, it must be really hard for them to fully appreciate what it's like (Laura)*

However, some participants reflected that even when they don't feel fully understood by their loved ones, it is a comfort to know that they can be relied upon for support:

> *loved ones and close family and partners don't necessarily understand your symptoms…But they are there, I know all of them are there should I want to go and talk about things. (Esther)*

Some participants spoke about the support they receive from their family and friends. Claire captured her family in Figure 21 and spoke about how they keep her "grounded". Jacqueline captured the support of her family and friends in an image from an MS Society fundraising event she organised, and they took part in (Figure 22). So, although participants largely felt misunderstood with regards to their invisible symptoms, many still felt supported in other ways.

> *it's just great that I have to say, all my friends are in this with me, together. So this is a really positive photograph…it captures, you know, what it's all about, it's 'let's try to stop this together'. (Jacqueline)*

Five participants spoke about the importance of pet ownership in their experience of invisible symptoms in which pets provided companionship, comfort, compart and unspoken support. Some highlighted that they felt understood and validated by their pets in a way that they did not by other humans, and that their pets would sense their internal struggles with invisible symptoms (Figure 23 and 24 show how some participants depicted this):

> *I think animals sense if there's something wrong…if you're ill or you're doing too much I'm sure that they sense it (Kate)*
>
> *And I think also he [her dog] knows when I have my ups and I have my downs. You know, he obviously can't talk but yeah, he knows. (Jacqueline)*

> *there's just something about Ben [her dog] and dogs…they show their understanding without [words] (Wendy)*

Figure 21

'Family' taken by Claire

Figure 22

'My MS Walk' taken by Jacqueline

Figure 23

'Rodney' taken by Kate

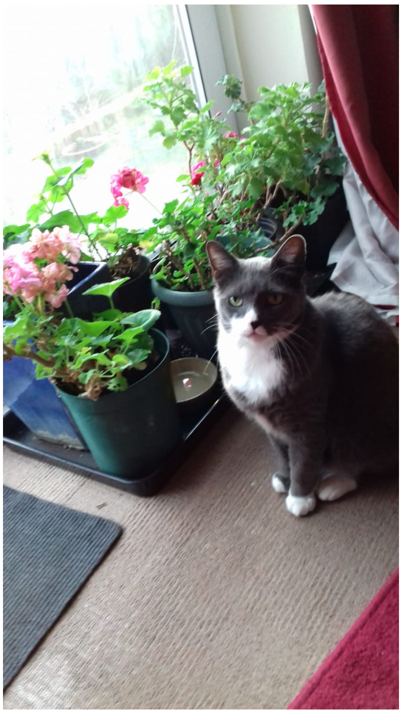


Figure 24

'Lakie' taken by Jacqueline

Participants expressed beliefs that others perceive disabilities to be more legitimate when there are visible indicators of this, and experience invalidation in the absence of this. Fay described that other people will “glare” at her when she parks in a disabled spot in a car park (which often has an image of a wheelchair), and that it is usually only when she gets her walking stick from her car that they “stop mumbling”, presumably because they have seen physical evidence of a disability. She expressed feeing that people who park in disabled spaces who do not use obvious mobility aids are regarded as “lazy” by members of the general public, which invalidates the reality of their struggles. Ann spoke about her optic neuritis (the experience of which she depicts in Figure 25) which resulted in temporary partial blindness, and how she was treated differently by others to how she would have been treated if she had a visible sign of her disability:

> *Nobody can tell there’s anything wrong, your eyes just look fine. So, nobody makes any kind of allowance for you. If someone is walking around with a white stick, then people know ‘right’. (Ann)*

Embedded within participants’ conflicts of legitimacy were fears and expectations of being delegitimized by others disbelieving them around their symptoms or judging them negatively, even if they had not experienced this directly. Some participants indicated that this inhibits how or whether they talk about their symptoms to others.

> *it’s more my perception of what people might think about me. You know “is she thick?” and “what’s going on with her? (Kate, speaking about how she expects people to judge her when she is struggling with cognitive symptoms and stops mid-sentence to word find).*

Even where participants hadn’t experienced another person being directly critical or dismissive towards them, others ignoring the existence of their invisible symptoms led to invalidation by omission:

> *I’ve never felt like people don’t believe me as such but it’s still quite rare that people ask how I am because they assume, they forget that I’ve got this. (Esther)*

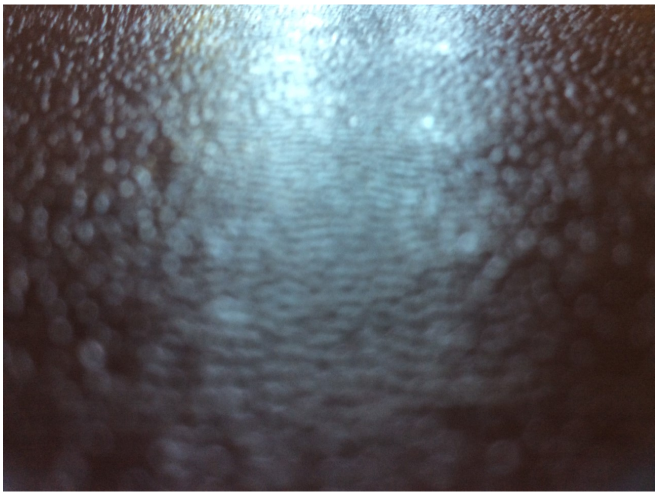
Where participants hadn't received negative comments from others, they spoke about their awareness of other people with invisible symptoms of MS receiving these, which they feel indirectly invalidated by.

Participants spoke about a range of emotional impacts the invalidation of their symptoms had on them, and how these impacts are invisible themselves. This creates a scenario where not only are the symptoms invisible, but also the emotional impact of living with these symptoms can't be seen by others:

> *it does worry me that people maybe aren't getting the support that they could benefit from just because they look fine and actually may even be that emotionally, they may need a bit more of a helping hand (Esther)*

Figure 25

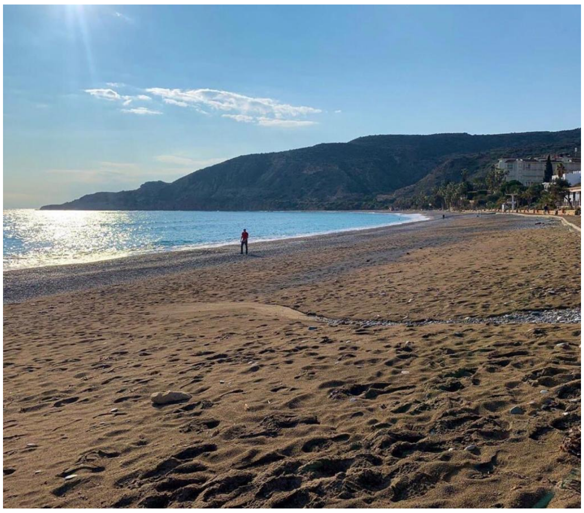
'Coming out of Optic Neuritis' taken by Ann to depict what she 'saw' when she had optic neuritis.



Jennie captured an image (Figure 26) to represent the loneliness she feels living with invisible symptoms where others do not understand her experience:

Figure 26

'Lonely' taken by Jennie to represent that even in the nicest of places, she still feels alone in her struggle.



3.4 Extended comments on theme 'Navigating life with invisible symptoms' and subthemes

The invisibility of symptoms offered people with MS a choice as to how they navigate living with the symptoms more generally, and also the issues of legitimacy they bring.

3.4.1 Extended comments on subtheme 'Making symptoms fit to life'

Some participants characterized the 'making symptoms fit to life' approach as a 'battle' and 'fight against' their symptoms, where they felt able to have a sense of control over their illness more generally. Whilst speaking about her reluctance to change her working hours in response to her fatigue, Claire reflected on her desire to 'fight back' against her symptoms:

> *I'm not gonna let the illness win…It's just…it's giving in, isn't it? It's giving in to the illness. (Claire)*

Some participants 'pushed through' and attended to daily tasks despite suffering with their invisible symptoms, however found that doing so sometimes exacerbated their symptoms and made them feel worse:

> *Often, I try and do things then I get over- fatigued. (Kate)*

Laura spoke about returning home from work and usually feeling fatigued. She described a "love-hate relationship" with her bed (depicted in Figure 27), where she fights against going to bed early so that she can spend time with her partner, despite knowing that she needs to rest. This was usually motivated by not wanting the invisible symptoms to "*rob*" her of or "*absorb*" her personal time:

> *if I don't go to bed and I hold off the sleep and put it off for as long as possible…then the quality of my interactions are worse, and actually it's not quality time with someone if you're just sat there kind of lying down on the sofa and not really saying very much…So it's like a bit of a vicious circle and I'll put off going to bed, just because I don't want to give in to the sleep, I don't want to give in to being tired (Laura)*

Not only does fighting against fatigue backfire in that the time she spends with her partner doesn't feel like quality time, but she also observed that staying awake when she is struggling causes the fatigue to worsen.

For some participants, 'pushing through' their invisible symptoms and continuing to pursue their regular interests was helpful in terms of managing their invisible symptoms. Claire spoke about attending 'Parkrun'[66] with her children at the weekends, and how doing this despite suffering with her symptoms helps her to manage the rest of the day (she captured this in Figure 28):

> *It gets me over that hump in the morning. When I wake up feeling like shit, if I don't go to parkrun I'm not gonna get over that hump and I'll be like "oh fuck it, I'll just stay in bed". So, if I get up and go to parkrun, I've got over that original hurdle of getting up and doing something, and I'm gonna feel less crap the other side. (Claire)*

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[66] 'Parkrun' is a collection of free weekly 5-kilometre events that take place worldwide and any member of the community can join.

Figure 27

‘Love-Hate’ taken by Laura

Figure 28

‘Parkrun’ taken by Claire

3.4.2 Extended comments on subtheme ‘Making space for symptoms in life’

A big part of making space for invisible symptoms for participants was to acknowledge when they needed to rest and ensure that they were able to do this. This could be a challenge when faced with the various responsibilities of life. Fay talked about going on a regular caravan holiday to ensure that she could have regular periods of rest without feeling the need to push through symptoms every day:

> *For me it just means that I can rest. Or not that I can rest, that I do rest. Whereas at home yes I can rest, I have days off work where I can sit and do nothing, but you can't really. You can't really sit and do nothing, because the jobs are there. (Fay)*

At the caravan, Fay can give herself permission to create space for her invisible symptoms and rest away from the daily demands of her life. Fay captured this in Figure 29:

Figure 29

'My bolt hole' taken by Fay

Claire took an image (Figure 30) of her bedside table to represent the times when she has to "*give up*" and get some rest to help her with her invisible symptoms "*instead of trying to fight on*":

Figure 30

Sleep taken by Claire

Stuart spoke about ensuring that he is able to take additional breaks as needed to rest at his voluntary job. Esther reflected that she has slowed her daily pace in order to make space for her invisible symptoms, representing this with an image of a snail (Figure 31):

Figure 31

'Snail' taken by Esther[67]

> *[The image represents] having to be slower, so making myself slow down… so not taking on quite as much stuff. (Esther)*

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[67] Esther also took this image to represent loneliness (related to the 'External invalidation' subtheme

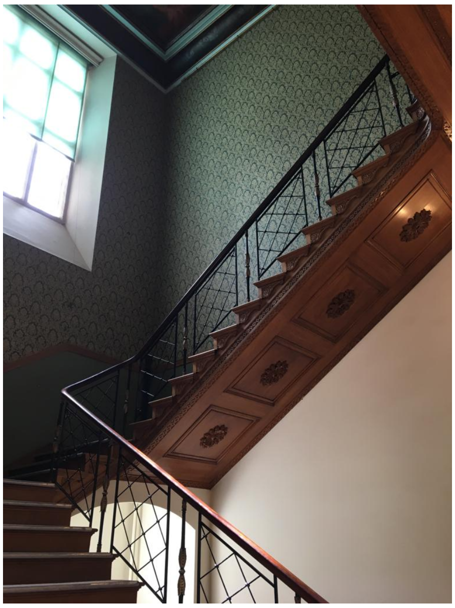
3.4.3 Extended comments on 'Implications of navigation styles on symptom invisibility'

Fay described her efforts for her invisible symptoms and her needs to 'be seen' as an "uphill battle", which she captured in an image of a staircase (Figure 32). Despite her struggles to make space for symptoms and be seen by others through applying for Personal Independence Payments[68] (she had to reapply when the government processes for accessing financial support with her disability changed), the sense of 'staying invisible' prevailed as an unintended consequence of 'making space':

> *I've now got to fight again to prove that I've got MS, that I can't do all these things that they expect me to do…If I was entitled to it 15 years ago, why should I have to have another fight and prove to people 'hey, I've got MS!'…Why should I have to fight these people?...that staircase is me on a blooming daily basis and anybody else out there with MS, fighting and saying "Look! It's me! I might look like a normal 51-year-old person, but I need this this and this'. And that picture to me, it just…I want to run up that flight of stairs and at the top shout "This is me!". (Fay)*

Figure 32

'Uphill Battle' taken by Fay



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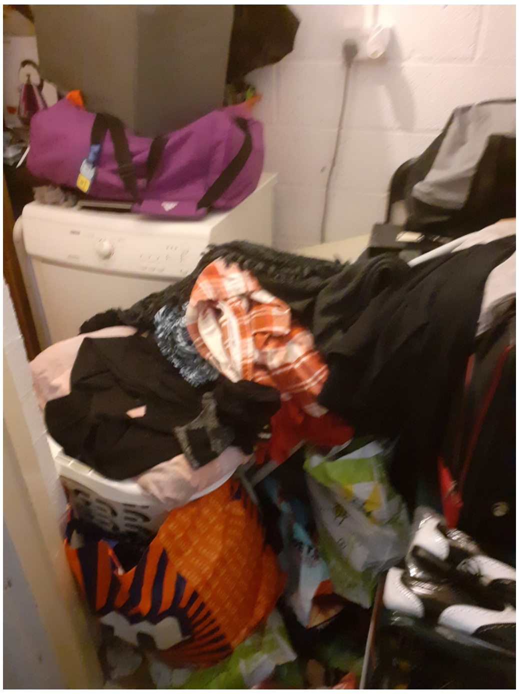
[68] Personal Independence Payment is a welfare benefit in the United Kingdom that is intended to help adults with the extra costs of living with a long-term health condition or a disability.

While many participants spoke about the implications of ‘pushing through’ symptoms or ‘making space’ for them on symptom invisibility, many also managed living with invisible symptoms by acting in ways to directly make their symptoms known (to ‘be seen’) or to keep them hidden (to ‘stay invisible’). Anniemac shared an image (Figure 33) which captured the way in which she ‘hides her life away’ from others in order to ‘stay invisible’ and avoid the judgment of others. She described that when she has visitors, she puts all of the things she feels too fatigued and depressed to tidy into a cupboard, and how this was a metaphor for her invisible symptoms themselves:

Figure 33

‘The Office’ taken by Anniemac



> *It’s just chucked in there, shut the door, nobody can see it, so it looks very tidy when you come in and it looks like life’s tidy. But in actual fact, it’s hidden in that cupboard, and it’s a mess…I don’t want anyone to see me living like that…I’ve isolated myself (Anniemac)*

3.4.4 Extended comments on ‘Finding a balance on a double-edged sword’

Participants chose from context to context whether to make space for symptoms or make them fit to their existing lives, taking into consideration the consequences of different navigation styles in terms of both their implications for symptom management more generally, and on symptom invisibility. Striking a balance requires many factors to be taken into consideration and was an ongoing and sometimes burdensome choice for participants.

Balancing daily tasks against energy levels and ensuring rest was important for participants. Stuart talks about the ways that he ‘pushes through’ his invisible symptoms to achieve some tasks but knows that he then must compensate by ‘making space’ and resting (captured in Figure 34). Esther also describes this balancing act:

> *[Before MS] I wouldn’t have to think ‘oh god I need to do the washing up now’ ‘I need to have a shower cause I’m not gonna have energy in the morning because I’ve then gotta do something else later on in the day’…I need to think about what I’m doing the rest of today to make sure I’ve got enough energy to go into the shower…I have to think ahead…So it’s always a constant balancing act if you like, to keep things at a reasonable level. (Stuart)*
>
> *When I know that I’ve done too much I need to take a rest. So, if I push myself too hard, I will have to just have an afternoon on the sofa. Or you have an early night and go to bed. (Esther)*

Stuart also spoke about needing to judge whether to push through fatigue and attend his voluntary job or to not go into work and to rest. He explained that although his job gives him a sense of purpose and value in society, he needs to balance the benefits of this against the disadvantages of pushing himself too much:

> *I have occasionally phoned in sick and said ‘you know, I just physically can’t do it’, but I’ll always try [to go in] (Stuart)*

Figure 34

'Sink' taken by Stuart

Claire appraised finding a balance with managing her fatigue as a learning process, and how it can be tipped by different factors which will make her more likely to either 'make space' or 'fit symptoms to life':

> *it's just learning to ring in sick to work instead of going in and coming back home again. I'm a lot more accepting. But then I still try and do things I shouldn't (Claire)*

Claire recognised that sometimes she 'makes space' and sometimes she 'pushes through' her symptoms at work. This decision is influenced often by other people's responses to her when she chooses to 'be seen', and her wish to be in control of her own workload:

> *If you say "look I've got a problem" they're gonna panic aren't they? There gonna go "oh god, what do we need to do? Are you okay?"…Cause the team leader is really bad, because you say to her "I'm struggling, I'm really tired" and then she's over then and she's like "well take more breaks". And it's like for fucks sake, I know, it's like "I'm telling you I'm tired because I have to, I don't need you on my back, I don't need you changing my workload because I'm tired… It's just them running around panicking when panicking is not really needed. And sometimes it's my own fault for going in when I really shouldn't do.*

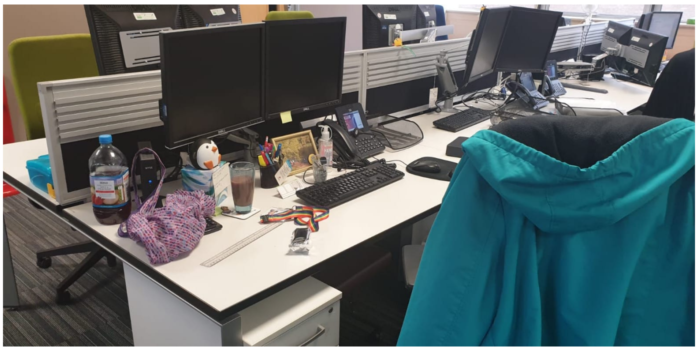
There is a conflict for Claire where she wants to be able to ‘be seen’ and ‘make space’ for her invisible symptoms by telling others when she is struggling at work, however does not want to invite unhelpful advice from others or have her decisions around her working patterns made for her. Here, ‘being seen’ and ‘staying invisible’ each have both potentially positive and negative outcomes and knowing how to balance this decision making against what is needed in the moment leaves Claire with a difficult task. She captured an image to represent her struggles at work in Figure 35.

Esther highlighted the double-edged sword as a conflict between the consequences for ‘staying invisible’ or ‘being seen’, where making symptoms known elicits pity from others, and keeping symptoms hidden leads to not being acknowledged:

> *you then get a load of pity or you get ignored…I just don’t want to be treated any differently, I want to be treated the same. (Esther)*

Figure 35

‘Work’ taken by Claire



Esther highlights a wish to maintain a sense of identity that is separate from her illness. This was discussed by others, who wish to strike a balance where their needs are acknowledged without being treated differently by others:

> *I don’t want the condition to just be me, I want to be a person as well. (Laura)*

3.5 Deductive findings and fit with the model

The deductive sweep, which involved extracting data that fit with Parker et al.'s (2020) previous findings from our meta-synthesis, largely supported the themes and conceptual framework from the meta-synthesis[69]. Each theme was developed from a combination of both the inductive and deductive coding.

All participants described a discrepancy between their outward appearances and what they experience internally as a result of their symptoms when conceptualising invisible symptoms. The invisibility of participants' experiences also extended beyond their symptoms and to other areas of their lives affected by their symptoms. This is completely consistent with the deductive framework, and the findings are presented in the subtheme 'Others' blindness to my internal struggles'.

Participants' experiences of invalidation as a result of other people's response to their symptoms were a key feature of their accounts. Participants also described a lack of understanding from others and the way in which the invalidation of their symptoms lead them to doubt their own legitimacy. These findings are presented in the subtheme 'External invalidation (experiential issues of conflict)'. Whilst some participants spoke about close loved ones being more understanding which does not fit with the deductive framework, their accounts largely fit with the framework as a whole.

The concept of 'hidden needs' was also a feature of participants' accounts, providing support for the framework. This often was in relation to participants' needs being overlooked in the workplace because of the invisibility of their symptoms. Fay described feeling annoyed and invalidated when her employers did not take account of her hidden needs when making a significant office move:

> *I used to work on the first floor, the main floor, the entrance was on our floor. To get into the building if I was in a wheelchair it would be impossible*

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[69] Please see appendix A & B for the deductive frame used in the analysis and the original conceptual framework

> *to do, but that's by the by. But they then decided to move our department up another floor. Nobody bothered to think 'will this affect Fay getting into work?". Because, the floor above now, the access into that from the staff car park means I have to walk up four flights of stairs to get into my office…nobody took that into consideration at all, or cared to be fair at all, that they were moving us up just another floor…it's things like that that are annoying, because this is my employer and they didn't take into consideration what effect this could have on me. (Fay)*

Claire described having to repeat herself and make her needs known each time there is a new HR staff member in order to have her hidden needs at work met:

> *You have to tell them [HR staff] all over again, don't you? They should know what the situation is and "oh look, here's another one that you need to tell all over again what's happening" (Claire)*

In-keeping with the deductive framework, participants spoke not only about the emotional impact of their invisible symptoms more generally, but also in relation to the invisibility of the symptoms and the fact that others cannot see them. Participants described the following emotional impacts in relation to living with the symptoms more generally (participants own words are used here):

- *Humiliation*
- *Guilt (about the impact the symptoms have on others)*
- *Hopelessness*
- *Sadness*
- *Annoyance*
- *Overwhelm*
- *Panic*
- *Stress*
- *Worry*
- *Frustration*
- *Anger*

More specifically in relation to the invisibility of their symptoms, participants reported '*annoyance', 'low mood', 'frustration'*, and '*exasperation'*. Jacqueline captured the emotional impact of her invisible symptoms in Figure 36, which depicts a tiny yacht in the middle of the sea:

> *that's what it [the image] represents, being this tiny little speck in this great big sphere of, well…dealing with these symptoms…the little speckness in a big, big, big blue sky and big, big, big blue ocean…we're little specks in a great big thing and how on earth then you deal with the tiny little specks that affect those little specks... that tiny smallness, that's how I feel… just this tiny thing and this great big thing that we're [people with invisible symptoms of MS] having to deal with. (Jacqueline)*

Figure 36

'Blue' taken by Jacqueline

Some participants recognised that the emotional impacts of their symptoms were invisible experiences in themselves, and ones they feel they have to cope with alone. In particular, these were depicted in '*Lonely'*, taken by Jennie (Figure 26), and in 'Overwhelmed and isolated', taken by Laura (Figure 8). Other participants' accounts of this were also heard.

