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Multiple challenges for people after transitioning to secondary progressive multiple sclerosis: a qualitative study

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Abstract

Objectives: Transitioning to secondary progressive multiple sclerosis (SPMS) is demanding for both patients and healthcare professionals. The particular challenges and the ways patients cope are poorly understood. The present study examines what challenges people face when diagnosed with SPMS, by exploring experiences of people who have transitioned recently (up to 5 years).

Design: Semi-structured qualitative interviews at two time points a year apart. Interviews were analysed using inductive thematic analysis.

Setting: United Kingdom

Participants: We interviewed twenty-one people at baseline and seventeen participated in the follow-up interviews.

Results: The majority of participants reported expecting to transition to SPMS, and the diagnosis did not make much difference to them. Participants described increasing emotional and physical challenges after transitioning to SPMS and between the first and second interview. Planning, using distractions and maintaining social roles helped participants cope with the increased challenges. The same coping strategies were used between the two interviews. Participants felt there was not much left to do regarding the management of their symptoms. A key theme was the sense of abandonment from health care services after transitioning to SPMS.

Conclusions: After transitioning to SPMS people are faced with multiple challenges. Participants described a lack of directions for symptoms management and lack of support from the healthcare system. An integrated multidisciplinary health care approach is crucial at

the progressive stage of the disease to alleviate feelings of helplessness and promote symptom management.

Strength and limitations of this study

- The present study is the first to use a longitudinal qualitative design to capture people's experience after transitioning to secondary progressive multiple sclerosis.
- The focus on people recently transitioned helped to narrow down people's experiences early on in diagnosis.
- We cannot determine whether the challenges described in this study are linked to having received an official diagnosis or to the challenges faced by increased disability, as we did not interview people just before being diagnosed and straight after.

Multiple Sclerosis (MS) affects more than 2.5 million people worldwide, including 127,000 in the United Kingdom [1]. MS is a condition that presents unique challenges for the individuals, as it usually starts with minimal physical and cognitive dysfunction which progresses over time. There are three types of MS. The most common type is relapsing-remitting (RR, 85%-90% of MS cases) which is characterised by unpredictable attacks (relapses) of new or increasing neurological symptoms that are followed by periods of partial or complete recovery (remissions) [2]. The secondary progressive (SP) course develops in approximately 82% of those with RRMS by 20 years of onset [3]. Finally, a minority of people (10%) will be diagnosed with the primary progressive (PP) course that involves a steady disability increase without attacks [4].

The transition to secondary progressive multiple sclerosis (SPMS) is an expected part of the disease trajectory. Epidemiological studies indicate nearly 10% of people with RRMS reach the SPMS stage after five years, which increases to 25% at ten years and 75% at 30 years [5]. A mean of 2.9 $\pm$ 0.8 years is a typical length of time when people are faced with the uncertainty of whether or not they have transitioned to SPMS [6]. The delay in the diagnosis is thought to be due to the subtle nature of early progression and absent of evidence-based treatment for SPMS [6]. People with SPMS are older, are less likely to be employed, and a lower proportion is female compare to people with RRMS [7]. When people transition to SPMS, they are likely to experience more severe neurological symptoms, more frequent hospitalisation [8], have more frequent and pronounced cognitive deficits [9, 10], and higher levels of fatigue [11]. The onset of SPMS has also been associated with fear, low mood [12], greater distress [13, 14], lower quality of life [15], and higher levels of depression and anxiety [16] than in both relapsing-remitting and primary progressive types of MS. Only three qualitative studies have been conducted to examine the experiences of people with SPMS. In

two of these studies, women with SPMS recognised the impact of the loss of meaningful activities on the sense of self [17, 18]. A recent qualitative study on patients' experiences while transitioning to SPMS showed that some people learned they had SPMS without any advanced knowledge or understanding of what SPMS meant for them, which sometimes caused confusion and upset [19].

We know little about people's experiences after transitioning to SPMS, and psychological and behavioural interventions aimed at people with SPMS are scarce. Further, a recent meta-analysis on the effectiveness of interventions for treating depression in MS identified 13 papers, with 8 of these excluding people with progressive MS from participating [20]. When people with SPMS are included in psychological interventions, they also seem to gain fewer benefits compared to people with RRMS [21]. The psychological challenges that people with SPMS face may, therefore, differ to the ones that people with RRMS face

In this study, we aimed to explore the experiences of people who have recently transitioned from RRMS to SPMS. Knowing the challenges that patients face at this stage of their MS trajectory makes it easier to identify information needs and tailor support interventions. Further, it is not well understood how transitioning to SPMS contributes to psychological challenges and adjustment attempts. The present study is the first to use a longitudinal qualitative design to capture people's experience shortly (within five years) after transitioning. The follow-up interviews allowed respondents to reflect on the changes (or lack of them) since the first interview. The focus on people recently transitioned helped to narrow down people's experiences early on in diagnosis.

Patients and public involvement

This research was part of a larger research project, and two individuals with SPMS are part of

our user involvement committee, and they have helped with different aspects of the project from the initial conceptualisation of the study, feedback on the grant application and feedback of the topic guide. They were also asked to assess the burden of participation in the study. The two people with SPMS gave us feedback on an initial version of our themes. They inquired about whether the experiences differ based on the age of diagnosis or whether the people who described more psychological challenges had also reported cognitive difficulties. Following this, we checked the emerging themes to identify any patterns or differences in themes of people with different demographic characteristics. The two people with SPMS also suggested exploring further the experience of diagnosis and the importance of receiving the diagnosis of a SPMS from a healthcare professional. Finally, they commented on the clarity of description and presentation of the results, for example, they made some suggestions about how themes linked together and asked for a few additional quotes at places to better illustrate themes.