Consistent with the deductive framework, many participants spoke about choosing to disclose their symptoms to others and choosing to keep them hidden from others. They also spoke about the dilemma they experience between ‘staying invisible’ and ‘being seen’. These findings are discussed in the theme “Navigating life with invisible symptoms’ and its related subthemes. Whilst not every participant offered accounts of ‘staying invisible’ and ‘being seen’, no contradictory accounts were present in the paper.

Overall, our findings provided substantial support for and fit with our deductive framework, with little deviation from this.

3.6 Additional findings on ways of managing life with invisible symptoms

Participants spoke about a range of ways in which they manage living with invisible symptoms more generally. Although these were relevant to our research aims and could be seen as an extension of ‘making space’ for symptoms, these did not form any coherent themes and so are better presented as a selection of miscellaneous ‘tips’ and considerations from the participants.

3.6.1 Pharmacological

Many participants spoke about pharmacological treatments as an important part of their invisible symptom management:

> *My infusion is vital for me to continue with things as they are in life really. So as long as that continues, it works. It makes a big difference to me. (Ann)*
>
> *I think I feel like I suffer from cognitive fog, so that’s like not really thinking very clearly at times. But I feel like the medication that I’m on, Tysabri, keeps all of the other symptoms at bay. (Esther)*

Medication was acknowledged as often helping with some, but not all invisible symptoms, and often would not remove the symptom entirely but would help these to be more manageable:

> *I have a constant migraine-style headache, which is managed with lots of pills, but at times it just gets worse and I haven't found anything that helps. (Ann)*
>
> *I take pain medication to knock the edge off the pins and needles, but they don't go away. (Kate)*

Participants also reported struggling with side-effects and sometimes feeling conflicted about taking their medications.

3.6.2 Behavioural

Using Practical aids

Some participants used aids in their daily lives to support them with living with invisible symptoms. Jennie spoke about the notetaking she does to aid her memory and the use of her 'Outlook' calendar every day. Anniemac makes use of a calendar diary (captured in Figure 37) to help organise her life, as it is difficult to remember appointments and events with cognitive symptoms:

> *any appointments I get I can put in on that, and I've only got to turn the page every week and I can see exactly what I've got for that week ahead. And it is wonderful. It's so simple, but it really helps... The organisation skills, the diary helps me there. (Anniemac)*

Anniemac also spoke about how making use of lists (Figure 38) helps her to plan around cognitive symptoms, and how using a 'fitbit' to prompt her to exercise helps with managing the impacts of depression (Figure 38):

> *There's actually my 'fitbit' on there…which helps me, keeps me moving because it nudges me occasionally, so I get up and move, so that's a good thing. Because of the depression, I'm not motivated to do anything. But*

> *because of the 'fitbit' every so often it just give me a little nudge and says 'move', so I do. (Anniemac)*

Figure 37

Taken by Anniemac

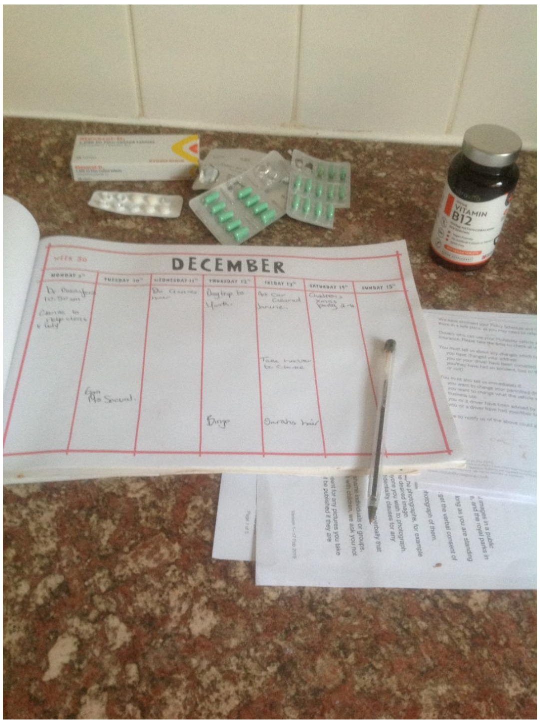
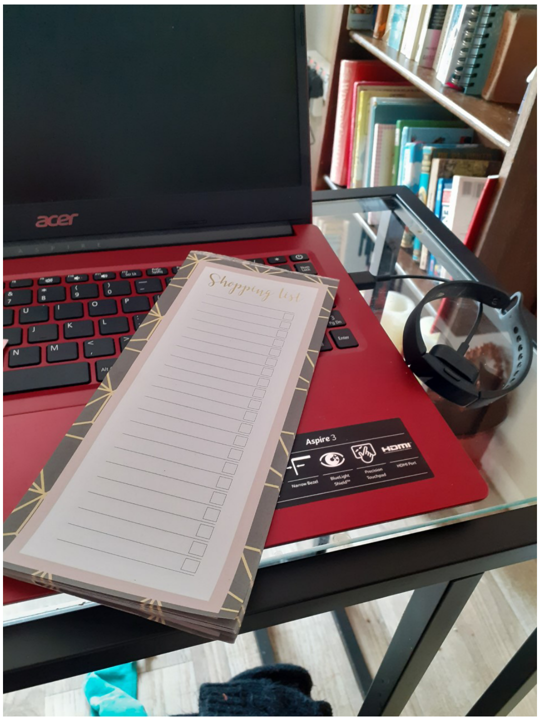


Figure 38

Taken by Anniemac



> *My shopping list is a shopping list, obviously. But it helps me to organise what I'm going to eat, where I'm going to buy it, that kind of thing (Anniemac)*

Fay described the adaptations she has made at work to help her manage pain and stiffness in her hands:

> *I use a small keyboard rather than the normal sized keyboard because it's easier to type than spreading my hands out typing. I've just got a little, one of the little compact keyboards… and laptops are smaller as well. (Fay)*

She also spoke about deliberately layering her clothing so that she could be more prepared for managing temperature sensitivity (i.e. adding or removing layers as needed). Ensuring that spare clothing was taken when leaving the house was important for people with bladder and bowel problems, in case of incontinence.

Meaningful Activity and other strategies

Participants spoke about exercise being a helpful way to manage their fatigue and balance difficulties. Kate spoke specifically about Pilates and yoga which help to strengthen her core muscles, and how she has made adaptations to the yoga positions so that she can still engage with this.

Self-soothing through listening to music and use of calming imagery was used by participants to help with managing the impacts of their invisible symptoms, particularly because it helped them to direct their attention away from the symptoms (this seemed especially true for those who suffered with chronic pain.

Engaging in meaningful activity that gave individuals a sense of value and purpose was important in terms of managing the global impact of invisible symptoms. This included work (paid and voluntary) and hobbies. Participants describe adapted their working patterns and hobbies to their symptom-related needs, for example:

> *I used to be able to do gardening, now I can grow things in tubs. (Fay)*

Stuart noted the importance of voluntary work for him:

> *One of the things that helped me with my depression was doing some volunteering, because one of the biggest problems for me when I stopped working was not having a purpose…the fact that I'm doing something, I've still got a purpose, I've still got some use…that is still massively beneficial to me…the fact that I'm being useful and I'm contributing to society if you like. (Stuart)*

3.6.3 Emotional/ Psychological

Support through MS Society and other groups

Although peer and group support contexts were not favoured by all participants, some participants spoke about the benefits of being a member of the MS Society. The MS Society offers support groups which some participants felt was helpful in managing the emotional impact of invisible symptoms. Stuart spoke about valuing the friendships and support he gets from the MS society members, and the sense of wellbeing he gets in being able to support others. He captured these aspects of his experience in Figure 39, an image he took following one of the regular MS Society meetings he is a representative at:

> *I've made a lot of friends through meeting people at the get togethers and things…If I'm the representative there that day…that's kind of good because it feels like I'm doing something, although I'm getting something out of it as well from socialising with friends and meeting new people… as well as making friends, you meet other people with MS as well so they have an understanding of what you're going through...And not just that, people know things, they've got experience of different things. They might be able to give you tips on 'I've done this, have you done that? Did you know you can apply for this?' all these sorts of things. So it's a fantastic opportunity for people to get together…So it's kind of a two-way street if you like, I'm helping other people, they're getting benefit from coming, but I'm also getting the benefit from the friendship and from the things I learn, and the fact that I'm being useful and I'm contributing to society if you like.*

Laura spoke positively about 'Shift.MS'; an online forum which has given her the opportunity to meet other people her age with MS and share experiences.

Figure 39

'Tables' taken by Stuart

Adopting a different 'mindset'

Many participants spoke about the importance of maintaining a "positive mindset", living in the present, and focussing on what they *can* do, rather than what they cannot do because of their invisible symptoms:

> *I don't dwell on it; I just take it as it comes. It is what it is. I make the best of a bad job. (Daniel)*
>
> *I'm just really grateful for all the people, and the countryside ,and the animals, and the farm animals, and the birds, that they're all around, you know? Because they all have difficult times as well, you know? And sometimes it's about putting it into perspective as well. Because it can be very easy to feel sorry for yourself. But if you put it into perspective, no one's got an easy life.* (*Wendy)*
>
> *I think it is useful to kind of think in the present, think about what's positive, you know, all that. Rather than think about ten years time, because in ten*

> *years time, who knows? Who knows what might be happening?...there's no point dwelling on that. So, I might as well look at what I've got, how I can improve what I've got, and make the most of what I've got. (Ann)*

Laura took an image (Figure 40) of all of the trips she has been on since being diagnosed with MS to highlight an important aspect of managing the impacts of invisible symptoms for her:

> *It was important for me to capture some of the positives of the condition because there can be positive effects from going through something quite challenging. I think it's the fact that it's motivated me to take my life into my own hands as much as possible, and really push myself to go places and do things. Because nobody knows what's going to happen in the future, with MS or without. So, it's just that kind of that 'seize the day' phrase but… just not letting it hold you back from doing things. (Laura)*

Figure 40

'Motivation' taken by Laura

Stuart spoke specifically about a mindfulness intervention he engaged with as part of a research trial, and how this helped him with the depression he identified as an invisible symptom:

> *When I was first diagnosed I spent a lot of time worrying about the past and worrying about the future, and I try to live more in the present now…there's no point in worrying about the past because it's gone, and there's no point worrying about the future if there's nothing you can influence on it because it's not happened, so there's no point wasting time now thinking things might get worse when it's not happened. You're wasting the better time you're having now, worrying about things that haven't happened…I will try and drop in a few bits of mindfulness…it allows me to focus rather than going onto autopilot, which again I do a lot…. Now I want to appreciate what I can now, you know, in case of whatever does happen in the future, which I'm not worrying about because it's not happened. (Stuart)*

Some participants felt that counselling had played a positive role in supporting them to navigate the impacts of their invisible symptoms and adopting alternative perspectives.

Laura spoke about the importance of being kind to oneself, and how social media accounts with positive messages can be helpful. She talked about a particular positive quote she saw on the 'Instagram' page of another person with MS:

> *it's just something to take as advice when you're going through a bad time, that actually you're still standing, like you're still here, and you're coping as best as you can….I guess it made me reflect on all the bad times that I've been through with my condition and that they have happened but there's still a lot of positives about life and still a lot of good things that will happen in the future… I think it's just not beating yourself up about each day. You might have a day where you feel really exhausted and tired, and not letting that kind of ruin your week. It might have spoilt your day a bit but actually you could… get a really nice healthy dinner, you could go to bed early tonight, and actually by tomorrow it might be fine. This is just one little bump, and the next day might be even better. (Laura)*

Wendy and Jaqueline took images to signify the way in which they live in "hope" with their invisible symptoms. Wendy's bird (Figure 41) symbolised her hope that she will continue to live well with her symptoms, and Jacqueline's rainbow (Figure 42) represented a hope for a cure for the disease:

> *…there is hope for me out there, and I can have a nice time, enjoy myself, be happy, because I'm no longer that frightened- I am frightened of the future but not as frightened as I was. (Wendy)*

Figure 41

'Garden Bird' taken by Wendy

Figure 42

'Rainbow' taken by Jacqueline

4. Extended Discussion

4.1 Further commentary on integrated conceptual framework

The conceptual framework from our previous systematic review (Parker et al., 2020) which informed the deductive framework was integrated with our new findings to produce the framework presented in the journal paper.

The original '*Looking healthy, feeling ill'* theme from the deductive framework was felt to relate to our main theme *'The challenges of conceptualising the invisibility of symptoms'*, whereby the discrepancy between what is felt by those with the invisible symptoms and what can be seen by others means that others are blind to the struggles of people with MS ('*Others' blindness to my internal struggles')*, and the language with which to communicate the abstract and strange experiences of the invisible symptoms is not available (*'Can't be seen, can't be spoken about'*).

The challenges of conceptualising the invisible symptoms creates a number of impacts, including '*conflicts of legitimacy*' (one of the main themes from our current findings) and also the '*hidden needs'* that are associated with the invisible symptoms and '*emotional impacts'* that people with MS invisible symptoms suffer with. All of these impacts were supported by our current findings and so all are presented in the framework.

People with MS invisible symptoms must then navigate the '*conflicts of legitimacy and other impacts'* as well at the physical management of the invisible symptoms, and do so either by '*making symptoms fit to life'* or '*making space for symptoms in life'* which can then have implications for and indirectly influence symptom invisibility ('*Implications for symptom invisibility*'). People with MS also might behave in ways to directly allow them to stay invisible or be seen. *'Staying invisible'* and '*being seen'* can have consequences in terms of compounding or resolving the impacts and conflicts of legitimacy associated with invisible MS symptoms, hence the dashed lines connecting these concepts on the framework to reflect these findings.

Not only did we find that choosing how to navigate their invisible symptoms can be a burden for people with MS (*'burden of choice'* from the previous framework), we found that people strive to '*find a balance on a double edged sword'* by weighing up the advantages and disadvantages of their navigation styles. The final framework reflects this new understanding.

This study has helped to further define the conceptual framework and our understanding of people's lived experience of MS invisible symptoms. In particular, we learned of the significance of the invisible symptoms' abstract and strange nature, and the way in which this limits the ability to conceptualise them for oneself and communicate them to others, which consequently impacts on the legitimacy conflict. We were also able to identify styles of navigating and the ways in which people with MS can directly or indirectly influence their symptom 'invisibility' according to their context and needs.

4.2 Relating findings to existing literature and theory

4.2.1 Types of invisible symptoms defined by participants

The range of invisible symptoms that participants reported were largely expected based upon the previous literature. Fatigue, pain, cognitive problems and sensory dysfunctions were most commonly reported by the participants, which is consistent with documented prevalence rates of these invisible symptoms. The number of participants who reported bowel and/or bladder dysfunction was lower than might have been expected based upon prevalence rates of up to 80% (bowel) and 75% (bladder) (Marrie et al., 2007; McClurg et al., 2017). Sexual dysfunction was also not reported, which is discussed in more detail in section 4.3.1. All participants reported experiencing or having experienced multiple invisible symptoms, supporting existing literature which states that symptoms are usually experienced in clusters rather than in isolation (Christogianni et al., 2018).

Interestingly, although balance problems and muscle stiffness are rarely defined in the extant literature as being invisible in nature, some of our participants defined them in this way. Some said that this was because although others might see them walking slowly, the symptom itself and the reason for the slower walking is invisible to others. This suggests that it is important to allow people with MS to self-define their invisible symptoms and their specific impacts for them, in both research and clinical domains.

4.2.2 The significance of language in conceptualising and communicating invisible symptoms

The findings highlight that the difficulties in conceptualizing and communicating invisible symptoms creates a sense of delegitimization around people's invisible symptoms, where people with MS question 'If one's symptoms cannot be seen or explained to others, then are they real?'. Language is said to provide a means to communicate, express, and consolidate a person's lived reality as a social phenomenon and is an objective form through which the human experience can be transferred into knowledge about their reality (Korneeva et al., 2019). Knowledge is thought to be reproduced through language, transferring information about a person's inner world in a way that can be understood and accepted by the listener (Korneeva et al., 2019). This theoretical understanding provides support for the idea that if people with MS do not have the language with which to accurately communicate their experiences, then what can be known about it or accepted as reality by others may be limited. This is consistent with our epistemological stance of critical realism; that our ability to access knowledge about the objective reality of one's experiences (in this case, of invisible MS symptoms) is mediated and therefore limited by what can be communicated about it (Al-Ababneh, 2020). Given that our findings highlight that invisible symptoms of MS are difficult to describe through language and the fact that many people with MS suffer with language and communication impairments as a symptom of the illness (Ntoskou et al., 2018), it is clear that people with MS are faced with a significant challenge in relation to communicating the validity of their lived realities to others.

It has been theorised that language also serves a cognitive function; that thought is materialized through language, and through this process becomes something accessible and concrete which allows an individual to access their reality

(Korneeva et al., 2019). Therefore, our thoughts exist in the form of words and this assists us to interpret our own experiences. Our findings suggest that people with MS struggle to access and interpret their invisible symptoms because of the difficulties in translating these strange experiences into a linguistic form, and therefore their tendency to doubt their internal experiences in the absence of physical evidence is understandable and compounds the legitimacy conflict around the reality of their symptoms.

4.2.3 Further discussion of invisible symptom management

As is consistent with the literature, we found that people with MS manage daily living by utilising a range of practical strategies and adaptations (Norton & Chelvanayagam, 2010; Stuifbergen & Rogers, 1997). This included making adjustments to their physical environments to be able to continue pursuing work and hobbies, and pacing activity to conserve energy (Frost et al., 2017). As is consistent with other research (Frost et al., 2017; Knaster et al., 2011), developing supportive social networks was important for people with MS in order to cope with the impacts of their invisible symptoms more generally. Whilst some support was noted from family and friends who do not have MS, participants in our study seemed to benefit more from receiving support from others who have MS, due to the shared understanding they have about invisible symptoms. This preference could perhaps be because there is less of a need or pressure to validate the existence of invisible symptoms when interacting with others who experience invisible symptoms, relieving the conflict of legitimacy for people with MS.

Engaging in activities that give a sense of purpose and meaning was highlighted as a strategy for managing the direct impacts of invisible symptoms such as depression and the indirect impacts of invisible symptoms on wellbeing. This is consistent with the evidence base around rehabilitation in chronic illness, where involvement in meaningful activity and the pursuit of valued roles is linked with better outcomes (Cardol et al., 2002). As engagement in personally meaningful activity is associated with greater psychological wellbeing more generally (Hooker et al., 2020), it is unsurprising that participants reported this to be helpful.

Some participants noted the benefits of attending to the present moment rather than dwelling on the past or worrying about the future, and of self-kindness in helping to manage the emotional impact of living with their invisible symptoms. The practice of Mindfulness (mentioned by one participant in our findings) originates from ancient Buddhist mediation principles, and over time is a practice that has been applied to various clinical settings to support wellbeing and stress management in long -term conditions (Goyal et al., 2014). Mindfulness is defined as 'paying attention in a particular way: on purpose, in the present moment, and non-judgmentally' (Kabat-Zin, 1994, p4). Mindfulness-based interventions aim to facilitate the development of mindfulness skills and have been shown to produce benefits for people with MS with regards to quality of life, mental health and some physical health measures (Simpson et al., 2014; Lorenz et al., 2021). Although these are not outcomes specific to invisible symptoms, mindfulness may present a promising way of helping to manage the wider impacts of MS, of which invisible symptoms are a significant part. Evidence also suggests that self-compassion is associated with better health related quality of life in people with MS, suggesting that supporting people to increase self-compassion may also help with managing the impacts of MS and the invisible symptoms which comprise a part of this experience (Nery-Hurwit et al., 2018).

4.2.4 Relating findings to stigma literature

As discussed in the journal paper, it was found that participants' ways of managing their invisible symptoms extended beyond the management of daily living with symptoms, to the management of the inherent invisibility of them. Participants expressed an awareness that their navigation styles in a given situation inherently influenced the degree of invisibility of their symptoms, and that their styles not only allowed them to navigate day-to-day living with the invisible symptoms themselves, but the legitimacy conflict they also bring, offering a sense of control over their 'illness identities'. The efforts of people with MS to conceal or try to show their invisible symptoms to others to manage other people's perceptions of them is consistent with the existing literature (Cook et al., 2016; Grytten & Maseide, 2005).

In line with Joachim and Acorn’s (1999) theoretical model for managing stigma with a chronic illness, people with MS recognised that the invisibility of their symptoms afforded them a choice as to how they navigate them, and they must decide whether to disclose or conceal their symptoms. In our findings, disclosure was linked with ‘making space for symptoms’ and ‘being seen’, and concealment was linked with ‘making symptoms fit to life’ and ‘staying invisible’. Participants reported that when they make their invisible symptoms known to others, this sometimes resulted in receiving support, but often they were met with invalidation and a lack of understanding. This is consistent with Joachim and Acorn’s (1999) model, as were our findings that ‘staying invisible’ can be stressful for people with invisible MS symptoms as they attempt to be perceived as ‘normal’, particularly when they are unintentionally ‘outed’ when their attempts to push through their symptoms create a struggle that is observable to others. Some participants identified that they deliberately disclosed their invisible symptoms to others (‘preventative disclosure’, according to Joachim and Acorn), and this was usually in order to manage expectations in the workplace and access the support required to do their jobs.

Some participant’s experiences echoed the process outlined in Corrigan et al.’s (2006) model of internalised stigma, whereby invalidating attitudes of others are internalised by the person experiencing the stigma. This manifested in our participants’ lives as questioning the reality of their invisible symptoms as a result of invalidation from others. Of course, the difficulties in conceptualising the invisible symptoms appeared to contribute to this questioning also (as discussed in section 4.2.2).

4.3 Implications of findings

4.3.1 Future Research

This research demonstrated that not only is it important to understand the potential impacts of symptom invisibility on a person with MS, but also to understand the different navigation styles used to manage life with the invisible symptoms. Whilst the present study offers us some understanding of this, we have not uncovered which contexts or situations influence or determine certain navigation styles over others, and which contexts certain navigation styles are most effective in (i.e. where the person with MS' desired outcome is achieved). Although it seems that the cost-benefit analysis people with MS perform is on an individual basis, it would be an interesting approach for future research to explore this process of decision-making around invisible symptoms, and what factors influence or 'tip the balance' in the direction of one navigation style over another.

It was interesting that although sexual dysfunction is a commonly reported invisible symptom of MS, and feedback from PPIE members suggested that this is regarded as a particularly important and impactful invisible experience, these difficulties were not mentioned by any of our participants in their interviews. It is possible that participants experienced these invisible symptoms, however, chose not to discuss them because of the personal and sensitive nature of the symptoms and their impacts on sexuality. Research suggests that people who experience sexual dysfunction, particularly when this is paired with a chronic and life limiting condition, find this difficult to discuss and it is often regarded as a taboo subject (Blackburn et al, 2018; Traumer et al, 2019). We might think of symptoms related to sexual dysfunction as invisible in nature due to the fact that they cannot be observed by onlookers, but also that there is an additional 'layer' of invisibility in that they are hidden and taboo subjects. We note that discussions around bladder and bowel dysfunction in interviews were relatively superficial in comparison to other symptoms, perhaps for similar reasons. As photovoice has been demonstrated here as a successful approach for gaining rich insights into people's experience of invisible symptoms, future research might apply this method towards specific symptoms that are perhaps even more hidden and invisible than others (e.g. sexual symptoms, bladder and bowel problems).

In the journal paper, Acceptance and Commitment Therapy (ACT) is suggested as a potential intervention to support people with MS to improve psychological flexibility in order to navigate their invisible symptoms. ACT interventions involve developing mindfulness skills, increasing self-compassion, and recognising and engaging with meaningful activity that is consistent with one’s values, all of which have been raised in our findings as being beneficial for participants (see section 4.2.3). Therefore, ACT and mindfulness-based interventions may not only be implicated for managing the legitimacy conflict brought about by the invisibility of symptoms, but in supporting people with managing the impacts of the invisible symptoms more generally.

4.3.2 Other Implications

It is important for photovoice studies to communicate the participants’ voices to a wider audience; not only to research health professionals, but to the public domain. Different photovoice studies have varied in the advancement of participant voice, and it is suggested that the least advancement occurs when a study manuscript is published with no further dissemination, and that photovoice researchers should expand their approaches to advance participant voice further (Evans-Agnew & Rosemberg, 2016). We intend to explore avenues for disseminating our findings and sharing participants’ images and corresponding narratives beyond the scope of journal publication, by liaising with key MS organisations and seeking to exhibit the images in a publicly accessible domain.

4.4 Extended Critique of Methodology

Our recruitment methods may have excluded people who are reviewed less regularly in MS clinics, and those who are not active members of the MS Society support groups, and thus may not have captured the views of those who have less accessible or regular avenues for formalised support for their MS and symptoms. Our study also did not recruit anyone with PPMS. Given that prognosis of PPMS is different to other sub-types, there may be differences in the way that invisible symptoms are experienced and managed. We are mindful, therefore, that our findings explore the experiences of white individuals with RRMS and SPMS, who either receive regular input from an MS clinic or are involved with the MS Society, and therefore do not represent experiences from other groups of people with MS. Whilst qualitative methodologies do not assume generalisability, a more diverse sample would have achieved a greater degree of transferability. Our inclusion criteria highlighted that participants needed to be able to independently use a camera device to take an image, due to the importance of capturing this for themselves and ‘through their own eyes’. This may have risked excluding people who were more physically disabled or suffering from severe tremor from participating in the study. Nobody who was unable to take an image by themselves expressed an interest in taking part in the study. If they had, we as a research team would have revisited the criteria to explore the use of equipment to support people to take an image.

Throughout the study, we employed reflexivity and ongoing evaluation of the research process. In addition to reflecting on the process in research meetings and maintaining a diary, the primary researcher’s (LSP) interview technique was independently evaluated to allow for identification of potential biases within the participant interviews themselves. Weaving this reflexive and evaluative process throughout the study served to improve the quality of the research.

Photovoice studies often report high attrition rates (Baker & Wang, 2006; Williams et al., 2014). The rate of attrition for this study was comparatively low, and could possibly be explained by the efforts to build a positive relationship between researcher and participant, ensuring regular opportunities throughout data collection to speak in person, via Skype, email, and telephone (as a lack of personal relationship with the researcher has previously been highlighted by Williams and Colleagues in 2014 as a likely factor in high attrition rates). We also followed advice from Baker & Wang (2006), reducing the number of images requested from participants so that participant burden was reduced, and using digital technology to take the photographs for ease (Boulos et al., 2011).

Although the PPIE in the study is a strength, with input at all stages including PPIE feedback on the analysis and findings, we recognise that this could have extended even further. Braun and Clarke (2013) suggest that checking the analysis with participants (or ‘member-checking’) can add credibility to research. On reflection, including a process whereby study findings are shared and discussed with the participants in advance of write-up may have improved the quality of the study.

The photovoice method created the novel opportunity for people with MS to capture and reflect upon their lived experiences as different situations arose and offered the creative freedom to communicate their experiences through their own 'lens'. The approach granted us access to people's experiences of invisible symptoms that are difficult to articulate through language and are described by participants as abstract in nature (Frith & Harcourt, 2007). The research also served a collaborative purpose, where participants communicated their experience 'through their eyes' and engaged in a process of sense-making with a researcher, in a way not offered before with this population.

4.5 Critical reflection

In this section, I will reflect upon my experience and learning throughout the research process. I kept a reflective diary from planning the study through to write-up. I will discuss my key reflections and share some brief excerpts from my diary here. The reflexive process, (including regular supervision and my diary) supported me to remain mindful about my own preconceptions (e.g. my own beliefs or assumptions about MS invisible symptoms and how these might be experienced or managed), and how these may have influenced the research process.

4.5.1 Overall Study Process

My stance on 'being a researcher'

Starting my DClinPsy training, I would not have described myself as someone who was particularly 'academically-inclined' in comparison to my peers, certainly viewing my clinical skills as a strength and the research element of training as something unpleasant to endure and tolerate in order to qualify as a Clinical Psychologist. Whilst I valued the importance of research in the profession, to marry my self-concept with the idea of being a ‘researcher’ stirred up a lot of anxiety about my aptitude for this. Unsurprisingly to me, therefore, the process of developing and completing a doctoral research project was challenging. The surprise, however, was how exciting it felt in parts too. I noticed that this feeling was usually there when I was engaging with members of the PPIE group and supervisors who expressed enthusiasm for the study, and when I wasn’t so caught up in thoughts of self-doubt. It was during these times I was able to come back to the purpose of this project and clinical psychology more widely- developing new insights and knowledge about a relevant subject area, in order to potentially benefit people in clinical populations. Below is a diary excerpt where I make some of these connections:

‘Helping people in clinical populations is a fundamental part of wanting to be a psychologist in the first place, right? So why am I treating research like it exists in a separate universe that doesn’t align with my values, or is something inaccessible to me?’

As I continued to make these links through the research process and the wider elements of the DClinPsy programme (teaching and clinical placements), the perceived ‘distance’ between the way I perceived my clinical role and the field of research lessened, and I began to better understand what it means to be a clinical psychologist and a scientist-practitioner.

Using Supervision

A valuable piece of learning from the overall research process for me is the importance of using the resources available to me in terms of support and research supervision. Of asking questions when I don’t understand, even if it means sitting with the discomfort that comes with ‘exposing’ my lack of knowledge to others. I realised early in the research process that my long-practiced response of retreating inwardly to work something challenging out (essentially, ‘muddling through without a clue’) was not going to be helpful, and of course, does not lend itself to the reflexive discourse between researchers that is an important part of qualitative research.

A break from research

Part-way through the data collection phase of the study, I took a break from the research process for 3-4 months while I recovered from a very literal break in my ankle bones. What struck me during this time, off the back of discussions with participants about the way in which visible indicators of symptoms provide validity to symptoms and the way in which people are treated as a consequence, was how differently I was treated by people close to me and members of the public now that I had a very visible cast and set of crutches to use when going about my daily life. The validity of my injury and mobility difficulties seemed not to be questioned for a moment, and those around me rushed to provide assistance when I approached a door, a flight of steps, or moved to get something I needed ("let me get that for you"). Even when I did not ask for help from others, they seemed to observe the crutches and respond almost automatically. Although by no means comparable to the experiences of those with invisible symptoms of MS, it prompted me to reflect on the differences between visible indicators of ill-health and those which are invisible:

'Would I have asked people for help with stuff if the crutches and cast were not there, or would I have struggled on with what I was doing? Did I feel more validated in requesting and accepting help because I could wave my cast at someone and they knew I was injured?' (diary excerpt).

I remained mindful of these thoughts on my return to the research process, using supervision and ongoing reflection to monitor their impact on my approach to interviews and interpretation of the data.

Covid-19

The coronavirus pandemic and associated restrictions began in the UK before my return to work when my data-collection had been halted and continued throughout the rest of the research process and the write-up. I found myself continuing the project in the absence of the coping strategies and social connections I rely on to maintain my wellbeing and emotional resilience, and to help me engage with the challenges of academic work more effectively. The absence of other activity certainly afforded me more time to immerse myself in the data and analysis, however maintaining self-care and boundaries around this was challenging. I found myself oscillating between immersing myself in the research process with little rest, and feeling overwhelmed and being avoidant of the work where I would not engage with it at all for a period of time. I noticed these patterns and began to actively apply the principles of Acceptance and Commitment Therapy (ACT) to my sense-making (I was using this a lot clinically at the time, and it made sense to practice what I was preaching). Over time, I started to behave more flexibly and effectively towards the research and writing the thesis. One of the ways in which I did this was noticing my internal experiences and how I was being avoidant of the work in response to these, then bringing myself back to reminders of the human beings at the heart of the project and my own values. I could remind myself of these things by viewing the images taken my participants, which helped me to connect with them and bring my attention and motivation back to the research.

I also note that I used supervision to maintain reflexivity in my interpretations and sense-making of the data, given that I was working with ACT clinically and did not want this to influence the conclusions I drew.

4.5.2 Planning the Study

I recall thinking multiple times throughout the planning stage (and more generally in the process) that ‘I always make more work for myself’. I have a tendency to put aside the familiar in favour of the challenge, even when this is to my detriment and causes me distress and discomfort. So naturally, when it came to developing a research project, I opted for qualitative research (something I had very little knowledge about) and within this field, a methodology I had never even heard of before (photovoice). I also had no experience of working or conducting research in physical health settings. I was nervous about embarking on the project and knew it would perhaps present me with more challenges than other ideas I might have developed, however, I felt that it would be a valuable learning experience and allow me to develop knowledge and skills I otherwise might not have explored on DClinPsy.

The biggest challenge initially was getting to grips with the methodology and exploring the existing relevant literature. I found myself wondering what would come up in the research based upon the literature, and also noted my own experiences that informed certain expectations. For example, I noted that I had an automatic assumption that photographs which were more metaphorical, abstract and artistic would be more interesting and uncover deeper meaning, and therefore knew that it would be important to keep a reflexive log of my responses to participant's images. Through my discussions with PPIE members in the planning stages, I noted that I had certain expectations around what kind of invisible symptoms might be described, and that people with invisible MS symptoms felt overlooked and ignored by others. I knew I needed to be mindful of this 'lens' when interviewing and conducting the analysis.

Planning the study also led me to reflect on the value of PPIE involvement and incorporating their feedback into the study design. In any future research, I will endeavour to have service user/PPIE involvement from the initial stages.

4.5.3 Gaining Ethical approval

The ethical approval process was new to me; however, I was aware that given we were asking participants to take images, there would be additional complexities to consider on top of what I already understood to be a frustrating and necessary process. It was helpful here to seek out ethical guidelines and considerations used in previous photovoice studies and discuss these in research supervision. I also decided to gain ethical approval via two channels (HRA and REC for HNS recruitment, and departmental approval for recruitment from MS charities). Whilst this approach did create more work (as the process and paperwork required differed for each route), it maximised avenues for recruitment. I had also hoped that recruiting from multiple sites would help to diversify the sample. The sample we had was fairly homogenous, however it did offer us a wider 'pool' from which to recruit, and so is an approach I would use again, particularly when using a methodology with a notoriously high attrition rate.

4.5.4 Recruitment

It was very encouraging to learn that everyone who expressed an interest in taking part in the study felt invested in the study, felt it would be of value and were particularly interested in being involved in a visual approach to MS research. Even those who had to withdraw due to personal circumstances expressed that they were saddened to do so. I think that the participants' enthusiasm for the project fed my own investment at this stage also. I felt that I developed positive relationships with the study participants, as there were multiple opportunities for contact and I used my own engagement skills which are a key part of my clinical roles. I think that this helped to reduce study drop-out despite the fact that this methodology can be burdensome for participants.

4.5.5 Data Collection

I kept a log recording all of my initial responses to each image I received from the participants, followed by a reflection after a second viewing approximately a week later, and after the interview itself. I found that I made assumptions about what the image related to and thought about which parts of the image I was interested to know more about. It also brought my attention to any potential biases that could occur should I lead the interviews with my own interpretations or questions about the image. For this reason, I shared my interpretations and points of interest in the image after the participant had spoken about them so as not to influence their responses.

Prior to doctoral training, my only experience of conducting interviews were clinical interviews in mental health settings, forming part of a psychological therapy process. A challenge I had during the interview process was ensuring that I stepped into a 'researcher role' as opposed to my more naturally held 'therapist role'. I noticed during the first interview I did, my propensity to ask questions in a therapeutic manner, and to formulate difficulties in my mind in the way I would in my clinical work. I noticed that this also led to me starting to actively ‘code’ during the interview, which could have influenced the questions I was asking and is certainly unhelpful in an inductive process. I was mindful of this during the interview, however sought supervision where my interview technique was reviewed and discussed. It was felt that whilst I did not unduly influence participants’ responses with my questions and comments, this is something I needed to continue to be mindful of throughout the process, holding my position as a researcher. I also have a tendency to offer summary statements of things participants have said (another clinical habit). As my diary extract shows, I needed to be cautious of this:

In my interview with Fay today, I summarised what she had said but used the word ‘upset’ when she had said ‘sad’. Luckily, I caught and corrected, but in future I need to be mindful- if you’re going to summarise what someone has said or check out understanding, use the exact words they did.’ ‘

I noted in my diary during the interview process that I had felt particularly sad after my interview with one participant who became tearful during our discussion. I reflected on the fact that we had many shared characteristics: both women, of a similar age, both working hard in a professional career, and both in the middle of discussions around family planning with our partners. Yet, her life differed from mine in so many ways, and she spoke about invisible challenges she experiences that I readily take for granted. I noted how I became emotional again when transcribing and coding her interview. I was conscious of not coding through an emotional lens and so ensured I came away from the work and returned to it. I specifically requested a supervisor to second check my coding of this interview. There was agreement about the coding and no sense that I had made interpretations that were not plausible, however reflexivity and noting down my feelings during and after interviews helped me to identify those potential sources of influence, and to seek out alternative interpretations of the data.

Aside from the challenges of data collection, I felt very privileged to have the participants share accounts of their lived experience with me. Collecting qualitative data gave me the opportunity to meet interesting people and to hear their stories, which I believe increased my investment in the research project itself and my sense of committed to representing their voices in a balanced and careful way. It certainly felt important to me that the project was something the participants would feel happy about having been involved in.

4.5.6 Analysis Process

My initial response to the data was that of overwhelm. The interviews had generated a lot of rich data, and it all felt significant, important, and vast. Initially, I found myself getting caught in my sense of wanting to give all of the stories shared a voice in the analysis, rather than answering the research aims. I discussed this in research supervision, where I was able to reflect on a very useful concept that once again, linked my researcher role with my clinical skills. We drew parallels between TA and leading clinical group interventions, in that when formulating clinically we cannot pull every individual strand of information into the mix, we must stick to the common themes which are useful and tell a cohesive story, and let go of the information that is although important to an individual in the group, does not serve the wider intervention and group. It was helpful to compare this to drawing themes in the TA and thinking in this way allowed me to start to shape the themes and let go of things that didn't address the aims of the research.

It was a challenge to remain in an inductive process given grounding in the literature and completing a systematic review of this. I valued having a reflexive log that I could make notes in throughout coding and developing themes, taking any pertinent issues to supervision. Regular supervision was helpful throughout analysis, as was using visual tools such as colour-coding, post-it notes and conceptual maps to organise my thinking.

As a person who likes to have a clear structure to follow when I work, qualitative analysis opened up a world of uncertainty and interpretation that I had to get comfortable sitting with. I think that my tendency to question my own interpretations, to go back and forth and consider all possible angles before firming up a stance, made for a rigorous analysis process, albeit a very exhausting one. Even as I conclude this final section of my thesis, I notice questions arising (‘was I reflexive enough?’ ‘did I address my aims fully?’). I acknowledge that it is in my nature to engage with this questioning process, but I can sit with acknowledging that I made good use of internal and external resources to complete this project.

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Appendices

Appendix A- Deductive Framework used for analysis

The deductive framework was developed from the findings of Parker et al.'s (2020) meta-synthesis and was applied as a series of questions asked of the data during a deductive sweep in the analysis phase. The bullet points represent examples of each occurrence in the data but were not exhaustive:

Deductive Framework

Does the participant describe a discrepancy between their outward appearances and what they experience internally as a result of their symptoms?

Does the participant make reference to specific symptoms as being invisible?

Does the participant speak about more general invisible experiences in relation to their symptoms/ symptom cluster?

Does the participant experience or anticipate being disbelieved, dismissed or invalidated by others with regards to their symptoms?

- Feeling others do not afford them the allowances or understanding that they would to someone with visible symptoms of illness/ visible symptoms are more legitimate.
- Invalidating experiences with healthcare professionals
- Describing symptom-related behaviours as being attributed to character flaws (e.g. being lazy).
- Beliefs that others will respond to them negatively (either publicly or privately)
- Others forgetting that they experience symptoms

Does the participant speak about feeling that others do not understand their experience of invisible symptoms?

- Unhelpful attempts of others to empathise with their experience of invisible symptoms

Does the participant doubt their own legitimacy in relation to their symptoms?

Does the participant speak about having hidden needs a result of their invisible symptoms?

- Having to make their needs known more explicitly around their symptoms in order for them to be met

Does the participant discuss the emotional impact of invisible symptoms?

Does the participant experience the emotional impact of MS as an invisible experience in itself (one they must cope with alone)?

Does the participant discuss choosing to disclose their symptoms to others?

- Convincing others of their symptoms
- Use of mobility aids when not needed

- Assuming responsibility to educate others

Does the participant discuss choosing to keep their symptoms hidden from others?

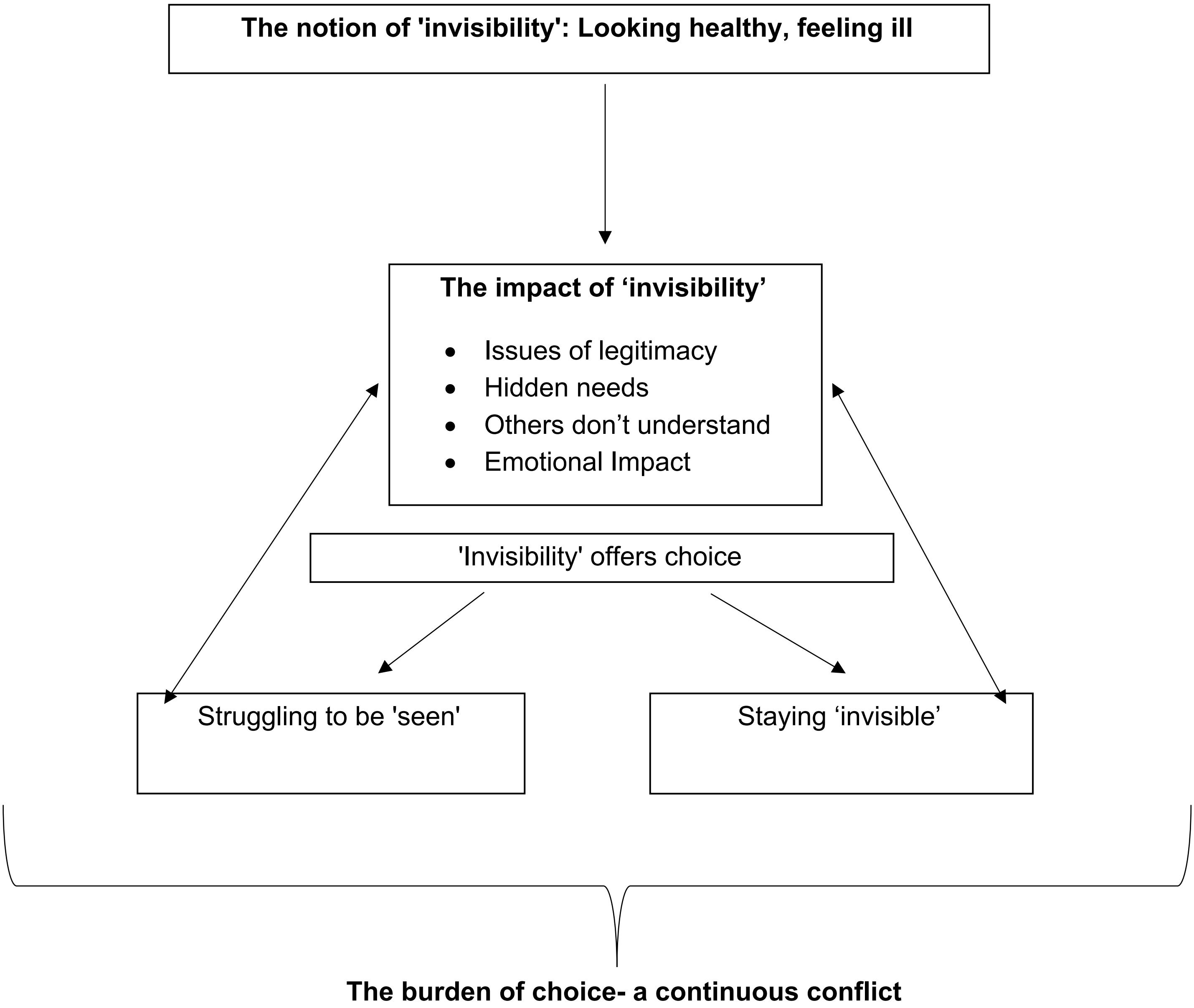
- Not speaking about existence of symptoms or their impact
- Minimising symptom-related behaviours/ saying ‘fine’ when not
- Shifting attention away from symptoms
- Continuing with daily activities despite exacerbation

Does the participant discuss the dilemma between being seen or staying invisible?