Participants and methods

This research is part of an MS Society UK funded junior research fellowship with a view to increase understanding of psychological challenges and adjustment of people with progressive MS. The current study was approved by the West of Scotland Research Ethics Committee (14/WS/0077). We recruited participants through online adverts on the MS Society UK website, adverts after speaking engagements at MS Society events and through study's information sheets provided by research nurses at MS hospital clinics (Northamptonshire Healthcare NHS Foundation Trust and Cumbria Partnership NHS Foundation Trust). Participants with primary progressive and secondary progressive MS were recruited, but for the aims of this study we focused on the interviews of people with

secondary progressive MS. Eighty-seven people with primary and secondary progressive MS expressed interest in participating in the study. Fifteen (17%) did not meet the inclusion criterion of diagnosis within five years of progressive MS. The remaining 72 completed a short screening questionnaire that included demographic questions, as well as questions about MS duration, symptoms and walking ability. We chose to interview people using maximum variation [22], i.e. sampling to represent different demographic characteristics. Interviewing ceased once data saturation was reached, defined as the point at which no new information or themes are observed in the data [23]. We interviewd 21 participants who were diagnosed with secondary progressive MS and had given informed consent to take part in the study.

As shown in Table 1, participants were between 40 and 77 years of age (mean: 57.38 years). More than half the sample was unemployed or retired due to MS (n=14, 66.66%) and lived with their partners (n=15, 71.43%). Seventeen participants were interviewed for the second time, four participants (19%) did not respond to the email invitations for a second interview.

Interviews

Participants were given the option of face-to-face or telephone interviews. In the first round, all participants opted for telephone interviews, and the interviews lasted between 26 minutes and 160 minutes (mean=79 minutes). The second set of interviews tended to be slightly shorter as they focused on changes in between interviews and lasted between 36 minutes and 103 minutes (mean=66.12). With one exception, participants in the second round also opted for telephone interviews. Table 2 shows the topic guide used for the two interviews. Examples people gave in the first interviews were also used as prompts to assess changes in emotions, thoughts or symptom-management. All interviews were audiotaped with consent and transcribed verbatim.

Data analysis

We used inductive thematic analysis [24]. An inductive approach means the themes identified emerge from the data themselves [25]. Therefore, the inductive analysis is the process of coding the data without trying to fit it into a pre-existing coding frame, or the researcher's analytic preconceptions. This form of thematic analysis is, therefore, data-driven [24].

Coding was undertaken by the first author (AB) under the supervision of the second author (MM) and with discussion of emerging themes with the third author (RMM). A variety of techniques were employed to increase familiarisation with the data. Audiotapes of each interview were listened repeatedly, and transcripts were read and reread. We used NVivo 10 for coding. The analysis of the transcripts was conducted in parallel with data collection. First, each coding unit in the first transcript was given a code name, using vocabulary as close as possible to that used by participants themselves [26]. Initial codes were then applied systematically to the entire dataset, and new codes developed and refined as appropriate. We also grouped participants' interviews that shared similar demographic characteristics and explored patterns or commonalities. We examined whether themes differed across sub-groups (e.g. method of recruitment, gender, and family circumstances). Figure 1 shows the steps followed during the analysis. The analysis was an iterative process and at times we had to go back and forth between steps. In order to document the analysis process, a detailed paper trail was kept with the notes taken after the interviews, the development of the codes and relationship between the raw data and the refined categories and codes. The quotes presented in the result section were chosen for typicality in illustrating the themes. Pseudonyms are used for the quotes.

Results

Two overarching themes about (1) Transitioning to SPMS and challenges and (2) Adaptive strategies over time. The first theme illustrated challenges involved in the transition and the second theme related to how participants responded to those challenges. Tables 3 provides examples of SPMS transition and challenges over time and Table 4 provides examples of self-management and changes over time.

Transitioning to SPMS and challenges

Initial reaction to the transition

For the majority of the participants the transition to SPMS was expected rather than shocking. For some, transitioning did not make much difference because at that point they had already lost essential relationships and activities they valued. For others transitioning to SPMS did not make much difference because the progressive worsening of their symptoms was slow and subtle.

We compared the interviews of people that found about the SPMS diagnosis in different ways, for example, accidentally when visiting the hospital for other issues, or in one of the annual consultations or they suspected they had transitioned and asked the neurologist for confirmation. In contrast to the majority, the five participants, who found out about the transition accidentally described being upset and shocked. These five people had MS between 3 to 21 years (median=12 years).

> *[Neurologist's name] started other MRI's and tests and things and said that I'd progressed to secondary progressive. So, I was shocked and- I was shocked that I got signed off work, and it progressed. I thought he'd just say, "You're signed off for a*

couple of weeks", not that and, I just thought, you know, "If I sleep then I'll be all right." (female, 45, T1)

Worsening of symptoms and emotional shifts

Transitioning to SPMS also meant worsening of symptoms, and people described progressive worsening of symptoms including mobility, fatigue, vision, and bladder dysfunction, urinary tract infections and falls. There were further worsening of symptoms reported between the first and second interview. This deterioration also brought more changes in participants' lives.

Transitioning to SPMS also meant worsening of emotional issues. A few participants had a diagnosis of depression before or after the initial diagnosis of MS, which made managing MS more difficult. However, the majority of participants described dealing with other difficult emotions in both interviews, including feeling like a burden, being afraid of having accidents, being trapped or feeling embarrassed. They also expressed low self-esteem and confidence, and some participants described the uncertainty of whether they have the skills to cope with MS, accept the condition and deal with stress.