- Weighing up costs and benefits between telling others versus keeping their symptoms private?
- Acknowledging the choice or control they have over their illness identities.
- Fears that disclosure of symptoms would affect employment status and the way they are treated
- A conflict between the desire for others to know, and the fear of losing the perceived or projected image of the ‘self’.

Appendix B- Conceptual Framework

Figure B1. Original conceptual framework developed as a result of Parker et al.'s (2020) meta-synthesis.



Appendix C- Author Guidelines for ‘Social Sciences & Medicine’ Journal

https://www.elsevier.com/journals/social-science-and-medicine/0277-9536/guide-for-authors

Journal Word Limit: 9000 (including abstract, figures, tables, and references)

Journal Paper Word Count: 8,731

Appendix D- Questionnaire for Patient & Public Involvement and Engagement (PPIE) members.

Feedback Form for Presentation #1: The experience of living with and managing the invisible symptoms of Multiple Sclerosis

We believe that your views would strengthen the quality and relevance of our research. Therefore, please could you answer these questions?

Please tick one:

○ I have MS
○ I care for someone with MS

Please circle one number for each of these questions.

1. Do you think the purpose of the research is clear?

Not at all: 1 : 2 : 3 : 4 : 5 :Very
Clear Clear

2. How likely are we going to get people to sign up for this research?

Extremely: 1 : 2 : 3 : 4 : 5 :Extremely
Unlikely Likely

3. Do you think the way we plan to conduct this research is appropriate?

Inappropriate: 1 : 2 : 3 : 4 : 5 :Appropriate

4. Do you think the outcomes (things we hope to change) are appropriate?

Inappropriate: 1 : 2 : 3 : 4 : 5 :Appropriate

5. How well do you feel we have planned to involve people with MS and carers during the development of the intervention?

Poor: 1 : 2 : 3 : 4 : 5 :Excellent

Please tell us if you have any other comments about our proposed research or how we could do better (you can write on the other side of this sheet)

Thank you for your time!

Appendix E – Study Advertisement

Study Advertisement
Final version 1.0:28/04/19

Title of Study: Living with and managing the invisible symptoms of Multiple Sclerosis

IRAS Project ID: 255380

Name of Researcher: Le-Sharn Parker

Seeing the Invisible

Would you be interested in taking part in a study exploring people's experiences of living with and managing the invisible symptoms of multiple sclerosis (MS)? If you have a diagnosis of MS and experience symptoms you feel no one else can see, we would like to hear from you.

What are invisible symptoms of MS?

These are the symptoms experienced by a person with MS that are not visible or obvious to other people. They might include things like fatigue, pain, bladder and bowel problems, low mood, cognitive problems (with memory, for example) and sexual problems. There may be others depending on the person as no two experiences are the same, and you may experience a mixture of things.

What would the study involve?

We would ask you to take photos of things that capture your experience of invisible symptoms of MS, and spend some time talking about these pictures and experiences with our researcher. You don't need to be actively experiencing these symptoms to take part, but need to have experienced them previously. We are hoping for voices to be heard and invisible symptoms to be 'seen' through this research.

What next?

If you are interested in taking part or would like to know more about this study, please contact Le-Sharn Parker on the details below:

Email: Le-Sharn.Parker@nottingham.ac.uk
Phone Number: [Redacted]
WhatsApp' username: Le-Sharn MS Research

Appendix F- Consent form for contacting participants

Consent for information sharing
Final version 1.0:16/07/19

Title of Study: Seeing the invisible: An exploration of living with, and how people manage, the invisible symptoms of multiple sclerosis.

IRAS Project ID: 255380

Name of Researchers: Le-Sharn Parker, Dr Gogem Topcu, Dr Danielle De Boos, , Dr Clare Bale, Professor Roshan das Nair.

I give my permission for a professional from my Multiple Sclerosis care team to pass on my contact details to Le-Sharn Parker, so that she can give me some more information about participating in the above study.

Name of participant	**Date**	**Signature**
________________________	______________	____________________

Name of MS professional	**Date**	**Signature**
________________________	______________	____________________

2 copies: 1 for participant, 1 for the medical notes

Appendix G- Eligibility screening script

Eligibility Screening
Final version 1.0:28/04/19

Title of Study: Seeing the invisible: An exploration of living with, and how people manage, the invisible symptoms of multiple sclerosis.

IRAS Project ID: 255380

Name of Researcher: Le-Sharn Parker, Dr Gogem Topcu, Dr Danielle De Boos, Dr Clare Bale, Professor Roshan das Nair.

The following questions will be asked if the participant confirms they would be interested in taking part in the study:

Opening statement:
It's great to hear that you are interested in taking part in the study. Before we continue, I'd like to ask a few questions to make sure you are able to take part if that is okay?

Questions:

Do you have a diagnosis of MS and experience/ have experienced one or more invisible symptoms (give examples if potential participant is unsure)?

Are you aged 18 or over?

Are you able to communicate in verbal and written English, or have any particular communication needs (verbal will be apparent from the phone conversation but will allow the person to highlight any subtle difficulties)?

Do you have access to a smart phone, electronic tablet (with camera function) or digital camera? Are you physically able to use this on your own to take a photo?

Are you physically able to use email, mobile phone (with camera function), electronic tablet (with camera function) or 'WhatsApp' (mobile phone application) to send photos?

Next, I will ask you for some personal information to give me an idea of the amount of diversity in the people taking part. If you would prefer not to answer these then please just say:

How would you describe your gender?
What is your age and date of birth?
What is your ethnicity?
What MS sub-type have you been diagnosed with?

If inclusion criteria are not met or exclusion criteria is met:

Thank you for taking the time to speak to me today. Unfortunately, you won’t be able to take part in this particular study [give reason].

If inclusion criteria are met:

Thank you for taking the time to speak to me today. You are eligible to take part in the study. I’ll be sending you and information pack in the post including a consent form and some instructions for the next steps- would this be okay? Can I please take your contacts details so that I am able to send you this information and contact you again on the phone? Do you have any questions at this point?

(To all) Thank you for your time today.

Appendix H – Participant consent form

CONSENT FORM
(Final version 2.0: 16/07/19)

Title of Study: Seeing the invisible: An exploration of living with, and how people manage, the invisible symptoms of multiple sclerosis.

IRAS Project ID: 255380

Name of Researcher: Le-Sharn Parker, Dr Gogem Topcu, Dr Danielle De Boos, Dr Clare Bale, Professor Roshan das Nair.

Name of Participant:

Please initial box

1. I confirm that I have read and understand the information sheet version number 3.0 dated 19/09/19 for this study and have had the opportunity to ask questions. ☐

2. I understand that my participation is voluntary and that I am free to withdraw at any time, without giving any reason, and without my medical care or legal rights being affected. I understand that should I withdraw or my capacity to give informed consent changes then the information collected so far will not be erased and that this information may still be used in the project analysis. ☐

3. I agree to provide evidence to confirm my diagnosis of multiple sclerosis for the purpose of the study (e.g. previous clinical letters) or give consent for a member of my MS care team to be contacted to confirm this. ☐

4. I understand that data collected in the study may be looked at by authorised individuals from the University of Nottingham, the research group and regulatory authorities where it is relevant to my taking part in this study. I give permission for these individuals to have access to these records and to collect, store, analyse and publish information obtained from my participation in this study. I understand that my personal details will be kept confidential. ☐

5. I understand that the interviews will be recorded and that anonymous direct quotes from the interviews may be used in the study reports. ☐

6. I understand that the data collected during the first telephone interview and the information collected about me will be used to support other research in the future, and may be shared anonymously with other researchers. ☐

☐

7. I agree to take part in the above study.

Name of Participant	Date	Signature
Name of Person taking consent	Date	Signature

2 copies: 1 for participant, 1 for the project notes

Appendix I- Participant Information Sheet

Participant Information Sheet
(Final Version 3.0: 19/09/19)

IRAS Project ID: 255380

Title of Study: Seeing the invisible: An exploration of living with, and how people manage, the invisible symptoms of multiple sclerosis.

Names of Researchers: Le-Sharn Parker, Dr Gogem Topcu, Dr Danielle De Boos, Dr Clare Bale, Professor Roshan das Nair.

This study is being undertaken as part of a Doctorate in Clinical Psychology.

We would like to invite you to take part in our research study. Before you decide we would like you to understand why the research is being done and what it would involve for you. One of our team will go through the information sheet with you and answer any questions you have. Talk to others about the study if you wish. Ask us if there is anything that is not clear.

If you would like to take part, please read, sign and date the enclosed consent form, and return to the researcher using the pre-paid, pre-addressed envelope provided. Please respond within one week of the date of this pack arriving. The researcher will contact you by telephone to confirm they have received your consent form. If we don't hear back from you within two weeks, will we contact you to check that you received this pack and whether or not you plan to take part. If you have decided not to take part, this is fine and the researcher will not contact you again after this phone call. Alternatively, if you would not like to be telephoned, please email the main researcher (Le-Sharn) to let them know that you will not be taking part.

What is the purpose of the study?
The purpose of the study is to increase our understanding of how people experience living with the invisible symptoms of Multiple Sclerosis (MS), and how they manage these symptoms. We hope to do this by working collaboratively with those who take part, supporting them take digital images/ photographs that capture their experiences. The purpose is to increase awareness and understanding of the invisible symptoms of MS, and to begin to understand what people might find helpful to manage them. We are interested in finding out more about both the positives and the challenges relating to people's experiences.

Why have I been invited?
You are being invited to take part because you have a diagnosis of MS and may experience invisible symptoms. We are inviting up to 16 participants like you to take part. If you agree to take part in this study, we will ask you to consent to either providing us with a document that confirms your diagnosis (e.g. a clinic letter), or for us to contact your MS professional to confirm your diagnosis.

What are the invisible symptoms of MS?

For this study, the invisible symptoms of MS mean any symptom you experience that you feel is not visible to other people. Some examples that people have given are: fatigue, pain, cognitive problems (such as problems with memory or problem solving), difficulties with mood, bowel and bladder problems, and sensory changes such as tingling or loss of senses.

Do I have to take part?

It is up to you to decide whether or not to take part. If you do decide to take part you are asked to sign the consent form included in your information pack and return this to the researcher using the pre-paid envelope provided. If you decide to take part, you are still free to withdraw at any time and without giving a reason. This would not affect your medical care and legal rights.

What will happen to me if I take part?

Your participation will be over approximately a two-week period and will involve a meeting with the researcher, taking digital photographs that capture your experiences and sending them to the researcher, and being interviewed about the photographs you take. Meetings and interviews in total should take no longer than 3 hours (including phone calls to arrange meetings), and it is up to you how much time you dedicate to taking and sending photos.

Initial Meeting

Once you have returned the consent form, a researcher (Le-Sharn Parker) will contact you to arrange an initial meeting lasting up to one hour. This can take place over skype, or in person either at your home address or at the University of Nottingham. This meeting won't be recorded or used in analysis. It will be used to discuss the study process, and to clarify information and answer any questions you have. You will be shown a number of example photos taken in other studies like this. You will then be asked to spend the next two weeks taking a minimum of five photos that capture your experience of living with and managing the invisible symptoms of your MS.

Phone call

A week after your initial meeting, the researcher will contact you on the telephone to discuss your experience of taking part so far, and to talk about any difficulties if they have come up. This will last up to approximately 30 minutes, and we will ask you permission to audio record this conversation. The audio recording from this phone call will be analysed by a researcher for a different study to this one. They will adhere to the same confidentiality and data protection guidelines as us. At the end of this phone call we will arrange to meet for an interview, and you will be asked to continue taking and sending photos if you have fewer than five.

Photograph Interview

This interview will last for up to one hour and will take place with the researcher either over skype, at your home or at University (Nottingham) premises, depending on your preference. This will be audio recorded and we will ask you for your consent to do this. Prior to the meeting, you will be asked to select five photos you've taken that you feel best capture your experience of invisible symptoms of MS and would most like to talk about in the interview. The researcher will make sure that your chosen photos are available to look at in the interview and will ask you some questions about them. You will not have to speak about any photos you do not wish to.

How will I take and send the photos?

You can take the photos using either your smartphone/tablet camera or digital camera. This will create the electronic image that can then be sent to the researcher via multimedia messaging services (MMS; text message on phone or tablet), email attachment, or private 'WhatsApp' message. These contact details are included at the end of the information sheet. Please feel free to caption each photo or send us a written description if you so wish and send this with the photo. We ask that you send each photo as soon after it is taken, and the researcher will confirm they have received it. We ask that if you have 'free data' on your phone this is used to send the picture via 'Whatsapp'. We ask that you take a minimum of five photos throughout the course of your participation. We would encourage you to use the cheapest option of sending your photo wherever possible.

Who will own the photos I take?

As the creator of the photo you will be the owner. We will therefore ask for your permission to use your photos for the following purposes only: to use in the interview to guide discussion, and to use and publish in our scientific papers. If you would like to be named as the person who took the photos when we publish them, we can do this, but it would mean that people would know that you took part in this study. An alternative would be to use a false name (pseudonym) which we would ask you to choose yourself. You will be able to select photos you are happy to be published. Similarly, if there is anything in the photos where others would be able to identify you from it, these will not be published unless you give additional written consent. We will not publish any photos that identify other people on private land who are not participants in this study. Please see the attached ethical guidelines for taking photographs.

Expenses and payments

You will be given a £10 Amazon gift voucher to thank you for participating in the study, as we realise we will be asking you for your time and input. Travel expenses will be offered for any costs you incur as a result of your participation (e.g. travel to meet with the researcher). If you do not have 'free data' on your phone and choose the send the photos by text (MMS) message, we will reimburse you up to the cost of 12 picture text messages (which we have valued at 50 pence per message). We will not reimburse for more than 12 messages.

What are the possible disadvantages and risks of taking part?

The interview will involve discussing events and experiences in your life that may be sensitive and personal to you. Talking about your experiences of invisible symptoms of MS could be distressing and difficult. You do not have to answer questions if you don’t want to, and you don’t have to give a reason. You will also be able to choose which photos to send to the researcher and choose the photos we talk about in the interview. The researcher will ask your permission first before discussing any others. You can have a break during the interview or can stop it completely if you are feeling distressed or tired. The researcher will support you to access further support should you feel this would be helpful.

If at any point you are feeling distressed in relation to taking the photos, you can contact the researcher (who is also a Trainee Clinical Psychologist) on the details provided to access support. You will also be asked about your wellbeing in relation to the research during the telephone interview so that any issues can be raised then too.

We also realise that the study asks for some of your time, and that it can be difficult to decide what kinds of photos to take. To support you with this, we will bring and discuss some examples with you during our initial visit. Please remember that there is no ‘right’ photograph to take. This could be anything that captures your experience of living with and managing invisible symptoms of MS (in line of course with the ethical guidelines provided for taking photos).

What are the possible benefits of taking part?
We cannot promise the study will help you but the information we get from this study may help us to better understand how people live with and manage invisible symptoms of MS, and what that experience is like for them. We hope that this research will give you and others like you a way to communicate your experiences. This may raise awareness in others (both professional and public) and also help others with MS to feel more understood.

What happens when the research study stops?
The study findings will be written up and submitted as part of a thesis to the Trent Doctorate in Clinical Psychology (University of Nottingham and Lincoln) for a Doctorate qualification. It will also be submitted for publication in a scientific journal. Your information will be anonymised using false names, unless you tell us in writing that you want your name to be included. We will ask you to choose your own false name for the study. Data relating to the study will be held for seven years at the University of Nottingham (and longer if this is required). Information containing your personal details will be held securely for one year, then destroyed. You will receive a summary of the findings at the end of the study and can tell us if you don’t wish to receive this.

What if there is a problem?
If you have a concern about any aspect of this study, you should ask to speak to the researchers who will do their best to answer your questions. The researchers contact details are given at the end of this information sheet. If you remain unhappy and wish to complain formally, you can do this by contacting Patient Advice and Liaison Services for [redacted] on [redacted], or at [redacted].co.uk.

In the event that something does go wrong, and you are harmed during the research and this is due to someone's negligence then you may have grounds for a legal action for compensation against the University of Nottingham but you may have to pay your legal costs

Will my taking part in the study be kept confidential?
We will follow ethical and legal practice and all information about you will be handled in confidence.

If you join the study, we will use information collected from you during the course of the research. This information will be kept **strictly confidential**, stored in a secure and locked office, and on a password protected database at the University of Nottingham. Under UK Data Protection laws the University is the Data Controller (legally responsible for the data security) and the Chief Investigator of this study (named above) is the Data Custodian (manages access to the data). This means we are responsible for looking after your information and using it properly. Your rights to access, change or move your information are limited as we need to manage your information in specific ways to comply with certain laws and for the research to be reliable and accurate. To safeguard your rights we will use the minimum personally – identifiable information possible.

You can find out more about how we use your information and to read our privacy notice at:

https://www.nottingham.ac.uk/utilities/privacy.aspx.

The data collected for the study will be looked at and stored by authorised persons from the University of Nottingham who are organising the research. They may also be looked at by authorised people from regulatory organisations to check that the study is being carried out correctly. All will have a duty of confidentiality to you as a research participant and we will do our best to meet this duty.

Your contact information will be kept by the University of Nottingham for one year after the end of the study so that we are able to contact you about the findings of the study and possible follow-up studies (unless you advise us that you do not wish to be contacted). This information will be kept separately from the research data collected and only those who need to will have access to it. Consent forms which contain personal identifiable information as well all other data (research data) will be kept securely for 7 years, or longer if required. After this time, your data will be disposed of securely. During this time, all precautions will be taken by all those involved to maintain your confidentiality, only members of the research team given permission by the data custodian will have access to your personal data.

In accordance with the University of Nottingham's, the Government's and our funders' policies we may share our research data with researchers in other Universities and organisations, including those in other countries, for research in health and social care. Sharing research data is important to allow peer scrutiny, re-use (and therefore avoiding duplication of research) and to understand the bigger picture in particular areas of research. Data sharing in this way is anonymised (so that you could not be identified) but if we need to share identifiable information, we will seek your consent for this and ensure it is secure. You will be made aware then if the data is to be shared with countries whose data protection laws differ to those of the UK and how we will protect your confidentiality.

If you have any questions or concerns, please don't hesitate to ask. We can be contacted before and after your participation at the email addresses above.

We must also highlight that if you choose to send your photos via 'WhatsApp', they will in part be responsible for the security and protection of these photos. WhatsApp state on their website:

"Your messages should be in your hands. That's why WhatsApp doesn't store your messages on our servers once we deliver them, and end-to-end encryption means that WhatsApp and third parties can't read them anyway… WhatsApp's end-to-end encryption ensures only you and the person you're communicating with can read what is sent, and nobody in between, not even WhatsApp.". For further details, please see www.whatsapp.com/security/

Although what you say in the interview is confidential, should you disclose anything to us which we feel puts you or anyone else at any risk of harm, we may feel it necessary to report this to the appropriate persons. This would mean breaching this confidentiality. We will always seek to discuss this with you first, unless notifying you in advance would increase risk of further harm.

What will happen if I don't want to carry on with the study?

Your participation is voluntary, and you are free to withdraw at any time, without giving any reason, and without your legal rights being affected. If you withdraw, we will no longer collect any information about you or from you, but we will keep the information about you that we have already obtained and this information may still be used in the final study analyses. To safeguard your rights, we will use the minimum personally-identifiable information possible.

What will happen to the results of the research study?

The research study will be written up as part of a Doctoral Thesis in Clinical Psychology.

A summary of the research findings will be sent to you after the study is completed. We intend to have the research published in a journal specific to Multiple Sclerosis/health research.

We intend to publish the results in 'Research Matters' magazine published by MS Society UK (or a similar magazine). We also intend to display the photos as part of an exhibition at public events raising awareness of MS (for example, displaying the photos and possibly their accompanying quotes in an exhibition at the Institute of Mental Health).

You will not be identified in any publication or dissemination unless you have explicitly stated you would like to be credited for the photo. If this is the case we will ask you for additional written consent. We will not publish any photos that identify other people on private land who are not participants in this study.

Who is organising and funding the research?This research is being organised by the University of Nottingham and is being funded by Health Education East Midlands.

Who has reviewed the study?
All research in the NHS is looked at by independent group of people, called a Research Ethics Committee, to protect your interests. This study has been reviewed and given favourable opinion by East Midlands and Derby Research Ethics Committee.

Further information and contact details
If you wish to contact a member of the research team regarding this study, please use the contact details below:

Le-Sharn Parker (Le-Sharn.Parker@nottingham.ac.uk): Trent Doctorate in Clinical Psychology, School of Medicine, Yang Fujia, Jubilee Campus, University of Nottingham. Tel: [redacted].

Under the supervision of

Professor Roshan das Nair (Roshan.dasnair@nottingham.ac.uk): Professor of Clinical Psychology and Neuropsychology, Institute of Mental Health, Nottingham. Tel 0115 8230589

Dr Gogem Topcu (Gogem.Topcu@nottingham.ac.uk): Research fellow, Faculty of medicine and health sciences, Institute of Mental Health, Nottingham. Tel 0115 8231299

Dr Danielle De Boos (Danielle.Deboos@nottingham.ac.uk): Deputy Course director, Trent Doctorate in Clinical Psychology, School of Medicine, Yang Fujia, Jubilee Campus, University of Nottingham. Tel 0115 8466696

Dr Clare Bale [redacted]: Patient and Public Involvement Group member (MS Notts).

Thank you for your time.

Appendix J- Example Images given to participants

Example Photos- Experience of studying for a Doctorate
Final version 1.0:28/04/19

Title of Study: Seeing the invisible: An exploration of living with, and how people manage, the invisible symptoms of multiple sclerosis.

IRAS Project ID: 255380

Name of Researchers: Le-Sharn Parker, Dr Gogem Topcu, Dr Danielle De Boos, Dr Clare Bale, Professor Roshan das Nair.

Below, we have provided a selection of example photos that Le-Sharn has taken to capture some aspects of her experience of studying for a Doctorate in Clinical Psychology. A brief narrative has been included to give an insight into Le-Sharn's thought process when deciding to take these photos, and her comments about what these photos capture for her.
Please remember that these are only examples, you are not expected to replicate them or change your own personal style of how you might approach the process. We are interested to know about *your* personal experience of living with and managing the invisible symptoms of MS. There is no right or wrong way to capture this. As you can see below, some photos are more concrete and some are more metaphorical. Please feel free to be as abstract or as concrete, as creative or as literal as is right for you when taking your photos.