I had a bad fall last week where I split all my head open, so I am feeling a bit lack of confidence in just going out for a little walk up the road and back with my walker on my own. (female, 54, T2)

Overall, in the second interview participants talked in more depth about their psychological difficulties and challenges in coping over the years and presented a more severe picture than in the first round of interviews. Further, people who described emotional challenges in the first interview described the same challenges in the second interview, a year later.

Arriving at the point of no help

After transitioning to SPMS, participants reported that they did not feel health services had anything more to offer since they could no longer have access to disease-modifying drugs. They saw their transition as a point beyond help.

> *I have an MS nurse but quite frankly you know I have come to an end what you can do, I don't feel I am getting any support at all at the moment. I feel I am being left to my own devices. Yes I see him every six months, every twelve months but there is nothing he can offer so you know it is a bit of a waste of time really. (female, 54, T2)*

When asked whether there is any support that they would like to receive in the future, one participant said: '*I think there is quite a lot of support when you're first diagnosed but I think it tails off I think people find it more difficult to support you the longer you have the MS I think people become quite cynical about the help that can be got and cynical about what the NHS has to offer and cynical about the help that can be obtained as to whether it's any use or not.*' (female, 65, T2). Most participants shared a similar opinion.

Adaptive tasks and changes over time

Planning activities or scaling down activities

Participants reported adjusting their holiday destinations and plans, to take into account their limitations, without changing their previous lifestyle too much. Sometimes, across the two interviews participants appeared to overestimate their limitations and underestimate their ability to cope in the future. This discrepancy is illustrated below by the quotes of the same participant in the first and second interview.

I don't go too far now, I can only cope with about three and half hours on the plane at the most. I can't go long distance or anything like that, you know long haul anymore, it's usually the Canary Islands or somewhere like that, somewhere nice and close (female, 54, T1)

I am still determined that I am going to travel and see as many places as I want. We went away for six weeks over January/February, we went to New Zealand and Australia (female, 55, T2)

However, at times, participants found it difficult to plan or modify their activities and instead scaled back, especially when the condition progressed rapidly, and almost all participants who had mentioned difficulties finding alternative activities in the first interview had the same difficulties in the second interview.

I can't read the same that I used to; I can't go on the computer; I can't really watch TV; gardening, the high impact exercises, you know, the long walk so everything's just had to be tailored down and you know brought into a shorter time period. The human is pretty good at finding ways around things…so yeah you just adapt um it's a struggle you know there's no two ways about it but it is that life is definitely a struggle but again you get used to that really as well (female, 59, T2)

Emotional regulation

Strategies to manage emotional difficulties included distraction such as playing video games, reading books, going out for a meal, or avoiding thinking and talking about the future, avoiding MS group meetings and spending the day sleeping. Some people tried to be positive by repeating 'things could have been worse'. People reported using the same strategies

between the two interviews. A few participants talked about how they would have liked access to psychological services.

> *It would be nice to talk about how you feel about it, would be it'd be nice if…do you know it would be nice if just to have somebody there saying you're doing a grand job [Laugh] with how you're doing. Because you are isolated (female, 53, T2)*

However, two participants said that they were offered psychological support, but they thought this was unnecessary.

Being social, fitting in and being of use

For most of the participants transitioning to SPMS also meant increased mobility problems and use of a wheelchair. Use of a wheelchair also posed accessibility issues leading to more carefully planned outings. Even though going out came with more difficulties, and at times people felt it was easier to stay in, most talked about the importance of being with friends and family and retaining relationships and also meeting new people. Participants described getting involved in charity work and helping others. They reported that this gave them a new purpose, they learned new things and met people. Participants who talked about the importance of being social or helping others in the first interview also talked about it in their second interview, as illustrated below by the same participant at the two time points.

> …going out with friends, going out for dinner, going and mixing with other people, which I would of quite enjoyed before and not thought anything about it, but now I think, "oh do I really want to go?" but then I force myself, you have got to go, because then when I do go, I do enjoy it and I do feel better because your mood's lifted and once you are moving around, things are easier. (female, 59, T1)

> We've got the local MS branch, we meet twice a month, first and third Tuesdays of the month. I find that very helpful. I do find that helpful to talk to other people that have got the same sorts of problems that you've got, gone through the same things. And again there's somebody there that, it's not perhaps a close friend, but has become or is becoming more of a close friend that we will see one another outside of the MS branch. (female, 59, T2)

Discussion

Reflecting on the SPMS diagnosis, people overall did not express trauma or distress, only on the occasions, the diagnosis came as a surprise. People who have recently transitioned to SPMS reported facing a worsening of the condition alongside increased emotional challenges. In the second interview, most people described the increased severity of their symptoms and more emotional difficulties. Participants reported detailed planning, distractions and remaining as active and as involved in the community as possible helped to cope with the condition. Some participants overestimated their limitations and underestimated their capacity to cope between the first and the second interview but overall most reported using similar coping strategies between the two time points. A common theme across all interviews was a sense of abandonment from the health care services and the sense that nothing can be done at this stage of their condition. Therefore, providing appropriate support after the transition diagnosis is vital.

A qualitative study of people recently diagnosed with MS showed that psychological well-being was described as precariously contingent on reasonable current and future health status [27], for example, people were not sure whether they would be able to cope if they lost their job or had to use a wheelchair. The current study examines what happens after

people have moved to SPMS. Participants in our study talked about increased disability since they have transitioned to SPMS or between the first and second interview, but despite these increased difficulties, people felt that health care professionals had nothing more to offer and there was nothing more they could do to control their emotional or physical symptoms. Focusing on well-being is often on diagnosable depression and anxiety. However, participants in the present study described having to deal with challenging emotions which may not reflect in these clinical diagnoses.

An essential finding of this study is that people who described some challenges with self-management in the first interview described the same challenges in the second interview, a year later. This can be linked to their perception of SPMS as the stage where nothing further can be done to manage things. According to the Common Sense Model [28]. Being diagnosed with an illness that is perceived as more controllable will lead to less distress and more control efforts such as seeking advice on symptom management. Participants interviewed here described SPMS as a condition they and their health care team did not have any control over. As people move from RRMS to SPMS, the capacity of personal and treatment control declines, and creates a significant challenge in adapting to the change by identifying areas that they can still have control while letting go of unattainable goals [29].

Participants in the current study reported that there is nothing they or health care professionals can do to manage their increasing limitations. Even though self-management interventions in progressive MS are scarce, there is some preliminary evidence of interventions that could help with symptom management. For example, endurance training can improve walking [30]; progressive resistance training may improve lower limb strength [31]; bodyweight-supported treadmill can reduce pain [32], and mindfulness training can

reduce anxiety, depression and impact of MS [33]. Where available, health care professionals should signpost patients to appropriate services that can help with MS symptoms but more needs to be done to find ways of developing these services, so they are more readily available.

The present study has certain significant strengths. First, to our knowledge, it is the first study attempting to investigate challenges and adaptive tasks shortly after people have transitioned to SPMS. The particular advantage of the present study was its longitudinal approach that allowed the identification of patterns over time. This showed that whereas more severe emotional and physical symptoms were reported in the second interview, these did not result in different or more challenges. While this may suggest particip ants feeling more comfortable in opening up at time two having built a report with the interviewer, it may also reflect the lack of support people were receiving. Finally, a strength of this study also lies in the specificity of the sample selected, who were all people who had transitioned to SPMS within the previous five years.

The study has certain limitations. Firstly, the qualitative exploratory nature of the study does not allow for causal relationships to be established. Second, getting a SPMS diagnosis may take years, in part due to substantial diagnostic uncertainty as a result of the subtlety of signs of early progressive disease [6]. Therefore, it is hard to determine whether the challenges described are linked to having received an official diagnosis or to the challenges faced by increased disability, as we did not interview people just before being diagnosed and straight after.

Regardless of the above limitations, the findings presenting here underline the additional physical and emotional challenges people face when transitioned to SPMS and that people try to adapt by planning, finding distractions and remaining social but they reported there

was nothing further to do regarding symptom management. Identifying physical and emotional challenges at the time of the diagnosis to SPMS will help referral to appropriate services within a multidisciplinary clinical team. Offering people options for continuing symptoms management at the time of SPMS diagnosis may help with feelings of helplessness accompany the diagnosis.

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Conflict of interest

We have no conflict of interest to declare.

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Author statement

AB and RMM designed the study. AB conducted the interviews, led the data analysis and drafted the manuscript. MM contributed to the development of the topic guide, oversaw data collection and analysis and contributed to drafts of the paper. RMM advised on the conduct of the research, contributed towards refining themes, interpretations of the findings, and drafts of the paper.

Data sharing statement

Paper trail and NVivo coding files available from the corresponding author

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Transition to secondary progressive MS

Table 1. Participants' characteristics

Participants' characteristics (T1)	*Number (%)*
Age (Mean, SD)	*57.33 (9.13)*
Gender-female	*16 (72.7%)*
Marital status (N, %):	
Married/cohabiting	*15 (71.4%)*
Single	*3 (14.3%)*
Divorced/separated	*3 (14.3%)*
Time since initial diagnosis:	
0-5 years	*5 (23.8 %)*
6-10 years	*3 (14.3%)*
11-15 years	*3(14.3%)*
16-20 years	*2 (9.5%)*
21years +	*8 (38.1%)*
Time since SPMS diagnosis:	
1 year	*4 (18.2%)*
2 years	*3 (13.6%)*
3 years	*3 (13.6%)*
4 years	*4 (18.2%)*
5 years	*7 (31.8%)*
Walking ability (with aid):	
0-5 meters	*9 (42.8%)*
20 meters	*3 (14.3%)*
100 meters	*3 (14.3%)*
200 meters	*4 (18.2%)*
500 meters	*1 (4.8%)*
unknown	*1 (4.8%)*

Transition to secondary progressive MS

Table 2. Topic guide

Questions	Prompts
Can you start by telling me all about what you thought and felt when you were first diagnosed with MS?	Main issues, explore concerns, feelings, (physical, psychological, family issues) what did he/she do about each problem that was identified?
Can you tell me about the time you found out you have moved to secondary progressive type of MS?	Explore how did they find out, period before receiving the diagnosis, thoughts and feelings after receiving the secondary diagnosis.
Can you tell me about what you think and feel about having MS now?	Examples of issues identified?
Can you tell me about all the things you found (un)helpful when dealing with challenges of MS?	Feelings and thoughts on support interventions they were offered or sought/ how did they apply the advice given (if given), support they would like to see in the future
Are there any other relevant issues we haven't covered that you would like to mention?	