Photo 1: *It often feels like I'm always staring at my laptop, doing the next piece of work, responding to the next email……*
It can get frustrating, sometimes I just want to look at something else.

Photo 2: *The people in my cohort are all so supportive of one another. There's an appreciation that we are all in the 'same boat'. I feel very lucky that I can count them as friends as well as colleagues.*

Photo 3: *Trying to balance my work and life commitments can leave me feeling like I'm trying to keep my head above water. I feel myself slipping under sometimes when things are really busy. I used to go under a lot more, but these days I can catch myself in time.*

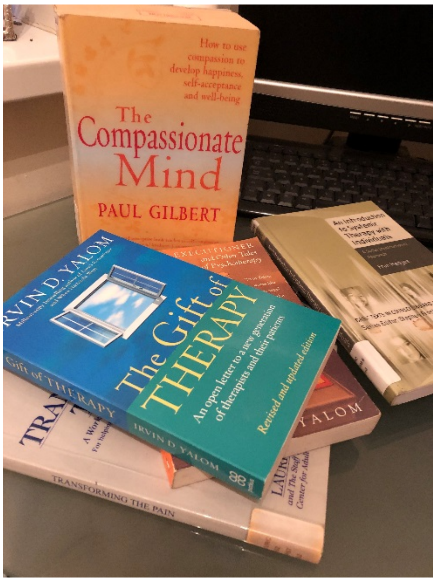


Photo 4: *I've cherished the opportunity to learn about so many of the things I'm interested in and to challenge myself. It's such a thrill to be able to use new learning in my clinical work with people!*

Photo 5: *For me this picture captures the excitement and reward of seeing someone benefit and progress through my clinical work with them.*

Photo 6: *I've learned so much about the importance of self-care this last year or so. I try to do something nice for myself regularly and it helps me to manage the demands of a doctorate and de-stress. It doesn't even have to be fancy. It's the little things*

Photo 7: *The list of deadlines and assessments in my first year felt never ending and daunting. It was such a good feeling to tick each one off as the year progressed. Each one felt like a mini milestone, and I felt proud to get to the end of that list.*

DEADLINES AND EXAMS

Date	Subject	Date	
	Reflective Essay	12/11/17	✓
	PBL Presentation	27/11/17	✓
	PBL Role Play 1	03/12/17	✓
	PBL Role play 2	10/12/17	✓
	PBL Role play 3	20/12/17	✓
	WAIS Passout	21/01/18	✓
	PBL Presentation	14/01/18	✓
	Research Panels	01/02/18	✓
	ICJ Roleplay 1	01/03/18	✓
	Research Protocol	18/03/18	✓
	ICJ Role Play 2	29/03/18	✓
	ICJ Role Play 3	01/04/18	✓
	ICJ Role Play 4	15/04/18	✓
	Case study 1	07/05/18	✓
	Written report	10/06/18	✓
	Systematic lit Review	12/08/18	✓
	Case Study 2	02/09/18	✓

Photo 8: *This was a care package my closest friend brought round for me when she knew I was feeling bogged down by a piece of work I was doing. Although this might not be an ideal package for most, it was deeply personal to me as I love to escape into a good book at the end of a day's work. The support of my family and friends whilst on the doctorate has been invaluable to me.*

Photo 9: *I think it's important to always find something lovely and bright in the middle of something that feels dark and difficult. With the hard work I put in there is so much reward, and I always try to look for the positive on the days that feel harder.*

Appendix K– Ethical Considerations for participants

Ethical Guidelines for Taking Photos
Final version 1.0:28/04/19

Title of Study: Seeing the invisible: An exploration of living with, and how people manage, the invisible symptoms of multiple sclerosis.

IRAS Project ID: 255380

Name of Researchers: Le-Sharn Parker, Dr Gogem Topcu, Dr Danielle De Boos, Dr Clare Bale, Professor Roshan das Nair.

Dear Participant,

Please find below some guidelines to adhere to when taking your photos.

Ethical Guidelines for taking photographs/ digital images

- In the UK, it is generally okay and legal to take photographs/digital images in a public space without asking (except Trafalgar square, Parliament Square, and the royal parks in London).
- You can take photographs of private property without consent as long as you are standing on public land.
- If you are taking a photograph on private property/land you must get the verbal consent of the owner.
- Please respect the privacy and dignity of others when taking a photograph of them.
- Don't take photographs of any illegal activities.
- Please do not compromise your wellbeing or safety to take the photographs, for example putting yourself in a problematic situation in order to get the desired image.
- In private spaces, you must obtain verbal consent from anyone you wish to photograph.
- Ensure that any photos you take are not violating any confidentiality clauses for any individual or organisation.
- Don't take a photograph of anyone who has indicated either verbally or nonverbally that they don't want to be photographed.
- Don't take any photographs that stigmatize, embarrass, or shame individuals or groups.
- Because of the added complexities around gaining consent with children, we ask you not to take images of children.
- The researchers will check with you that you got verbal consent for any pictures you take of others in non-public spaces, and images of others will not be published if they are identifiable from the photograph.

Thank you.

Appendix L- Interview Schedule

Semi-Structured Interview Schedule- Photo-elicitation

Final version 1.0:28/04/19

Title of Study: Seeing the invisible: An exploration of living with, and how people manage, the invisible symptoms of multiple sclerosis.

IRAS Project ID: 255380

Name of Researcher: Le-Sharn Parker, Dr Gogem Topcu, Dr Danielle De Boos, Dr Clare Bale, Professor Roshan das Nair.

All photo-elicitation interviews will be opened and closed with the following statements:

Opening statement: Thank you for agreeing to be interviewed today to discuss the digital photographs you've been taking over the last couple of weeks and have chosen to talk about. Before we start, do you have any questions or concerns? If you'd like to take a break at any point please let me know and we can stop. If you are finding something too distressing to talk about, please let me know. If there are any photos you do not wish to talk about, please let me know. I would like to check again that you are happy for us to audio-record this interview and to use this in our analysis [wait for response- if answer is no then thank for participation and inform them that they can withdraw from the study if they wish to]? Are you able to confirm that verbal consent was gained from any other people where relevant [if no, do not discuss these images in the interview and explain this to the participant]? Are you ready to begin?

What MS symptoms do you or have you experienced that you consider to be invisible?

Closing statement: Thank you very much for your time today. Is there anything we haven't covered that you wanted to? Do you have any questions? How are you feeling after speaking with me today? (if participant indicates distress then offer immediate support, refer and signpost to support as appropriate). Would you be willing to decide today which photos you are happy for us to use when we publish and disseminate this research?

Below are some example questions that may be asked during the interview to elicit discussion. The following schedule may be followed about each of the photos in turn. The researcher may add prompt/ elaborative questions as appropriate.

1. Can you tell me about this photo?
   (prompts may be what context was this taken in/ what was going on when you took it? Can you describe the photo for me? The researcher may point the specific elements of the photo and ask about them.)

2. How do you think this photo captures your experience of living with the invisible symptoms of MS? (prompts may be what was your intention when taking this photo? Why did you take it? Why did you choose to speak about it today?)

3. (If not already covered) Do you think this photo captures your experience of managing the invisible symptoms of MS? If so, how?

4. Why is X (depicting something in the photo) an important aspect of your experiences of invisible symptoms of MS?

5. Is there anything else you'd like to say about this photo and how it relates to your life?

After each image has been discussed in turn.

6. Is there anything else you would like to say about your experiences of living with and managing invisible symptoms of MS that isn't captured in the images you have taken?

The researcher may choose to reflect their interpretations or select out images of interest to them to discuss once those selected by the participant have been discussed.

Appendix M- Consent for photo publication

Consent form for publication/dissemination of specific photos
Final version 1.0: 28/04/19

Title of Study: Seeing the invisible: An exploration of living with, and how people manage, the invisible symptoms of multiple sclerosis.

IRAS Project ID: 255380

Name of Researchers: Le-Sharn Parker, Dr Gogem Topcu, Dr Danielle De Boos, Dr Clare Bale, Professor Roshan das Nair.

Please indicate below the types of publication and dissemination you are happy for us to use your photos for:

	Type of dissemination consented to (Initial box/cell as appropriate)				
Photo Name/Identifier	Publishing in academic journals	Publishing in publicly available articles (e.g. 'research matters' magazine	Sharing in teaching/ conferences	Public domains/spaces (exhibition)	All specified domains

Initial boxes to indicate consent:

I understand that photo(s)……………..……..contain identifiable information about me, and I wish for them to be published, even though this means other people will know I have taken part in the study. ☐

None of the photos stated above contain personally identifiable information. ☐

I wish to be credited by my full/first name (delete as appropriate) name for the photos: ……………………………………………….. in all publication. I understand that a pseudonym (false name) will be used for the photos I don't specify here. ☐

I wish for my full/first (delete as appropriate) name to be used for any of my quotations published in the research. ☐

I understand that I can withdraw my consent for the publication of the above photos up to two weeks after signing this consent form. If I have not withdrawn consent after two weeks, the photos will be used. ☐

Name of participant	**Date**	**Signature**
______________________	____________	__________________
Name of person taking consent	**Date**	**Signature**
______________________	____________	__________________

2 copies: 1 for participant, 1 for the project notes

Appendix N – Participant Debrief

Participant Debrief
Final version 1.0:28/04/19

Title of Study: Seeing the invisible: An exploration of living with, and how people manage, the invisible symptoms of multiple sclerosis.

IRAS Project ID: 255380

Name of Researchers: Le-Sharn Parker, Dr Gogem Topcu, Dr Danielle De Boos, Dr Clare Bale, Professor Roshan das Nair.

Thank you for taking the time to participate in this study. Your contribution is important to us and we hope to further our understanding of people's experiences of living with invisible symptoms of MS based on what you have shared with us. Please accept this £10 Amazon voucher enclosed as an expression of our gratitude.

The interviews you took part in will be used in the analysis and write up of the research, and you will receive a summary of the research findings via email (or post, if you don't have an email address) in April 2020. If you do not wish to be sent a summary, please contact us on the details at the bottom.

If you change your mind about specific images we have discussed using, please do not hesitate to contact me as you can withdraw consent for this up to two weeks from our interview.

If you have any further queries or concerns, or would like support with any aspect of the study then please feel free to contact me or another member of the research team on the details below in the future. Le-Sharn will not be contactable via the mobile number provided to you previously, but will be by email. We have also included some useful contacts for support (some specific to MS) at the bottom of this letter.

Best wishes,

The research team

Further information and contact details

Le-Sharn Parker (Le-Sharn.Parker@nottingham.ac.uk): Trent Doctorate in Clinical Psychology, School of Medicine, Yang Fujia, Jubilee Campus, University of Nottingham.

Under the supervision of

Professor Roshan das Nair (Roshan.dasnair@nottingham.ac.uk): Professor of Clinical Psychology and Neuropsychology, Institute of Mental Health, Nottingham. Tel 0115 8230589

Dr Gogem Topcu (Gogem.Topcu@nottingham.ac.uk): Research fellow, Faculty of medicine and health sciences, Institute of Mental Health, Nottingham. Tel 0115 8231299

Dr Danielle De Boos (Danielle.Deboos@nottingham.ac.uk): Deputy Course director, Trent Doctorate in Clinical Psychology, School of Medicine, Yang Fujia, Jubilee Campus, University of Nottingham. Tel 0115 8466696

Support services and helplines

- MS Society UK (emotional, practical and financial support): 0808 800 8000: www.mssociety.org.uk:helpline@mssociety.org.uk
- MS Trust (links to a range of online and local support groups): 0800 032 3839: www.mstrust.org.uk: infoteam@mstrust.org.uk.
- Samaritans (24 hours a day): 08457 909090: www.samaritans.org
- You may wish to contact your GP or a member of your health team.

Appendix O– Receipt for gift voucher

I can confirm that I have received one £10 Amazon gift voucher as an expression of gratitude for taking part in the study entitled ‘Seeing the invisible: An exploration of living with, and how people manage, the invisible symptoms of Multiple Sclerosis’.

Participant Print name:
Participant Signature:
Signature Date:

Researcher Print name:
Researcher Signature:
Signature Date:

Appendix P- Relevant approvals

Sponsor confirmation:

Research and Innovation
University of Nottingham
East Atrium, Jubilee Conference Centre
Triumph Road
Nottingham
NG8 1DH

Our reference: R&I: 19030
IRAS Project ID: 255380

0115 8467906
sponsor@nottingham.ac.uk

Health Research Authority
Research Ethics Committee

Professor Roshan das Nair
Professor of Clinical Psychology and
Neuropsychology
Room C22,
Institute of Mental Health
Innovation Park, Triumph Road
Nottingham, NG7 2TU

9th May 2019

Dear Sir or Madam,

Sponsorship Statement

Seeing the Invisible: An exploration of living with, and how people manage the invisible symptoms of Multiple Sclerosis

I can confirm that this research proposal has been discussed with the Chief Investigator and agreement to sponsor the research is in place.

An appropriate process of scientific critique has demonstrated that this research proposal is worthwhile and of high scientific quality.*

Any necessary indemnity or insurance arrangements will be in place before this research starts. Arrangements will be in place before the study starts for the research team to access resources and support to deliver the research as proposed.

Wording has been included in the participant information sheets to address the requirements of GDPR for transparency information and has been drafted by the sponsor to ensure consistency and compliance with the University's privacy notice, HRA guidance and the expectations of other organisations, therefore the HRA template wording has not been used verbatim.

Arrangements to allocate responsibilities for the management, monitoring and reporting of the research will be in place before the research starts.

The duties of sponsors set out in the UK Policy Framework for Health and Social Care Research will be undertaken in relation to this research.**

* Not applicable to student research (except doctoral research).
** Not applicable to research outside the scope of the Research Governance Framework.

Yours faithfully

Angela Shone
Head of Research Governance
University of Nottingham

world-changing research
from The University of Nottingham

University Ethics Approval:

University of Nottingham
UK | CHINA | MALAYSIA

DPAP Committee

17/05/2019

Supervisor: Roshan das Nair

Applicant : Le-Sharn Parker

Project: Project Id Seeing the Invisible: An exploration of living with, and how people manage the invisible symptoms of Multiple Sclerosis

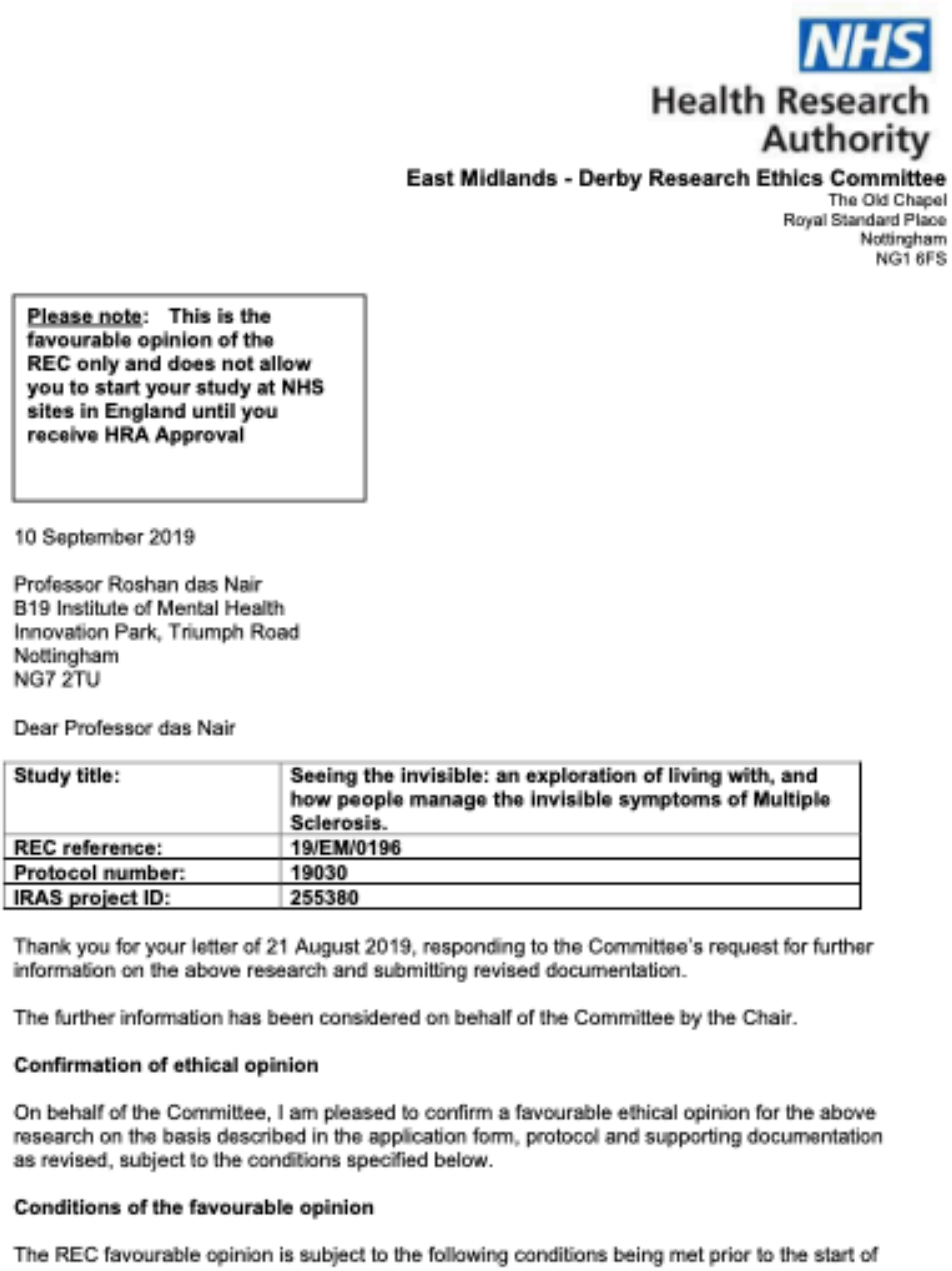
A favourable opinion is given to the above named study on the understanding that the applicants conduct their research as described in the above numbered application. Applicants need to adhere to all conditions under which the ethical approval has been granted and use only materials and documentation that have been approved. If any amendments to the study are required, an amendment should be submitted to the committee for approval. An end of study form will be required once the study is complete.

yours

David Daley

Professor David Daley

REC approval:



Health Research Authority
East Midlands - Derby Research Ethics Committee
The Old Chapel
Royal Standard Place
Nottingham
NG1 6FS

Please note: **This is the favourable opinion of the REC only and does not allow you to start your study at NHS sites in England until you receive HRA Approval**

10 September 2019

Professor Roshan das Nair
B19 Institute of Mental Health
Innovation Park, Triumph Road
Nottingham
NG7 2TU

Dear Professor das Nair

Study title:	**Seeing the invisible: an exploration of living with, and how people manage the invisible symptoms of Multiple Sclerosis.**
REC reference:	**19/EM/0196**
Protocol number:	**19030**
IRAS project ID:	**255380**

Thank you for your letter of 21 August 2019, responding to the Committee's request for further information on the above research and submitting revised documentation.

The further information has been considered on behalf of the Committee by the Chair.

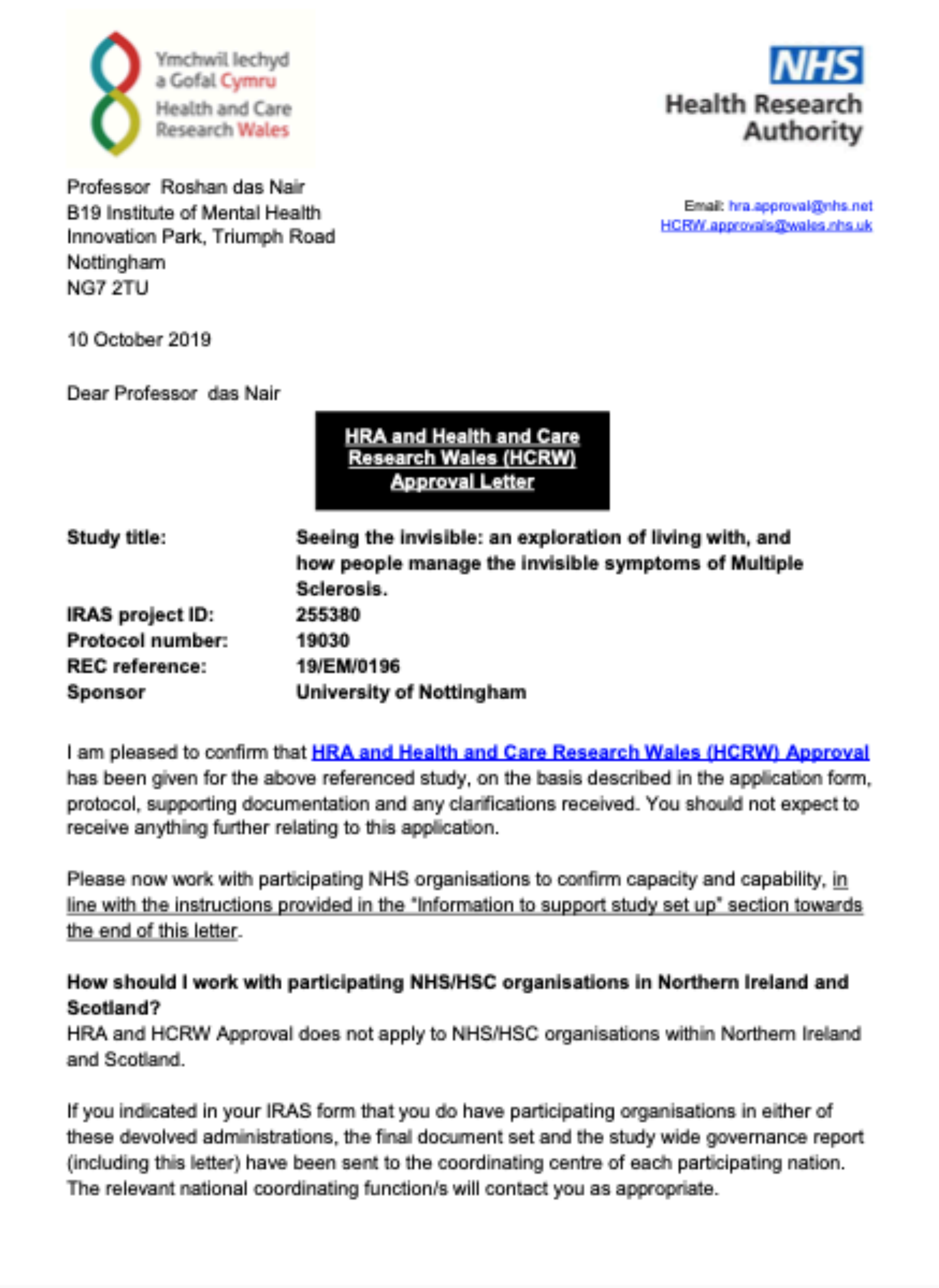
Confirmation of ethical opinion

On behalf of the Committee, I am pleased to confirm a favourable ethical opinion for the above research on the basis described in the application form, protocol and supporting documentation as revised, subject to the conditions specified below.