Transition to secondary progressive MS

Table 3. Examples of SPMS transition and challenges over time

Table 4. Examples of self-management and changes over time

Figure 1. Data analysis flow

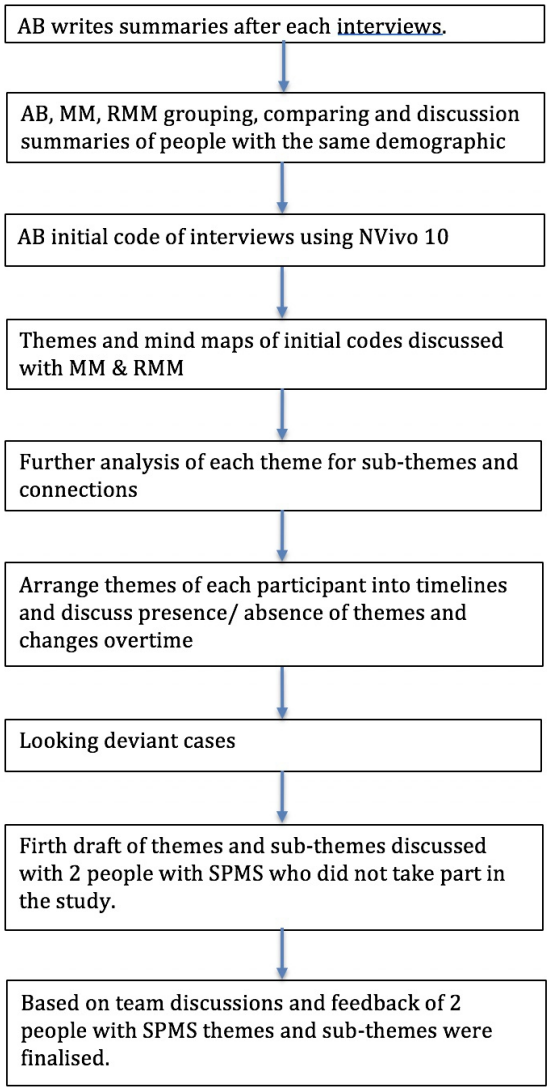

Figure 1. Data analysis flow

Figure 1. Data analysis flow

336x523mm (72 x 72 DPI)

Consolidated criteria for reporting qualitative studies (COREQ): 32-item checklist

Developed from:
Tong A, Sainsbury P, Craig J. Consolidated criteria for reporting qualitative research (COREQ): a 32-item checklist for interviews and focus groups. *International Journal for Quality in Health Care*. 2007. Volume 19, Number 6: pp. 349 – 357

No. Item	Guide questions/description	Reported on Page #
Domain 1: Research team and reflexivity		
Personal Characteristics		
1. Interviewer/facilitator	Which author/s conducted the inter view or focus group?	Angeliki Bogosian
2. Credentials	What were the researcher's credentials? E.g. PhD, MD	Angeliki Bogosian PhD, Senior lecturer Myfanwy Morgan PhD, Emeritus Professor Rona Moss-Morris PhD, Professor
3. Occupation	What was their occupation at the time of the study?	Angeliki Bogosian Research fellow Myfanwy Morgan Professor Rona Moss-Morris Professor
4. Gender	Was the researcher male or female?	Female
5. Experience and training	What experience or training did the researcher have?	The researcher has completed qualitative methodologies training as part of the MSc in Health Psychology course (Southampton University) and attended a two-day workshop on longitudinal

		qualitative research organized by the Methodological Innovation Network and delivered at Southampton University (Nov 2012). At the time of this study the researcher had completed and published 3 qualitative studies as the lead researcher (conducting of interviews, analysis and write-up) and supervised 1 MSc student and 2 PhD students on their qualitative methodologies.
Relationship with participants		
6. Relationship established	Was a relationship established prior to study commencement?	No
7. Participant knowledge of the interviewer	What did the participants know about the researcher? e.g. personal goals, reasons for doing the research	Participant information sheet and consent form
8. Interviewer characteristics	What characteristics were reported about the inter viewer/facilitator? e.g. Bias, assumptions, reasons and interests in the research topic	Participants and Methods

Domain 2: study design		
Theoretical framework		
9. Methodological orientation and Theory	What methodological orientation was stated to underpin the study? e.g. grounded theory, discourse analysis, ethnography, phenomenology, content analysis	Data analysis
Participant selection		
10. Sampling	How were participants selected? e.g. purposive, convenience, consecutive, snowball	Participants and Methods
11. Method of approach	How were participants approached? e.g. face-to-face, telephone, mail, email	Participants and Methods
12. Sample size	How many participants were in the study?	Participants and Methods
13. Non-participation	How many people refused to participate or dropped out? Reasons?	Participants and Methods
Setting		
14. Setting of data collection	Where was the data collected? e.g. home, clinic, workplace	Interviews
15. Presence of non-participants	Was anyone else present besides the participants and researchers?	No
16. Description of sample	What are the important characteristics of the sample? e.g. demographic data, date	Participants and Methods
Data collection		
17. Interview guide	Were questions, prompts, guides provided by the authors? Was it pilot tested?	Table 2, Patient & involvement
18. Repeat interviews	Were repeat interviews carried out? If yes, how many?	Methods