Conditions of the favourable opinion

The REC favourable opinion is subject to the following conditions being met prior to the start of

HRA Approval:



Ymchwil Iechyd a Gofal Cymru
Health and Care Research Wales

NHS
Health Research Authority

Professor Roshan das Nair
B19 Institute of Mental Health
Innovation Park, Triumph Road
Nottingham
NG7 2TU

Email: hra.approval@nhs.net
HCRW.approvals@wales.nhs.uk

10 October 2019

Dear Professor das Nair

HRA and Health and Care Research Wales (HCRW) Approval Letter

Study title: **Seeing the invisible: an exploration of living with, and how people manage the invisible symptoms of Multiple Sclerosis.**
IRAS project ID: **255380**
Protocol number: **19030**
REC reference: **19/EM/0196**
Sponsor **University of Nottingham**

I am pleased to confirm that HRA and Health and Care Research Wales (HCRW) Approval has been given for the above referenced study, on the basis described in the application form, protocol, supporting documentation and any clarifications received. You should not expect to receive anything further relating to this application.

Please now work with participating NHS organisations to confirm capacity and capability, in line with the instructions provided in the "Information to support study set up" section towards the end of this letter.

How should I work with participating NHS/HSC organisations in Northern Ireland and Scotland?

HRA and HCRW Approval does not apply to NHS/HSC organisations within Northern Ireland and Scotland.

If you indicated in your IRAS form that you do have participating organisations in either of these devolved administrations, the final document set and the study wide governance report (including this letter) have been sent to the coordinating centre of each participating nation. The relevant national coordinating function/s will contact you as appropriate.

Approval from NHS recruitment site:

Via Email sent to sponsor 21/10/19

Dear ***Angela,***

RE: IRAS 255380 Confirmation of Capacity and Capability at [redacted]

R&I Ref: 19NS017
Full Study Title: Seeing the invisible: an exploration of living with, and how people manage the invisible symptoms of Multiple Sclerosis.
Sponsoring Organisation: University of Nottingham

This email confirms that [redacted] has the capacity and capability to deliver the above referenced study. Please find attached our agreed Statement of Activities as confirmation.

We agree to start this study on a date to be agreed when you as sponsor give the green light to begin.

Please be aware this confirmation of capacity is provided on the understanding and provision that you will follow the conditions set out in the attached document ([redacted] R&I of Confirmation of Capacity and Capability Conditions, v2).

The following documents were reviewed:

Document	*Version*	*Date*
Copies of advertisement materials for research participants [STUDY ADVERTISEMENT Living with and managing the invisible symptoms of MS v1.0 28.04.19]	1.0	28 April 2019
Evidence of Sponsor insurance or indemnity (non NHS Sponsors only) [2018 To Whom It May Concern - EL PL PI]	1.0	28 April 2019
HRA Schedule of Events [SCHEDULE OF EVENTS Living with and managing the invisible symptoms of MS v1.0 28.04.19]	1.0	28 April 2019
HRA Statement of Activities [STATEMENT OF ACTIVITIES Living with and managing the invisible symptoms of MS v1.0 28.04.19]	1.0	28 April 2019
Interview schedules or topic guides for participants [INTERVIEW SCHEDULE PHONE CALL Living with and managing the invisible symptoms of MS v1.0 28.04.19]	1.0	28 April 2019
Interview schedules or topic guides for participants [INTERVIEW SCHEDULE PHOTO-ELICITATION Living with and managing the invisible symptoms of MS v1.0 28.04.19]	1.0	28 April 2019

IRAS Application Form [IRAS_Form_18072019]		18 July 2019
Letter from sponsor [19030 Sponsor Letter HRA REC v3.0]	1.0	09 May 2019
Other [REC Responses and amendments]	2.0	21 August 2019
Other [Participant contact details form]	1.0	21 August 2019
Other [REC Responses and amendments]	3.0	19 September 2019
Other [PARTICIPANT DEBRIEF Living with and managing the invisible symptoms of MS v1.0 28.04.19]	1.0	28 April 2019
Other [ETHICAL GUIDELINES Living with and managing the invisible symptoms of MS v1.0 28.04.19]	1.0	28 April 2019
Other [EXAMPLE PHOTOS Living with and managing the invisible symptoms of MS v1.0 28.04.19]	1.0	28 April 2019
Other [ELIGIBILITY SCREENING QUESTIONS Living with and managing the invisible symptoms of MS v1.0 28.04.19]	1.0	28 April 2019
Participant consent form [PARTICIPANT CONSENT FORM Living with and managing the invisible symptoms of MS v1.0 28.04.19]	2.0	16 July 2019
Participant consent form [PUBLICATION CONSENT FORM Living with and managing the invisible symptoms of MS v1.0 28.04.19]	1.0	28 April 2019
Participant information sheet (PIS) [PARTICIPANT INFORMATION Living with and managing the invisible symptoms of MS v1.0 28.04.19]	3.0	19 September 2019
Research protocol or project proposal [PROTOCOL Living with and managing the invisible symptoms of MS v1.0 28.04.19]	2.0	16 July 2019
Summary CV for Chief Investigator (CI) [Staff - CV Roshan das Nair 1802 IRAS (Feb18)]	1.0	28 April 2019

If you wish to discuss further, please do not hesitate to contact me.

Kind regards,
[redacted]

Dr [redacted]
Managing Director Research and Innovation

Appendix Q- Blank Coding Template

Line Number	Transcript	Code(s)
1		
2		
3		
4		

Appendix R – Coding Extract

Note- Emboldened codes are those identified from deductive sweep

283	LSP: Could you tell me about this picture	
284	and why you took it?	
285	P: So this is trying to symbolise like strange	P14P7L285- Metaphor to communicate experience
286	sensations or pains, or sort or stings and	of symptoms
287	there's like different concepts around it. So	P14P7L287- Sensory symptoms are hard to explain
288	whilst the strange sensations I have I	P14P7L287- Symptoms are strange sensations
289	can't…it's really hard to explain them to	
290	someone who hasn't had them. It's not like	P14P7L290- Metaphor to translate symptom into
291	a nettle sting as such but I was trying to	something accessible to others
292	symbolise something that is a sensation	
293	that people can sort of relate to. So I had	P14P7L293- Numbness as invisible symptom
294	numbness around all of my torso which	**P14P1L293- Identification of invisible symptom**
295	was one of the things that made me think	
296	'something's not quite right'. It made me	P14P7L296- Symptoms are a strange sensation
297	feel like…I'd been running and it made me	
298	feel like I hadn't got any trousers on, it was	
299	quite strange. And I had tingling toes and	
300	numbness in my legs at certain times as	
301	well. I had the MS hug but that's another	
302	picture so I won't go into that yet. Just	
303	about strange sensations, really. And then	P14P7L303- Pain with optic neuritis as invisible
304	with the optic neuritis as well, it started as	
305	a dull ache at the back of my eye, and then	
306	I started to lose the vision in the eye. But	P14P7L307- Using common experience to
307	there was a dull ache like a dull ache pain, a	communicate symptom
308	bit like a headache over your eye. And	
309	then, so some of the treatment has pain	P14P8L310- Self-injected medication can be
310	linked to it. So the first medication I was	painful
311	put on to was Rebif which is an injection,	
312	and with that you have to inject yourself…	
313	on various different sites of the body I had	
314	to do, so legs, arms, on the top of your bum	
315	and on your tummy, and they..that's a	
316	very…it's a machine where you sort of	
317	press a button and it jabs you with the	P14P8L318- Metaphor to describe experience of
318	injection. And that is like a bee sting, like a	self-injecting medication
319	very sharp sting. And then you..everywhere	
320	you inject you'd have a reaction to that, so	
321	you'd have like a double the side of a 50p	
322	pieces, like two 50p pieces sort of sized red	
323	mark where you'd injected yourself. And	
324	the sting of that, it's a bit like a bee sting or	
325	like a bit more than a nettle sting. But just	

the injections and having to do that. So whilst it's visible, they're in places that are invisible to other people so you wouldn't necessarily be showing your tummy or the tops or your legs and stuff. But those are feelings that go on for weeks after injecting and you've still got the big red mark sting there. So that's a frustration, you feel like you're doing the right thing because the medication should be helping you but you know, it's something you would be covered up and that people wouldn't be seeing. I've had copaxin as well which is an injection that you have to…similarly inject yourself. And then with Tysabri I have an infusion which is having to go and have a needle put into my vein in my arm, so that's like a sharp sort of like a sting to have the treatment. And just thinking slightly more metaphorically, some of the stings of having MS, some of the annoyances… for me an annoyance I felt and this seems a bit crazy.. because my actual diagnosis, by the time I was diagnosed I'd already thought 'Yes it's MS' because of all the symptoms and having looked on google, I thought 'yeah, it will be'. So when I was diagnosed I was fairly relieved, so it wasn't like a big shocker for me to me diagnosed. The symptoms were shocking to start with but the actual diagnosis wasn't.	P14P8L327- Marks left by injecting medication are generally hidden to others **P14P8L327- People don't see that you've taken medication** P14P8L330- Injecting medication can cause stinging sensation for weeks P14P8L330- Enduring pain of the medication in order to help with symptoms P14P8L333- Taking medication is a frustration P14L8L333- Tension between taking medication and managing side effects **P14P8L336- People don't see that you take medication** P14P8L340- Pain associated with medication treatment P14P8L344- Metaphor to communicate the annoyances of having symptoms and the impact on life P14P8L348- Diagnosis was a relief as it gave an explanation for symptoms P14P9L355- Symptoms were a shock at first

Appendix S – Looking for Themes

Individual typed codes were printed and colour coded by participant, and then group together where codes shared features and conceptual similarities.

Figure S1. Grouping codes conceptually

Poster

Seeing the Invisible: Using Photovoice to explore the invisible symptoms of Multiple Sclerosis

The University of Nottingham
UNITED KINGDOM · CHINA · MALAYSIA

Le-Sharn Parker, Gogem Topcu, Danielle De Boos, Clare Bale, Roshan das Nair
Trent Doctorate in Clinical Psychology & Institute of Mental Health Nottingham

UNIVERSITY OF LINCOLN

Background

People with Multiple Sclerosis (MS) experience invisible symptoms not observable to others[1]. The hidden nature of invisible symptoms presents challenges for those with MS including stigma, invalidation, and hidden needs[2]. Research shows that people find adaptive ways of managing individual invisible MS symptoms[3], but no study has explored how people experience and manage clusters of invisible symptoms.

Objective-To explore how people with MS experience **living with** and **managing** clusters of invisible symptoms in daily life.

Methods

We used a participatory qualitative method: Photovoice[4]. Twelve adults with MS produced digital images to capture their experiences of living with and managing invisible symptoms. Participants provided short written narratives for each image and discussed their images with a researcher in semi-structured interviews. We developed themes from the narratives and interview data using inductive-deductive thematic analysis.

Results

"it can feel like because it's all hidden that it's all just a figment of your imagination."

The challenges of conceptualising invisibility of symptoms

People with MS face difficulties when conceptualising and communicating their invisible symptoms due to these experiences being unobservable, and because it is challenging for them to accurately communicate their abstract experiences through language.

Conflicts of legitimacy

People with MS discussed issues of conflict in the perceived legitimacy of their invisible symptoms, where they struggle to confirm the 'realness' of their symptoms to both themselves and to others, and they feel invalidated as a result.

"Everything is a balancing act"

Navigating Life with Invisible Symptoms

People negotiated their lives with invisible symptoms in dynamic ways, choosing to try to fit their symptoms to their lives or 'make space' for their symptoms depending upon the context- a decision often influenced by their desire for their symptoms to 'stay invisible' or to 'be seen' by others. Flexibility was important for adopting different navigation styles for different situations.

Discussion

People with MS use different ways to navigate their lives with invisible symptoms and the legitimacy conflicts they bring. It is important that clinicians are aware of the challenges created by symptom invisibility and offer appropriate support to people with MS. People with invisible MS symptoms might benefit from interventions that support them to navigate invisible symptoms flexibly both psychologically and behaviourally. Future research might seek to explore how improved psychological flexibility can support people with MS to live with and manage their invisible symptoms.

Conclusions

This study offers a unique exploration of experiences that cannot be captured through words alone, where people with MS invited us to 'see the invisible' they live with and manage every day. Our research highlights the potential importance of supporting people with MS to validate their invisible symptoms and respond flexibly to the issues they present.

References

[1]Werfel, P.B., & Trettin, L. (2020). Working with Clients with Multiple Sclerosis. *Journal of Health Service Psychology, 46*(1),5–12 https://doi.org/10.1007/s42843-019-00001-1

[2]Parker, L-S., Topcu, G., De Boos, D., & das Nair, R. (2020). The notion of "invisibility" in people's experiences of the symptoms of multiple sclerosis: a systematic meta-synthesis. *Disability and Rehabilitation*, 1-15. https://doi.org/10.1080/09638288.2020.1741698. Online ahead of print.

[3]Norton, C., & Chelvanayagam, S. (2010). Bowel problems and coping strategies in people with multiple sclerosis. *British Journal of Nursing, 19*(4), 220-226. https://doi.org/10.12968/bjon.2010.19.4.46783.

[4]Wang, C.C. (1999). Photovoice: a participatory action research strategy applied to women's health. *Journal of women's health, 8*(2), 185-192. https://doi.org/10.1089/jwh.

Small Scale Research Project

Difficulties with mental wellbeing in people with Multiple Sclerosis: an evaluation of patient need and service responses.

Le-Sharn Parker[a], Gogem Topcu[b], Danielle De Boos[c], Nikolaos Evangelou[d], Roshan das Nair[e*]

[a]*Doctorate in Clinical Psychology trainee, Division of Psychiatry and Applied Psychology, School of Medicine, University of Nottingham, Nottingham, England*

[b]*Research Fellow, Division of Psychiatry and Applied Psychology, School of Medicine, University of Nottingham, Nottingham, England*

[c]*Assistant Professor, Research & Academic Tutor, Division of Psychiatry and Applied Psychology, School of Medicine, University of Nottingham, Nottingham, England*

[d]*Clinical Associate Professor and Consultant Neurologist, Faculty of Medicine & Health Sciences, Queen's Medical centre, Nottingham, England.*

[e] *Professor of Clinical Psychology & Neuropsychology, Division of Psychiatry and Applied Psychology, School of Medicine, University of Nottingham, Nottingham, England; Institute of Mental Health, Nottinghamshire Healthcare Trust, Nottingham, England.*

*Prof Roshan das Nair can be contacted using the following:

Email: Roshan.dasNair@nottingham.ac.uk; Tel: +44 (0) 115 82 30589; Address: B19, Institute of Mental Health, Jubilee Campus, University of Nottingham, Nottingham, NG7 2TU, UK

Purpose: A high prevalence of mental health difficulties exist in the Multiple Sclerosis (MS) population. These are often not adequately identified and addressed by services. We investigated how often patients' difficulties with mental wellbeing were recorded in clinic appointments of a UK-based National Health Service (NHS) MS service, and how these problems were addressed by clinicians.

Methods and materials: A quantitative manifest content analysis was undertaken on a sample of MS clinic letters. Clinicians completed a questionnaire to indicate the frequency with which mental wellbeing difficulties are asked about, mentioned, and recorded in their clinics, and their confidence related to managing these processes.

Results: From 295 clinical notes sampled, the number of patients reported by clinicians to have mental wellbeing difficulties was 22 out of 50 (44%); consistent with the literature, however the frequency with which these issues were reported in clinic letters was not. Clinicians addressed mental wellbeing difficulties in 70% of cases and offered a range of responses. They reported that they do not always ask patients about mental wellbeing or record this.

Conclusions: There is patient need within the service in terms of screening for and addressing mental wellbeing. Although the service captures and responds to some of these needs, there are barriers to this. Further research is required to establish the underlying mechanisms that influence asking about, recording, and responding to mental wellbeing difficulties.

Keywords: multiple sclerosis; mental wellbeing; mental health; service evaluation; MS professionals.

Introduction

Background

Multiple Sclerosis (MS) is a chronic inflammatory disease of the central nervous system that develops when an abnormal response of the immune system causes damage to the myelin sheaths protecting nerve fibres [1]. The disruption of impulse transmissions from the brain to the rest of the body leads to cognitive, affective, sensory and motor impairments [2]. MS is estimated to affect 2.3 million people worldwide [3], and 167 in every 100,000 in the United Kingdom [4]. People are usually diagnosed between ages 20 and 50, and the peak age of onset is 30 years [4,5]. Four courses of MS are recognised (clinically isolated syndrome, relapsing-remitting, primary progressive, and secondary progressive) [6].

People with MS experience debilitating symptoms, including fatigue, cognitive impairment, pain, bladder and bowel dysfunction, blurred vision, muscle weakness and spasticity, ataxia, and sensory deficits [1,7,8]. Within 15 years after onset, approximately 20% of people are bedridden, and a further 20% require mobility aids [9].

Historically, the emotional impact of MS and effect that living with the condition can have on mental wellbeing has been poorly understood. In recent years, acknowledgment of these difficulties and their prevalence has grown [10]. Mental health difficulties in MS are associated with reduced quality of life and treatment adherence, increased symptom severity, increased disability progression, and mortality [11-17]. Epidemiological data also suggest that suicide in MS is approximately twice that of the general population, with depression, severity of depression, social isolation, and alcohol abuse being associated with thoughts of suicide [18].

Therefore, the consequences of mental health problems in people with MS are severe but impacts on mental wellbeing can be overlooked or dismissed as an understandable response to the disease, despite evidence that large proportions of people with MS experience clinically significant symptoms related to their mental health [10]. These difficulties often worsen and persist if adequate support is not offered [19]. Phillips et al. found people with MS demonstrated significantly more difficulty regulating their emotional state than controls [20]. Inappropriate or exaggerated affect are also common experiences with MS [21].

Although there is some heterogeneity in prevalence rates for anxiety and depression in people with MS, rates are significantly higher than in the general population [22-26] and the associated burden is also higher [27]. Prevalence rates for depression in people with MS are reported as 15%-47% [22,23,28-30], and 19.3%-54% for anxiety [22, 31-33]. In the largest systematic review to date, pooled mean prevalence of depression was 30.5%, and 22.1% for anxiety [34]. A UK MS Register study [31] found 54.1% and 46.9% of participants with MS scored in the clinical range on the Hospital Anxiety and Depression Scale (HADS) [35] for anxiety and depression respectively. The authors concluded that the mental health needs of people with MS could be better identified and addressed by health professionals. Another systematic review found a 4.3% prevalence rate for psychosis [21] and it is two-to-three times more likely to develop in those with MS compared to the general population [36].

Research suggests that the provision of treatment for depression and anxiety in people with MS has increased [37,38], having previously taken a primary focus on managing physical symptoms [23,39]. Recent research found that the presence of symptoms of anxiety and depression, rather than meeting diagnostic thresholds for these disorders, were predominant factors associated with a perceived need for mental health support in people with MS, [40]. Therefore, research suggests that clinicians should be vigilant and responsive to individual mental health difficulties and psychological distress, even when patients do not meet criteria for psychiatric diagnosis [41].

Explanations for Anxiety and Depression in MS

A number of psychological theories have been proposed to account for the aetiology of mental wellbeing difficulties in the context of MS, specifically with regards to depression and anxiety [42-44]. Theories largely share an emphasis on avoidance of undesirable internal states, unhelpful thinking styles and negative appraisals. The proposed Working Model of Adjustment to Multiple Sclerosis focuses on psychological distress specific to adjustment to MS and its symptoms [45]. The model assumes (as with other longitudinal cognitive behavioural models) that early experiences shape an individual's core beliefs about themselves, others and the world [44]. Core beliefs are thought to influence a person's values, assumptions and behaviours. Dennison et al. suggest receiving a diagnosis, having a symptom relapse, or experiencing disease progression are critical events that can challenge an individual's assumptions and behaviours [45]. This leads to disruptions to the person's emotional equilibrium and creates distress as they attempt to adjust. Dennison et al. highlight that temporary distress is to be expected until the individual adjusts to their new circumstances, develops alternative beliefs/ makes positive reappraisals of existing beliefs and engages in adaptive behaviours and management strategies [45]. If the individual struggles to adjust, engaging in avoidance behaviours and appraising the disease as threatening, this can prolong distress and lead to longer term mental health difficulties. Wilkinson and das Nair's metasynthesis also points towards the role that unpredictability of the disease plays in initiating and maintaining distress [46].

There is growing evidence for the use of CBT-based psychological therapies in improving the mental wellbeing of people with MS, improving adjustment, and reducing symptoms of depression and anxiety [47-50]. This indicates that psychology provision is useful in the support of people with MS, as is practitioners' awareness of effective interventions. It seems important for services and health professionals to be aware of the mental wellbeing needs of their MS patients and how to respond to these needs.

Policy context

The National Institute for Health and Care Excellence (NICE) recommend a tailored and multidisciplinary approach to MS symptom management and rehabilitation, including pharmacological and physiological treatments [51]. NICE guidelines state professionals should be aware that people with MS can present with mental wellbeing difficulties [51]. They stipulate that a wide range of MS symptoms should be regularly assessed, and patients who experience fatigue should undergo assessment of anxiety and depression. Where depression and anxiety are indicated, clinicians are referred to NICE general guidance surrounding interventions for Generalised Anxiety Disorder (GAD) and are directed to NICE guidelines for depression comorbid with chronic physical health problems [52,53].

The service/ service context

The evaluation was undertaken at an NHS Multiple Sclerosis service led by a multidisciplinary team of 15 clinicians, including neurology and neuro-rehabilitation consultants, specialist MS nurses, occupational therapists and specialist physiotherapists.

The outpatient service provides assessment and ongoing MS treatment to over 2,800 patients, delivering this through several MS clinics per week. After diagnosis, patients are typically invited to attend clinic appointments with a specialist from the team every 6-12 months for review, or more frequently depending upon individual care needs. A summary letter is sent to the patient's GP after every clinic appointment.