19. Audio/visual recording	Did the research use audio or visual recording to collect the data?	Interviews
20. Field notes	Were field notes made during and/or after the inter view or focus group?	Data analysis
21. Duration	What was the duration of the inter views or focus group?	Interviews
22. Data saturation	Was data saturation discussed?	Participants and Methods
23. Transcripts returned	Were transcripts returned to participants for comment and/or correction?	No
Domain 3: analysis and findings		
Data analysis		
24. Number of data coders	How many data coders coded the data?	Data analysis
25. Description of the coding tree	Did authors provide a description of the coding tree?	n/a
26. Derivation of themes	Were themes identified in advance or derived from the data?	Data analysis
27. Software	What software, if applicable, was used to manage the data?	Data analysis
28. Participant checking	Did participants provide feedback on the findings?	Patient and public involvement
Reporting		
29. Quotations presented	Were participant quotations presented to illustrate the themes/findings? Was each quotation identified? e.g. participant number	Results
30. Data and findings consistent	Was there consistency between the data presented and the findings?	Discussion
31. Clarity of major themes	Were major themes clearly presented in the findings?	Results
32. Clarity of minor themes	Is there a description of diverse cases or discussion of minor themes?	Results

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Multiple challenges for people after transitioning to secondary progressive multiple sclerosis: a qualitative study

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Multiple challenges for people after transitioning to secondary progressive multiple sclerosis: a qualitative study

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Keywords: secondary progressive, Multiple Sclerosis, transition, self-management, qualitative research, health services

Word count: 3,850

Abstract

Objectives: Transitioning to secondary progressive multiple sclerosis (SPMS) is demanding for both patients and healthcare professionals. The particular challenges and the ways patients cope are poorly understood. The present study examines what challenges people face when diagnosed with SPMS, by exploring experiences of people who have transitioned recently (up to 5 years).

Design: Semi-structured qualitative interviews at two time points a year apart. Interviews were analysed using inductive thematic analysis.

Setting: United Kingdom

Participants: We interviewed twenty-one people at baseline and seventeen participated in the follow-up interviews.

Results: The majority of participants reported expecting to transition to SPMS, and the diagnosis did not make much difference to them. Participants described increasing emotional and physical challenges after transitioning to SPMS and between the first and second interview. Planning, using distractions and maintaining social roles helped participants cope with the increased challenges. The same coping strategies were used between the two interviews. Participants felt there was not much left to do regarding the management of their symptoms. A key theme was the sense of abandonment from health care services after transitioning to SPMS.

Conclusions: After transitioning to SPMS people are faced with multiple challenges. Participants described a lack of directions for symptoms management and lack of support from the healthcare system. An integrated multidisciplinary health care approach is crucial at

the progressive stage of the disease to alleviate feelings of helplessness and promote symptom management.

Strength and limitations of this study

- The present study is the first to use a longitudinal qualitative design to capture people's experience after transitioning to secondary progressive multiple sclerosis.
- The focus on people recently transitioned helped to narrow down people's experiences early on in diagnosis.
- We cannot determine whether the challenges described in this study are linked to having received an official diagnosis or to the challenges faced by increased disability, as we did not interview people just before being diagnosed and straight after.

Introduction

Multiple Sclerosis (MS) affects more than 2.5 million people worldwide, including 127,000 in the United Kingdom [1]. MS is a condition that presents unique challenges for the individuals, as it usually starts with minimal physical and cognitive dysfunction which progresses over time. There are three types of MS. The most common type is relapsing-remitting (RR, 85%-90% of people with MS are diagnosed with this type) which is characterised by unpredictable attacks (relapses) of new or increasing neurological symptoms that are followed by periods of partial or complete recovery (remissions) [2]. The secondary progressive (SP) course develops in approximately 82% of those with RRMS by 20 years of onset [3]. Finally, a minority of people (10%) will be diagnosed with the primary progressive (PP) course that involves a steady disability increase without attacks [4].

The transition to secondary progressive multiple sclerosis (SPMS) is an expected part of the disease trajectory. Epidemiological studies indicate nearly 10% of people with RRMS reach the SPMS stage after five years, which increases to 25% at ten years and 75% at 30 years [5]. A mean of 2.9 $\pm$ 0.8 years is a typical length of time when people are faced with the uncertainty of whether or not they have transitioned to SPMS [6]. The delay in the diagnosis is thought to be due to the subtle nature of early progression and absent of evidence-based treatment for SPMS that could increase patients' anxieties [6]. People with SPMS are older, are less likely to be employed, and a lower proportion is female compare to people with RRMS [7]. When people transition to SPMS, they are likely to experience more severe neurological symptoms, more frequent hospitalisation [8], have more frequent and pronounced cognitive deficits [9, 10], and higher levels of fatigue [11]. The onset of SPMS has also been associated with fear, low mood [12], greater distress [13, 14], lower quality of life [15], and higher levels

of depression and anxiety [16] than in both relapsing-remitting and primary progressive types of MS. Only four qualitative studies have been conducted to examine the experiences of people with SPMS. In two of these studies, women with SPMS recognised the impact of the loss of meaningful activities on the sense of self [17, 18]. A qualitative study on patients' experiences while transitioning to SPMS showed that some people learned they had SPMS without any advanced knowledge or understanding of what SPMS meant for them, which sometimes caused confusion and upset [19]. In the fourth study, MS health professionals highlighted the value of adequate information and support around the transition period, while patients talked about the reclassification with SPMS as a turning point [20].