Rationale for evaluation

Although literature indicates a large proportion of people with MS will experience mental wellbeing difficulties, the service had not routinely recorded or systematically investigated the frequency with which its patients present with these problems. There are no existing service protocols or guidance around how psychological issues should be responded to by professionals in MS clinics. The present service evaluation was undertaken to investigate the level of need for support with mental wellbeing difficulties within the service, the existing processes by which these needs are met, and to identify areas for development.

Aims and hypothesis

We aimed to investigate how often patients' experiences of mental wellbeing difficulties are discussed in MS clinic appointments, and how they are responded to by clinicians. Although the majority of research relates to anxiety and depression, we did not wish to be exclusive of other mental health difficulties, as studies show many people with MS who do not meet diagnostic criteria for a mental illness still express a need for support [40]. We therefore extended the focus of this evaluation to include all mental wellbeing difficulties. We conceptually defined 'mental wellbeing difficulties' as problems relating to how people feel, think, behave and function on a personal and social level, and how they evaluate their lives as a whole that extends beyond just the absence of mental health pathology [54-58]. In its simplest form, a widely accepted definition of mental wellbeing has been defined as 'feeling good and functioning well' [57,p.1].

Based upon existing literature we hypothesised that anxiety and depression would be captured in appointments in 22.1% and 34.5% of cases, respectively [34]. Although we focussed on anxiety and depression to inform our predictions, by broadening our definition to include other types of mental wellbeing difficulties, we expected the frequency with which these mental wellbeing difficulties are mentioned to be equal-to or greater than the noted percentages.

If discrepancies transpired between our expectations of how frequently difficulties with mental wellbeing were identified in clinics and the actual frequency, we also aimed to explore explanations for this.

Materials and methods

Design

The study was split into two phases. The first phase was a quantitative content analysis of a sample of summary letters from the service's MS clinics. Content analysis allows us to make valid and reliable inferences from texts and apply these to their social contexts by examining them systematically through a classification process of coding according to categorical distinctions [59,60]. This approach enabled us to quantify the frequency with which mental wellbeing difficulties were raised in clinics, and how they were responded to, through coding the clinic letters' content. The study was supplemented by questionnaires competed by clinicians in the service, to allow for discussion of information omitted from the summary letters.

The second phase was the aforementioned questionnaire to help highlight any discrepancies between what is captured in the letters and what is experienced by clinicians in the actual appointments, and to provide insight into the mechanisms underpinning any loss of information between the appointment and letter summary. They were also used to help identify factors which may influence how often mental wellbeing difficulties are mentioned and if/how they are responded to in clinics.

Materials

We accessed patient clinic letters via the service's electronic medical notes platform. A coding scheme was developed for coding types of mental wellbeing difficulties that could be mentioned in the letters. This was derived from empirical studies, *a priori* knowledge and theories, and the data itself [60]. The scheme was based upon existing literature around MS and mental wellbeing difficulties, specifically anxiety disorders, depression, and psychosis. As symptom profiles for these diagnoses do not differ between people with MS and the general population [34] we used key words from the International Statistical Classification of Diseases and Related Health Problems criteria to develop codes [61]. Jackson et al. developed and validated a collection of codes used to extract 'symptoms of mental illnesses' from clinical text summaries produced in healthcare settings [62]. Their codes were also incorporated into our coding scheme (see appendix 1 for coding scheme). So as not to exclude data around other mental wellbeing difficulties that may be relevant but not captured in the pre-determined codes, codes were also developed inductively during the process of data extraction.

A coding scheme was not pre-defined for the responses given by clinicians in their letters, as no literature was available to base this upon. Codes were developed inductively and were data-led. A data extraction form (see appendix 2) was generated which was used to record the relevant manifest (overt and quantifiable) content of the letters [63].

For the second phase, a Likert scale questionnaire was developed, related to clinician's experiences in clinics of the frequency with which mental wellbeing difficulties are asked about, mentioned, and recorded, as well as their self-reported confidence in doing so and the value of having support with this.

Procedures

Systematic sampling was used where we identified each MS clinic that ran between 1st and 30th November 2018 (a random sampling frame within a year of study commencement). This sampling frame enabled us to capture patients who were newly diagnosed with MS and those attending follow-up appointments. We excluded bowel and bladder clinics to avoid bias towards non-emotional letter content, as these clinics tended to take a clear focus on physical symptoms. This provided 37 clinics to sample 50 clients from. We sampled from a number of clinics across a period of a month to capture letters written by all clinicians in the service, thus allowing us to draw inferences about the service as a whole rather than individual clinicians. We sampled the first patient seen at each clinic. If two were seen at the same time by different clinicians, we sampled the first patient listed out of the two on the electronic clinic list. One patient was sampled from each of the 37 clinics, and the remaining 13 were taken by sampling the second patient seen in clinics 1-13. Each patient's electronic clinic letters were reviewed from 1st November 2013 to the date the data extraction started (5th July 2019). This was in order to sample letters over a period of time, so they were more representative. Once the content of the resulting 295 clinic letters had been extracted, the content analysis was completed. The clinician questionnaire was given to the attendees of a departmental meeting, and nine clinicians completed this out of a possible 15.

Validity and reliability

It is crucial in content analysis that the analytical constructs that give shape to the coding scheme are valid [59]. To improve the validity of this study, we used existing validated codes [62] and diagnostic criteria [61] to develop the final coding scheme. Validity and reliability are also improved when the coding scheme is applied consistently, and coding rules are followed [59]. The analyst developed coding rules (see appendix 1 and 2), which helped to reduce the influence of coding ambiguity, and increased transparency and reproducibility [64]. The clinician questionnaires were anonymous to minimise demand characteristics that could reduce the validity of the findings [65].

Reliability refers to the extent to which a procedure or study would yield the same results if they were to be replicated or repeated [66]. The inductive portion of the analysis involved idiosyncratic interpretation and subjectivity, which can lead to unreliable findings when multiple interpretations are possible [59]. To minimise these effects, we stated in coding instructions that the coder should be familiar with MS and have knowledge of mental wellbeing difficulties and MS [67].

Ethical issues

This study was considered to be a service evaluation, as we sought to measure an established service, rather than new interventions or changes to patient treatment [68]. Ethical approval was therefore not required, however approvals from the host trust of the service were gained, and the study was conducted in accordance with Helsinki Ethical Principals [69]. All patient data remained confidential and anonymised questionnaires were used.

Analysis

Content Analysis

Each clinic letter (the sampling unit) was subjected to a quantitative manifest inductive-deductive content analysis, following steps outlined by Weber [64]. Coding for mental wellbeing difficulties was approached deductively with pre-determined codes, and also inductively where the coder drew on their own knowledge and was led by the data in forming coding categories. Coding for clinician responses to the mention of mental wellbeing difficulties was inductive so that analysis was grounded in the data. The letters were read sentence by sentence and coded. Sentences that did not mention mental wellbeing difficulties were not entered into the coding frame, and the letter as a whole was coded as not mentioning them. If the content of a sentence fitted more than one coding category, it was split and included in both. Each sentence could be coded for multiple constructs.

The coding scheme was organised in a hierarchical manner. Text was coded into two main categories (the presence or absence of mental wellbeing difficulties). These are mutually exclusive categories and allowed us to answer our research question about the frequency of general mental wellbeing difficulties in the sample. Sub-categories were also developed in the coding scheme, where individual difficulties and symptoms were coded. This allowed us to make comparisons to prevalence figures available in the literature for specific presentations.

A descriptive statistical analysis of the frequency of codes was carried out [70]. This approach was deemed appropriate as it allowed us to make basic inferences about the frequency with which patients discuss mental wellbeing difficulties and identify responses from clinicians. We did not seek to uncover meanings that latent or qualitative analysis would be suitable for. Clinicians were coded by profession.

Clinician questionnaire

Descriptive analysis was used for data collected from the clinician questionnaires, and correlational analysis to identify basic trends only in the data. Due to the small sample size (*n*=9) the study was underpowered, and therefore the use of inferential statistics was not appropriate.

Results

Content analysis

Coding mental wellbeing difficulties

Within the sampling frame 295 clinic letters were extracted for analysis, written between 1st November 2013 and 5th July 2019. The average number of letters per patient was six.

MS Nurses had written 154 (52.2%) of the letters, Consultant Neurologists had written 120 (40.7%), and Registrars had written 21 (7.1%).

The resulting codes from the first part of analysis were arranged into tiers of categories (see figure 43). Tier 1 comprises of codes for specific symptoms found in the clinic letters (e.g. sleep disturbance, low mood). Tier 2 contains the categories that these symptoms fit into based on diagnostic criteria, as well as other categories that were inductive where the data did not fit into a diagnostic category. Tier 3 comprises of two mutually exclusive categories; whether difficulties with mental wellbeing were mentioned, or not mentioned in the letter. For each letter, the relevant text was subjected to a decision scheme (see figure 44). Tier 2 codes encompass Tier 1 codes, and Tier 3 codes encompass both Tier 2 and Tier 1 codes. For example, if low mood was reported, this was coded in Tier 3 as a symptom, then again in Tier 2 as Depression (as this is a symptom of depression according to the coding scheme), and then coded in Tier 1 as mentioning mental wellbeing difficulties. Some letters yielded multiple codes which would be coded into Tier 1 and 2 as appropriate but would only be coded into Tier 3 once. Tables 9, 10 and 11 present the frequencies of each category extracted from the texts.

Table 9. Categories by frequency for Tier 3 of MS clinic letter content analysis

Category	**Frequency of letters falling into category (%)**
Mention of mental wellbeing difficulties	53 (18%)
No mention of mental wellbeing difficulties	242 (82%)

Figure 43. Tiered codes resulting from content analysis of MS clinic letters

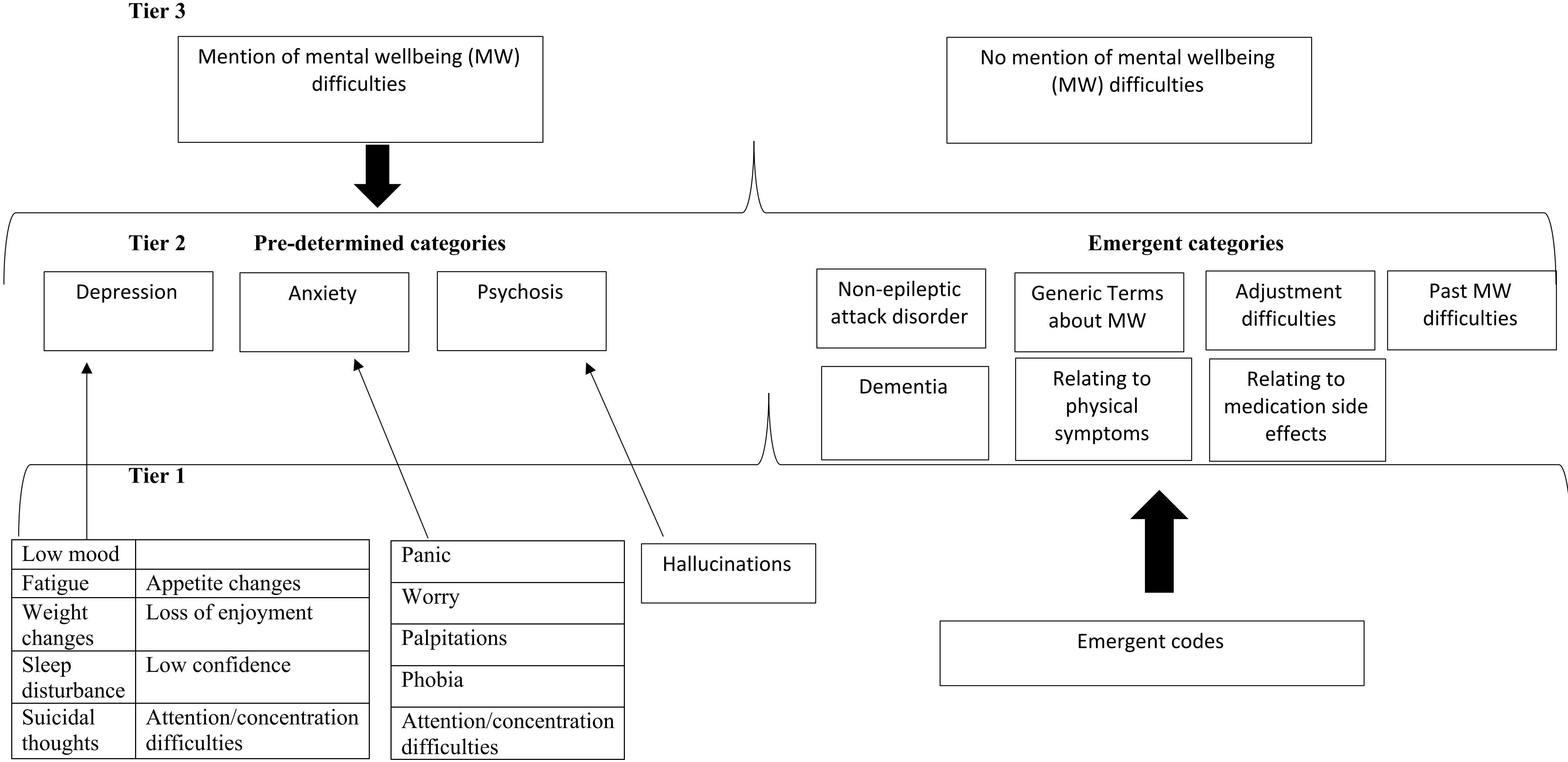
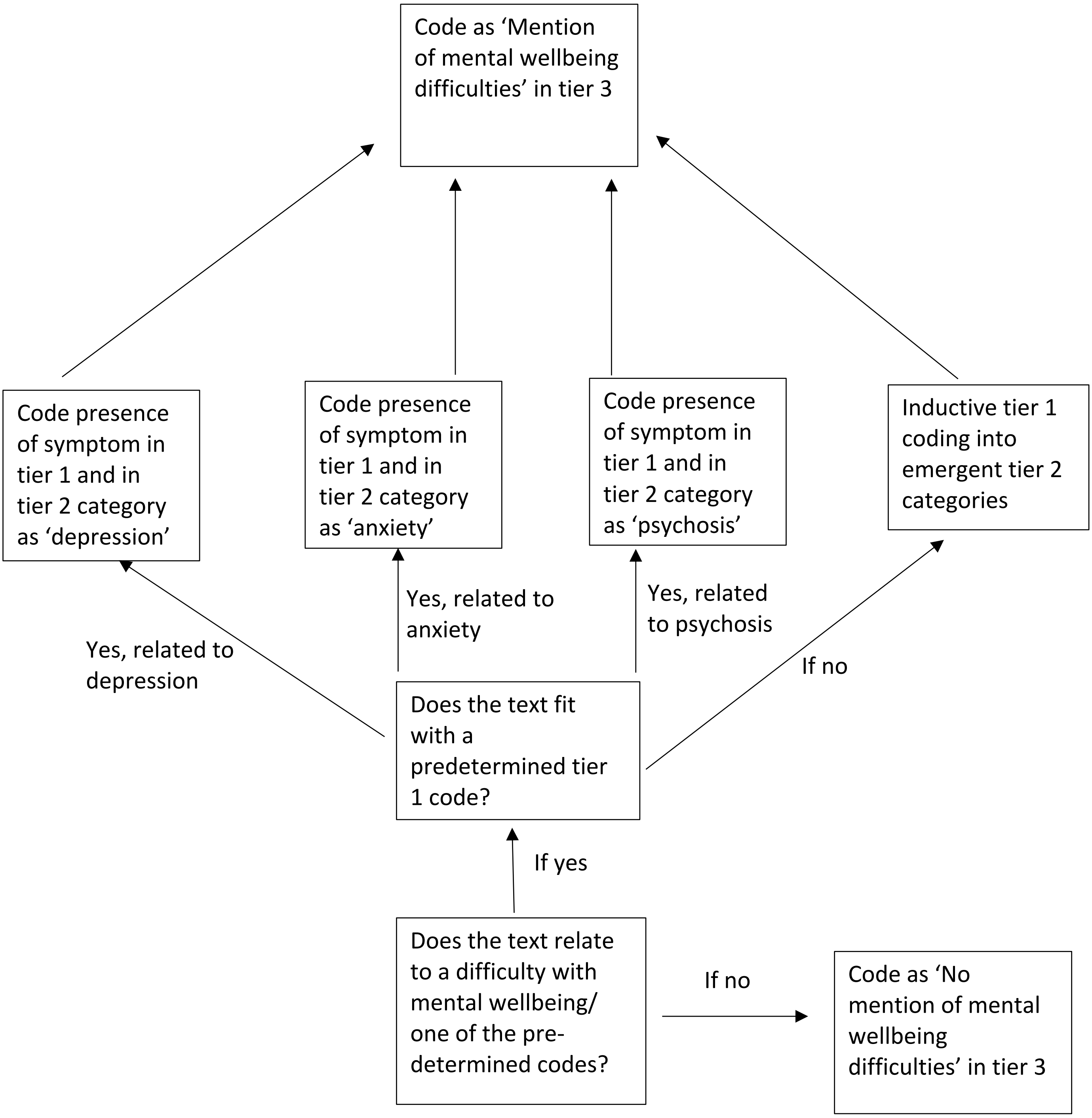


Figure 44. Decision scheme for coding text relating to mental wellbeing difficulties in MS clinic letters



Mental wellbeing difficulties were mentioned in 53 (18%) of the clinic letters. Of the 50 patients sampled, it is recorded that 22 (44%) patients discussed mental wellbeing difficulties at some point in their clinics over the sampling period. For Tier 2 coding, symptoms or statements consistent with depression were recorded in letters more frequently than symptoms of anxiety. The clinic letters also contained text relating to psychosis, dementia and non-epileptic attack disorder.

Table 10. Categories by frequency for Tier 2 of MS clinic letter content analysis

Category	**Frequency of times mentioned across all letters (%)**
Depression	27 (9.2%)
Anxiety	17 (5.8%)
Psychosis	5 (1.7%)
Dementia	1 (0.3%)
Non-epileptic attack disorder	2 (0.7%)
Generic terms for mental wellbeing difficulties	25 (8.5%)
Adjustment difficulties	5 (1.7%)
Past mental wellbeing difficulties	5 (1.7%)
Difficulties relating to physical symptoms	8 (2.7%)
Difficulties relating to medication side effects	3 (1%)

Inductive coding found that the letters reported generic terms for mental wellbeing difficulties, difficulties adjusting to MS and its symptoms, and mental wellbeing difficulties as a direct side effect of medication and/or related specifically to their physical MS symptoms. Past difficulties that were not being actively experienced were also coded.

A number of letters (*n*=10, 3.4%) mentioned patient experiences of symptoms consistent with more than one mental health diagnosis. The percentage of patients reported to experience symptoms of more than one mental health diagnosis in their letters was 18% (*n*=9).

In the Tier 1 analysis, the most commonly reported symptom was fatigue. As fatigue, and attention/concentration difficulties are common symptoms of MS, the data was also analysed with these symptoms removed. When symptoms common as a normal part of MS pathology were removed, the frequency of reported mental wellbeing difficulties reduced to 11.5% of the letters (*n*=34).

Table 11. Categories by frequency for Tier 1 of MS clinic letter content analysis

Category	**Frequency of times mentioned across all letters (%)**
Low Mood	11 (3.7%)
Fatigue	17 (5.8%)
Weight change	1 (0.3%)
Sleep disturbance	3 (1%)
Appetite	1 (0.3%)
Loss of enjoyment	1 (0.3%)
Attention/concentration difficulties	2 (0.7%)
Low confidence	1 (0.3%)
Suicidal feelings	1 (0.3%)
Worry	3 (1%)
Phobia	3 (1%)
Palpitations	1 (0.3%)
Panic	3 (1%)
Hallucinations	2 (0.7%)

Coding clinician responses

Of 53 letters mentioning mental wellbeing difficulties, clinicians offered a response to this in 70% of cases (*n*=37). For cases in which a response was given, these were coded into categories as to the type of response. Table 12 and 13 shows the frequency and type of responses coded. Table 13 shows the frequencies of the main categories but also provides a breakdown of the sub-categories within these. Some letters included more than one response.

Table 12. Categories by frequency for responding to mental wellbeing difficulties in MS clinic letters

Category	**Frequency (%)**
No response when mentioned	16 (30%)
Responded to when mentioned	37 (70%)

Table 13. Categories by frequency for types of responses given by clinicians to mental wellbeing difficulties in MS clinic letters

Category/ *sub-category*	**Frequency (% of overall responses given)**
Referral to health professional/ team	**11 (25.6%)**
Referral to GP	9 (20.9%)
Referral to Crisis team	1 (2.3%)
Referral to specialist team for physical symptoms	1 (2.3%)
Review of current care package	**5 (11.6%)**
Patient already seeing counsellor	4 9.3%)
Patient already under a mental health team	1 (2.3%)
Pharmacological intervention	**7 (16.3%)**
Psychiatric medication requested from GP	1 (2.3%)
Psychiatric medication advised	3 (7%)
Change to physical medication	3 (7%)
Psychological interventions advised	**8 (18.6%)**
Counselling advised	4 (9.3%)
Psychologist advised	1 (2.3%)
Mindfulness course advise	1 (2.3%)
Acceptance and Commitment Therapy advised	1 (2.3%)
Referral to 'Improving Access to Psychological Therapies' (IAPT) advised	1 (2.3%)
Practical strategies advised/offered	**7 (16.3%)**
Unspecified	2 (4.7%)
Cognitive Exercises	1 (2.3%)
Sleep Hygiene	2 (4.7%)
Information about physical symptoms given	1 (2.3%)
Information about psychological symptoms given	1 (2.3%)
Other	**5 (11.6%)**
Entered into clinical trial for 'support' intervention	1 (2.3%)
Request to psychiatrist for mental health diagnosis	1 (2.3%)
Explained difficulties in context of diagnosis	1 (2.3%
Reassurance given	2 (4.7%)
Total number of responses	**43**

Of the cases where a difficulty mentioned was not responded to, five were in relation to past difficulties rather than being actively experienced. Referrals to other health professionals/ teams was the most common response, however clinicians offered a range of responses.

Table 14 shows the frequency with which difficulties with mental wellbeing were mentioned and responded according to profession. The proportion of clinic letters where mental wellbeing difficulties was mentioned was highest for registrars and MS nurses had the highest response rate. For three clinic letters where a registrar did not record a response, the difficulties were related to a past experience rather than an active one. This was also true of two of the MS Nurses' letters.