We know little about people's experiences after transitioning to SPMS, and psychological and behavioural interventions aimed at people with SPMS are scarce. Further, a recent meta-analysis on the effectiveness of interventions for treating depression in MS identified 13 papers, with 8 of these excluding people with progressive MS from participating [21]. When people with SPMS are included in psychological interventions, they also seem to gain fewer benefits compared to people with RRMS [22]. The psychological challenges that people with SPMS face may, therefore, differ to the ones that people with RRMS face

In this study, we explored psychological and physiological challenges people experience after transitioning to SPMS and the strategies they use to manage those challenges over time. Knowing the challenges that patients face at this stage of their MS trajectory makes it easier to identify information needs and tailor support interventions. Further, it is not well understood how transitioning to SPMS contributes to psychological challenges and adjustment attempts. The present study is the first to use a longitudinal qualitative design to capture people's experience shortly (within five years) after transitioning, so recollection of

transition is recent but also there is time for people to have developed new coping strategies. The follow-up interviews allowed respondents to reflect on the changes (or lack of them) since the first interview. The focus on people recently transitioned helped to narrow down people's experiences early on in diagnosis.

Patients and public involvement

This research was part of a larger research project, and two individuals with SPMS were part of our user involvement committee, and they have helped with different aspects of the project from the initial conceptualisation of the study, feedb`ack on the grant application and feedback of the topic guide. They were also asked to assess the burden of participation in the study. The two people with SPMS gave us feedback on an initial version of our themes. They inquired about whether the experiences differ based on the age of diagnosis or whether the people who described more psychological challenges had also reported cognitive difficulties. Following this, we checked the emerging themes to identify any patterns or differences in themes of people with different demographic characteristics. The two people with SPMS also suggested exploring further the experience of diagnosis and the importance of receiving the diagnosis of a SPMS from a healthcare professional. Finally, they commented on the clarity of description and presentation of the results, for example, they made some suggestions about how themes linked together and asked for a few additional quotes at places to better illustrate themes. Participants of the study were sent the final results of the study but were not asked to provide feedback.

Participants and methods

This research is part of an MS Society UK funded junior research fellowship awarded to the first author with a view to increase understanding of psychological challenges and adjustment

of people with progressive MS. The current study was approved by the West of Scotland Research Ethics Committee (14/WS/0077). We recruited participants through online adverts on the MS Society UK website, adverts after speaking engagements at MS Society events and through study's information sheets provided by research nurses at MS hospital clinics (Northamptonshire Healthcare NHS Foundation Trust and Cumbria Partnership NHS Foundation Trust). At that stage through the participants' information sheet, potential participants learnt the aims of the research project and what the researchers were planning to use the information for, i.e. develop a psychological intervention tailored to the needs of people with progressive MS. Participants who self-reported that they had received a diagnosis of primary progressive and secondary progressive MS from their neurologist or MS nurse were recruited, but for the aims of this study we focused on the interviews of people with secondary progressive MS. Eighty-seven people with primary and secondary progressive MS expressed interest in participating in the study. Fifteen (17%) did not meet the inclusion criterion of diagnosis within five years of progressive MS. The remaining 72 completed a short screening questionnaire that included demographic questions, as well as questions about MS duration, symptoms and walking ability. We chose to interview people using maximum variation [23], i.e. sampling to represent different demographic characteristics. No one refused participation after they have been invited to the interview. Participants were recruited from a wide range of locations in the UK, including more rural (small towns) and urban areas, therefore there was also a variability on the services they were able to access. Interviewing ceased once data saturation was reached, defined as the point at which no new information or themes are observed in the data [24]. We interviewed 21 participants who were diagnosed with secondary progressive MS and had given informed consent to take part in the study.

As shown in Table 1, participants were between 40 and 77 years of age (mean: 57.38 years) at the time of the interview. More than half the sample was unemployed or retired due to MS (n=14, 66.66%) and lived with their partners (n=15, 71.43%). Half of the participants interviewed (n=12, 57.1%) were able to walk for 20 meters or less, which is reflective of the demographics of people with SPMS. Most people with MS accumulate no more than moderate disability (fully ambulatory) during relapsing remitting phase [25] and the onset of SP phase represents the key determinant of severe disability accumulation [25-27]. Seventeen participants were interviewed for the second time, four participants (19%) did not respond to the email invitations for a second interview, and no reason was provided.

Interviews

Researchers had not established a relationship with the participants prior to study commencement. Participants were given the option of face-to-face or telephone interviews. In the first round, all participants opted for telephone interviews, and the interviews lasted between 26 minutes and 160 minutes (mean=79 minutes). Interviews were conducted by the first author (AB, female). AB has conducted her MSc, PhD and post-doctoral studies in the area of MS and through her contact with people with MS and her knowledge from the literature has come to the conclusion that people with progressive MS are underrepresented in research literature and they feel left out when it comes to treatments and health care services. This sparked the interest in the research topic. The initial interviews were conducted between April 2014 and April 2015. The second set of interviews tended to be slightly shorter as they focused on changes in between interviews and lasted between 36 minutes and 103 minutes (mean=66.12). With one exception, participants in the second round also opted for telephone interviews. There was no one else present during the interview besides the

participant and the researcher (AB). The second round of interviews were conducted between July 2015 and April 2016. There was some variation in the intervals between the first and second interview, due to staff changing institute and participants' availability. One interview was conducted 13 months after the first interview, seven interviews were conducted 14 months after the first interview, 8 interviews were conducted 15 months after the first interview and one interview was conducted 18 months after the first interview. Table 2 shows the topic guide used for the two interviews. Examples people gave in their first interviews were also used as prompts to assess changes in emotions, thoughts or symptom-management. All interviews were audiotaped with consent and transcribed verbatim. Transcripts were not sent back to participants to comment.

Data analysis

We used inductive thematic analysis [28]. An inductive approach means the themes identified emerge from the data themselves [29]. Therefore, the inductive analysis is the process of coding the data without trying to fit it into a pre-existing coding frame, or the researcher's analytic preconceptions. This form of thematic analysis is, therefore, data-driven [28].