Table 14. Frequencies of mention of mental wellbeing difficulties and response according to profession in MS clinic letters

Code	Professional	Frequency (n)	As % of own appts
Mention mental wellbeing difficulties	Consultant	28	23.3%
	MS Nurse	17	11%
	Registrar	8	38%
Number of responses given to difficulties	Consultant	19	67.9%
	MS Nurse	13	76.4%
	Registrar	5	62.5%

Clinician questionnaires

We obtained nine responses in total from Consultant Neurologists (*n*=4), MS Nurses (n=4) and a physiotherapist (n=1). The frequency of each answer given in the clinician's questionnaire is presented in table 15. In table 16 we report the median response for each question. This measure of central tendency was chosen as the Likert scales were not assumed to represent constructs that were fixed intervals apart.

Table 15. Frequencies of each clinician response given the questionnaire about patient mental wellbeing difficulties in MS clinic appointments

Question	**Response frequency**				
	Every Appointment	51-99% of my appointments	50% of my appointments	1-49% of my appointments	I never ask
1. How often do you ask patients in MS clinic appointments if they experience any mental wellbeing difficulties?	N=2 (22.2%)	N=3 (33.3%)	N=3 (33.3%)	N=1 (11.1%)	N=0
	Every Appointment	51-99% of my appointments	50% of my appointments	1-49% of my appointments	Never
2. How often do your patients mention experiencing mental wellbeing difficulties in their MS clinic appointments with you?	N=0	N=2 (22.2%)	N=5 (55.5%)	N=2 (22.2%)	N=0
	Strongly agree	Agree	Neither agree nor disagree	Disagree	Strongly disagree
3. To what extent do you agree with the following statement: Patients who attend the MS clinics are generally affected by mental wellbeing difficulties	N=1 (11.1%)	N=8 (88.8%)	N=0	N=0	N=0

Question	Response frequency				
	Every time they are mentioned	51-99% of the time	50% of the time	1-49% of the time	Never recorded
4. When mental wellbeing difficulties are raised or discussed in an MS clinic appointment, how often are these conversations recorded in that patient's clinic summary letters which are sent to their GPs?	N=1 (11.1%)	N=2 (22.2%)	N=4 (44.4%)	N=2 (22.2%)	N=0
	Very confident	Confident	Unsure	Unconfident	Very unconfident
5. How confident do you feel in responding to patient mental wellbeing difficulties directly when they are raised in the MS clinic appointment?	N=0	N=6 (66.6%)	N=3 (33.3%)	N=0	N=0
	Very confident	Confident	Unsure	Unconfident	Very unconfident
6. How confident do you feel in recording discussions around patients' mental wellbeing difficulties in patient clinic letters when they do arise?	N=0	N=7 (77.7%)	N=2 (22.2%)	N=0	N=0

Question	Response frequency				
	Would value very much	Would value somewhat	Unsure	Would not particularly value	Would really not value
7. To what extent would you value having a clear process or guideline for responding to mental wellbeing difficulties when they are raised in MS clinics?	N=4 (44.4%)	N=4 (44.4%)	N=1 (11.1%)	N=0	N=0
	Would value very much	Would value somewhat	Unsure	Would not particularly value	Would really not value
8. To what extent would you value having training around responding to mental wellbeing difficulties when they are raised in MS clinics?	N=6 (66.6%)	N=3 (33.3%)	N=0	N=0	N=0

Table 16. The most common clinician responses to the questionnaire about patient mental wellbeing difficulties in MS clinics

Question	**Median response**
1. How often do you ask patients in MS clinic appointments if they experience any mental wellbeing difficulties?	51-99% of my appointments
2. How often do your patients mention experiencing mental wellbeing difficulties in their MS clinic appointments with you?	50% of my appointments
3. To what extent do you agree with the following statement: Patients who attend the MS clinics are generally affected by mental wellbeing difficulties.	Agree
4. When mental wellbeing difficulties are raised or discussed in an MS clinic appointment, how often are these conversations recorded in that patient's clinic summary letters which are sent to their GPs?	50% of the time
5. How confident do you feel in responding to patient mental wellbeing difficulties directly when they are raised in the MS clinic appointment?	Confident
6. How confident do you feel in recording discussions around patients' mental wellbeing difficulties in patient clinic letters when they do arise?	Confident
7. To what extent would you value having a clear process or guideline for responding to mental wellbeing difficulties when they are raised in MS clinics?	Would value somewhat
8. To what extent would you value having training around responding mental wellbeing difficulties when they are raised in MS clinics?	Would value very much

We investigated relationships and trends within the data. The Shapiro-Wilk test of normality was performed on the data to determine the appropriate statistical tests to use. Due to the small sample size and the fact that not all data was normally distributed, non-parametric tests were most appropriate. The relationship between specific variables was assessed by inspecting scatterplots to check assumptions were met and conducting a series of Spearman's Rho tests.

A non-significant negative correlation was found between clinician confidence in responding to mental wellbeing difficulties and asking patients about mental wellbeing difficulties (r_s= -.24). A significant positive correlation was found between clinician confidence in recording and recording mental wellbeing difficulties when mentioned (r_s= .76 p<.01). A non-significant negative correlation was found between clinician confidence in responding to mental wellbeing difficulties and their value of further training (r_s = -.16). A non-significant negative correlation was found between clinician confidence in responding to mental wellbeing difficulties and their value of guidelines or processes responding to them (r_s= -.10).

Discussion

Frequency of mental wellbeing difficulties

We found that a large proportion of patients with MS were reported in clinic letters to experience mental wellbeing difficulties, and percentages of patients experiencing symptoms relating to depression and anxiety were consistent with existing literature in this area [34]. However, our findings also show reporting of these issues only occurred in a small proportion of the letter sample as a whole, which did not meet our predictions. This may suggest that whilst these difficulties are captured at some point over the course of a patient's care, they are not followed up or monitored in every appointment. Nearly a fifth of patients were reported to experience symptoms related to more than one mental health diagnosis over time.

Adjustment difficulties emerged as a theme from the content analysis. The Working Model of Adjustment [45] suggests that adjustment is a common difficulty in people with MS and struggling with this is indicative of more persistent difficulties such as depression and anxiety if support and intervention is not offered early. The findings highlight a need within the service in terms of support relating to patient mental wellbeing. It is therefore important that clinicians are able to respond appropriately to these needs.

Fatigue was the most commonly reported individual symptom. Whilst a symptom of depression, this is also a common symptom of MS and so can be difficult to disentangle from a mental health difficulty [71]. This could lead to overestimation of depression in the MS population. However, research has shown a strong relationship between depression and fatigue in MS [23,72]. One study found that MS fatigue did not bias the assessment of the somatic symptoms of depression [71]. It is suggested that the relationship between fatigue and mental health is dynamic and multidimensional, and therefore clinicians should assess physical and mental fatigue separately [73,74]. We note that fatigue related to depression may be overlooked as a common MS symptom, and so further assessment of this is needed when raised.

A higher proportion of patients were reported to mention mental wellbeing difficulties with registrars and consultant neurologists in comparison to MS nurses. It is unclear why this is, e.g. due to differences in reporting accuracy, frequency of asking patients about their mental wellbeing across professional groups, or if patients are more likely to mention difficulties with specific profession types. Further research would allow exploration of this finding.

Responses to mental wellbeing difficulties

Clinicians did not record their responses to mental wellbeing difficulties raised in clinics in the clinic letters in almost a third of cases. It is possible that clinician's responded to patients' mental wellbeing difficulties verbally but did not record them in the clinic letter. There seemed also to be instances where difficulties with mental wellbeing were disclosed early in a patient's treatment pathway, and it is possible that the patients were already accessing support by other means. In such events, a clinician might be less likely to focus on or record these difficulties in clinic appointments. Although some of these cases were in relation to a past difficulty, people with MS who have previously experienced mental wellbeing difficulties are more likely to experience them again [75], and therefore may require ongoing monitoring and support. This suggests the service could be missing opportunities to offer support to patients who might need it, as research highlights that patients do not need to be experiencing a constellation of mental health symptoms to express a need for intervention [40,41].

Clinicians offered a range of responses, from practical advice and support to evidence-based psychological approaches [48,76]. This indicates there is some awareness of appropriate interventions for people with MS who are having difficulties with their mental wellbeing. It would be helpful to further explore the factors that influence and govern clinician's decision-making around how to respond to these difficulties.

Clinician perspectives

Clinicians indicated that they record mental wellbeing difficulties when mentioned in clinics in 50% of the resulting letters. This suggests that the content of the clinic letters are not a true representation of the level of need for mental wellbeing support, and indeed how the service responds to these. We must therefore draw conclusions tentatively, as clinician reports suggest our findings may underestimate the prevalence of mental wellbeing problems in their patients. This finding also suggests that there are issues with accuracy of recording clinic appointments within the service. A positive relationship was found between self-reported clinician confidence in recording and self-reported frequency of recording, suggesting that confidence may be a barrier to reporting.

No relationship was found to exist between clinician confidence in responding to mental wellbeing difficulties and asking patients about this in appointments, however, it is apparent from our findings that these issues aren't asked about in every appointment and are sometimes not responded to when raised. Further exploration around barriers and facilitators of asking about mental wellbeing in clinics could help us to understand the mechanisms underpinning these discrepancies.

Implications

As this is a small and underpowered study, the broader implications of our findings must be tentative. A larger, multicentre study focusing on the original aims would be helpful in terms of generalizing our findings and informing wider service development.

There are no specific guidelines within the service for responding to mental wellbeing difficulties, however it appears clinicians engage in some form of decision-making to come to their responses. NICE offer stepped guidelines to support clinicians in decision making around how to respond to symptoms of depression in MS patients [53]. As clinicians indicate they would value further guidance to support them in responding to such difficulties, a study to develop guidelines, templates or prompts for this could be warranted.

As confidence was not highlighted as a possible explanation for why clinicians do not ask each patient about mental wellbeing, it is important to understand other factors involved and generate solutions. For example, if clinicians have a tendency to focus primarily on physical symptoms and forget to ask about mental wellbeing, standardised prompt sheets for clinics could be developed. The service could also consider using validated and reliable measures with their patients to capture psychological health, such as the General Health Questionnaire [77-79].

Clinicians expressed they would value training around responding to mental wellbeing difficulties. Given our findings a training package delivered by a mental health professional should be considered by the service.

Limitations and strengths

Due to the level of data gathered, there are inherent limitations as to the analysis that could be completed. Therefore, we can speak to the findings only in terms of the service that has been evaluated and cannot make wider inferences about our findings or generalisations to other settings or contexts. The correlations only offer us information about general trends in the data rather than allow us to make statistically sound inferences.

The reliance of self-report in our clinician questionnaire may have created a social desirability bias, where clinicians wished to present more favourably in terms of their confidence and reporting.

One researcher coded the clinic letters which presents limitations in terms of assessing reliability of the content analysis [80]. Multiple independent coders would allow inter-coder reliability to be calculated, enhancing the scientific rigour of the analysis [59,81].

Reliability problems in content analysis can also develop as a result of ambiguous word meanings and coding rules [64]. The use of human coders and factors such as fatigue and subtle subjectivity can also affect reliability [64]. The effects of these were minimised by clear coding instructions and word definitions. The coder attended to their reflexive position and was mindful of their own expectations about the data in order to safeguard the quality of the analysis.

Although not without limitations, the study demonstrated some strengths. The use of a coding scheme derived from relevant literature and existing schemes improved validity and helped to ensure structural correspondence between the construct we investigated and the context we placed our findings in [59]. The inductive coding processes involved making conceptually logical decisions based in expert knowledge the coder has as a mental health professional, allowing us to place confidence in the coding scheme used.

Conclusion

Our results indicate that there is a large proportion of patients within the MS service who present with needs relating to mental wellbeing and require appropriate support for this. Whilst the service appears to have captured and responded to some of these difficulties, our findings indicate that there are discrepancies between asking about, recording and responding to mental wellbeing difficulties. We suggested potential barriers to the accurate identification of and response to mental wellbeing difficulties and made recommendations for further research. We suggest the service may benefit from producing clear guidelines for staff around recording and responding to these difficulties and investing in staff training around appropriate responses and interventions.

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Declaration of Interest Statement

The authors report no conflicts of interest.

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Supplemental Information

Appendix 1. Pre-defined coding scheme using diagnostic criteria and validated codes, including definitions [82].

Tier 3 coding scheme

Tier 3 categories (code if tier 2 code present)	
Mention of mental wellbeing difficulties	No mention of mental wellbeing difficulties

Tier 1 and 2 coding schemes

The * symbol indicates definition is quoted directly from APA Psychological Dictionary [81].

	Tier 2 category		
	Depression	**Definition**	**Example variations**
Tier 1 codes relating to Tier 2 category	Irritability	‘A state of excessive, easily provoked anger, annoyance, or impatience.’*	Short temper; ill temper
	Low Mood	Persistent feelings of low spirits and sadness	Depressed mood; low affect
	Loss of enjoyment	‘The inability to enjoy experiences or activities that normally would be pleasurable.’*	Anhedonia; loss of pleasure
	Fatigue	‘A state of tiredness and diminished functioning’.*	Loss of energy; decreased energy
	Reduced concentration	‘Problems in the act of bringing together or focusing, as, for example, bringing one’s thought processes to bear on a central problem or subject.’*	Trouble focussing
	Reduced attention	‘Problems with the state in which cognitive resources are focused on certain aspects of the environment rather than on others and the central nervous system is in a state of readiness to respond to stimuli.’*	Distractibility; inattentiveness
	Reduced self-esteem	‘Reduction in the degree to which the qualities and characteristics contained in one’s self-concept are perceived to be positive.’ *	Poor self-concept; poor self-regard
	Reduced self-confidence	‘Reduction in belief that one is capable of successfully meeting the demands of a task.’*	Not as self-assured
	Feelings of guilt	‘An emotion characterized by a painful appraisal of having done (or thought) something that is wrong and often by a readiness to take action designed to undo or mitigate this wrong.’*	
	Feelings of unworthiness	Feeling lacking in worth, excellence or deservedness	
	Pessimistic views	‘The attitude that things will go wrong and that people’s wishes	Pessimism

		or aims are unlikely to be fulfilled.'*	
	Suicidal Ideation	'Thoughts about or a preoccupation with killing oneself.'*	Suicidal thoughts
	Suicidal acts	'An attempted suicide or similar self-destructive behaviour, especially when the risk of death is low.'* 'The risk of suicide, usually indicated by suicidal ideation or intent, especially as evident in the presence of a well-elaborated suicidal plan.'* 'A situation in which suicide is threatened or attempted.'*	Suicidal gesture; parasuicide; Suicidality
	Disturbed sleep	'A persistent disturbance of typical sleep patterns (including the amount, quality, and timing of sleep) or the chronic occurrence of abnormal events or behaviour during sleep.'*	Insomnia; reduced sleep; increased sleep
	Diminished appetite	Decrease in desire for food/ to eat	
	Decreased emotional reactivity	Reduction in emotional reactions. Experiencing a narrower range of emotions	Blunted affect; flat affect
	Weight loss	Reduction in body mass	
	Loss of libido	'Loss of/ reduction in sexual instincts.'*	
	Feelings of uselessness	Feeling an inability to achieve an intended purpose or desired outcome.	
	Poor motivation	'Reduction in the impetus that gives purpose or direction to behaviour'*	Not goal driven
	Social withdrawal	'Retreat from interpersonal relationships, usually accompanied by an attitude of indifference, detachment, and aloofness.'*	

	Tier 2 category		
	Anxiety	**Definition**	**Example variations**
Tier 1 codes relating to Tier 2 category	OCD (Obsessive Compulsive Disorder)	Code if explicitly mentioned	
	GAD (Generalized Anxiety Disorder)	Code if explicitly mentioned	
	Phobia	‘A persistent and irrational fear of a specific situation, object, or activity (e.g., heights, dogs, water, blood, driving, flying), which is consequently either strenuously avoided or endured with marked distress.’*	Phobic; irrational fear
	Panic	‘A sudden, uncontrollable fear reaction that may involve terror, confusion, and irrational behaviour, precipitated by a perceived threat.’*	
	Palpitations	‘A rapid or irregular heartbeat.’*	
	Chest pain		
	Dizziness		
	Depersonalisation	‘A state of mind in which the self appears unreal. Individuals feel estranged from themselves and usually from the external world, and thoughts and experiences have a distant, dreamlike character.’*	Dissociation (determine closest fit)
	Derealisation	‘A state characterized by a diminished feeling of reality; that is, an alteration in the perception or cognitive characterization of external reality so that it seems strange or unreal (“This can’t be happening”).’*	Dissociation (determine closest fit)
	Nervousness	‘A state of restless tension and emotionality.’*	
	Worry	A state of mental distress or agitation due to concern about an impending or anticipated event, threat, or danger.’*	
	Light-headedness		
	Trembling		

	Muscular Tension	‘A feeling of physical strain accompanied by discomfort.’*	
	Apprehension	‘Uneasiness or dread about an upcoming event or the future generally.’*	
	Difficulty concentrating	‘Problems in the act of bringing together or focusing, as, for example, bringing one’s thought processes to bear on a central problem or subject.’*	
	fidgeting	‘A state of increased motor activity.’*	
	Tension headaches	‘A persistent headache produced by acute or prolonged emotional or physical strain.’*	
	Inability to relax	A’ form of activity that appears purposeless and limited in time or intensity. A human being may constantly move, become distractible, or pace the floor.’*	Restlessness
	Tachycardia	Abnormally increased heart rate	Racing heart
	Tachypnoea	Abnormally rapid breathing.	Heavy breathing; hyperventilation
	Obsessional thoughts	‘A persistent thought, idea, image, or impulse that is experienced as intrusive or inappropriate and results in marked anxiety, distress, or discomfort. ‘*	
	Compulsive acts	‘A type of behaviour (e.g., hand washing, checking) or a mental act (e.g., counting, praying) engaged in to reduce anxiety or distress. Typically, the individual feels driven or compelled to perform the compulsion to reduce the distress associated with an obsession or to prevent a dreaded event or situation.’*	
	Hypochondria	‘Morbid concern with the state of one’s health, characterized by unfounded beliefs of ill health’*	
	Social withdrawal	‘Retreat from interpersonal relationships, usually accompanied by an attitude of indifference, detachment, and aloofness.’*	

	Tier 2 category		
	Psychosis	**Definition**	**Example variations**
Tier 1 codes relating to Tier 2 category	Schizophrenia	Code if explicitly mentioned	
	Thought echo	‘An auditory hallucination in which an individual hears his or her own thoughts repeated in spoken form.’*	
	Thought insertion	‘A delusion in which the individual believes that thoughts have been irresistibly forced into his or her mind and ascribes these thoughts to outside sources.’*.	
	Thought withdrawal	‘The delusion that one’s thoughts are being removed from one’s mind by other people or forces outside oneself.’*	
	Thought broadcasting	‘The delusion that one’s thoughts are being disseminated throughout the environment (e.g., by means of television, radio, or other media) for all to hear.’*	
	Delusions (general)	‘An often highly personal idea or belief system, not endorsed by one’s culture or subculture, that is maintained with conviction in spite of irrationality or evidence to the contrary.’*	
	Hallucinations	‘A false sensory perception that has a compelling sense of reality despite the absence of an external stimulus.’*	Auditory/visual/olfactory/ gustatory/ tactile hallucinations
	Incoherence	‘Inability to express oneself in a clear and orderly manner, most commonly manifested as disjointed and unintelligible speech.’*	
	Breaks in train of thought		
	Neologisms	‘A newly coined nonsensical word or expression.’*	
	Catatonia	‘A state of muscular rigidity or other disturbance of motor behaviour, such	

		as catalepsy, extreme overactivity, or adoption of bizarre postures.'*	
	Social withdrawal	'Retreat from interpersonal relationships, usually accompanied by an attitude of indifference, detachment, and aloofness.'*	
	Paucity of speech	A general lack of additional, unprompted content seen in normal speech.	Poverty of speech
	Thought disorder	'A cognitive disturbance that affects communication, language, or thought content.'*	Derailment of speech, circumstantial speech, poverty of thought
	Blunted emotions	'A disturbance in which emotional responses to situations and events are dulled.'*	Emotional withdrawal, flat affect
	Incongruent emotions	'Lack of consistency or appropriateness, as in inappropriate affect.'*	
	Grandiosity	'An exaggerated sense of one's greatness, importance, or ability.'*	
	Euphoria	'Extreme happiness and an elevated sense of well-being which is exaggerated.'*	Elation; Elevated mood
	Echolalia	'Mechanical repetition of words and phrases uttered by another individual.'*	
	Poor motivation	'Reduction in the impetus that gives purpose or direction to behaviour.'*	
	Pressured speech	'Accelerated and sometimes uncontrolled speech.'*	
	Paranoia	'Experience in which the person develops a persistent, well-systematized, and logically constructed set of persecutory delusions, such as being conspired against, poisoned, or maligned.'*	

Appendix 2. Coding Instructions and Data Extraction form for content analysis of MS clinic letters

Instructions for coders

- Coders should have expert knowledge surrounding mental health and wellbeing and/ or mental wellbeing in Multiple Sclerosis, either through clinical practice or research.
- Use the data extraction form and coding scheme to code the text sentence by sentence.
- Each time a potential code is identified, refer to the decision scheme to code into the appropriate corresponding category.
- If code is not captured in the pre-defined coding scheme, then code inductively and record emergent categories led by the data.
- Some variations and definitions of words and word meanings are provided. Do not be limited by the example variations and use the definitions and your knowledge to determine coding.
- Be mindful that some MS symptoms may overlap with pre-determine codes. Use your discretion around the context in which these are mentioned to inform coding. For example, if a patient experiences trembling as a result of cerebellar damage, you would not code this as ‘anxiety’ despite the coding frame.

Data extraction form complete with examples (italicised codes emerged through an inductive process)

Patient Number	Letter number	Clinician	Relevant content (raw data)	Code (s)	Category (Tier 1, 2 or 3)	Response recorded
34	183	MS Nurse	N/A	N/A	3- No mention	N/A
	184	Consultant Neurologist	Experiencing overwhelming sense of fatigue at times associated with low mood	Fatigue; low Mood	3- Mentioned 2- Depression 1- Fatigue; low mood	Yes; *physical medication reviewed; referred to GP*
35	185	Registrar	She is under increased stress	*Stress*	3- Mention 2- *Generic*	No response
36	186	MS Nurse	He has been experiencing anxiety	Anxiety	3- Mentioned 2- Anxiety	*IAPT recommended*