Coding was undertaken by the first author (AB) under the supervision of the second author (MM) and with discussion of emerging themes with the third author (RMM). AB has extensive experience analysing qualitative methodologies. She completed qualitative methodologies training as part of the MSc in Health Psychology course (Southampton University) and attended a two-day workshop on longitudinal qualitative research organized by the Methodological Innovation Network and delivered at Southampton University (Nov 2012). At the time of this study the researcher had completed and published 3 qualitative studies as the lead researcher (conducting of interviews, analysis and write-up), supervised 2 MSc

student and 2 PhD students on their qualitative methodologies, and was teaching a series of qualitative methodologies lectures for MSc and Doctorate students. A variety of techniques were employed to increase familiarisation with the data. Audiotapes of each interview were listened repeatedly, and transcripts were read and reread. We used NVivo 10 for coding. The analysis of the transcripts was conducted in parallel with data collection and refinement of the themes continued after the end of the data collection, during writing up. First, each coding unit in the first transcript was given a code name, using vocabulary as close as possible to that used by participants themselves [30]. Initial codes were then applied systematically to the entire dataset, and new codes developed and refined as appropriate. We also grouped participants' interviews that shared similar demographic characteristics and explored patterns or commonalities. We examined whether themes differed across sub-groups (e.g. method of recruitment, gender, and family circumstances). Figure 1 shows the steps followed during the analysis. The analysis was an iterative process and at times we had to go back and forth between steps. The coding tree in table 3, shows how we moved from codes to refined categories. In order to document the analysis process, a detailed paper trail was kept with the notes taken after the interviews, the development of the codes and relationship between the raw data and the refined categories and codes. The quotes presented in the result section were chosen for typicality in illustrating the themes. Pseudonyms are used for the quotes.

Results

Two overarching themes about (1) Transitioning to SPMS and challenges and (2) Adaptive strategies over time. The first theme illustrated challenges involved in the transition and the second theme related to how participants responded to those challenges. Tables 4 provides examples of SPMS transition and challenges over time and Table 5 provides examples of self-

management and changes over time.

Transitioning to SPMS and challenges

Initial reaction to the transition

For the majority of the participants the transition to SPMS was expected rather than shocking. For some, transitioning did not make much difference because at that point they had already lost essential relationships and activities they valued. For others transitioning to SPMS did not make much difference because the progressive worsening of their symptoms was slow and subtle.

We compared the interviews of people that found about the SPMS diagnosis in different ways, for example, accidentally when visiting the hospital for other issues, or in one of the annual consultations or they suspected they had transitioned and asked the neurologist for confirmation. In contrast to the majority, the five participants, who found out about the transition accidentally described being upset and shocked. These five people had MS between 3 to 21 years (median=12 years).

> *[Neurologist's name] started other MRI's and tests and things and said that I'd progressed to secondary progressive. So, I was shocked and- I was shocked that I got signed off work, and it progressed. I thought he'd just say, "You're signed off for a couple of weeks", not that and, I just thought, you know, "If I sleep then I'll be all right." (female, 45, T1)*

Worsening of symptoms and emotional shifts

Transitioning to SPMS also meant worsening of symptoms, and people described progressive worsening of symptoms including mobility, fatigue, vision, and bladder dysfunction, urinary

tract infections and falls. There were further worsening of symptoms reported between the first and second interview. This deterioration also brought more changes in participants' lives.

Transitioning to SPMS also meant worsening of emotional issues. A few participants had a diagnosis of depression before or after the initial diagnosis of MS, which made managing MS more difficult. However, the majority of participants described dealing with other difficult emotions in both interviews, including feeling like a burden, being afraid of having accidents, being trapped or feeling embarrassed. They also expressed low self-esteem and confidence, and some participants described the uncertainty of whether they have the skills to cope with MS, accept the condition and deal with stress.

> *I had a bad fall last week where I split all my head open, so I am feeling a bit lack of confidence in just going out for a little walk up the road and back with my walker on my own.* (female, 54, T2)

Overall, in the second interview participants talked in more depth about their psychological difficulties and challenges in coping over the years and presented a more severe picture than in the first round of interviews. Further, people who described emotional challenges in the first interview described the same challenges in the second interview, a year later.

Arriving at the point of no help

After transitioning to SPMS, most participants reported that they did not feel health services had anything more to offer since they could no longer have access to disease-modifying drugs. A few participants talked about the benefits of physiotherapy but either the sessions offered in the NHS were too few or physiotherapy was expensive to access privately. They saw their transition as a point beyond help.

I have an MS nurse but quite frankly you know I have come to an end what you can do, I don't feel I am getting any support at all at the moment. I feel I am being left to my own devices. Yes I see him every six months, every twelve months but there is nothing he can offer so you know it is a bit of a waste of time really. (female, 54, T2)

However, a few participants reported doing their own research and identifying treatments that could potentially help them (e.g. oxygen therapy, incontinence nurse) and then asked their healthcare team to refer them accordingly. Also, a few participants attended self-management courses run by local MS charities or access psychological treatments as part of research projects.

When asked whether there is any support that they would like to receive in the future, one participant said: '*I think there is quite a lot of support when you're first diagnosed but I think it tails off I think people find it more difficult to support you the longer you have the MS I think people become quite cynical about the help that can be got and cynical about what the NHS has to offer and cynical about the help that can be obtained as to whether it's any use or not.*' (female, 65, T2). Most participants shared a similar opinion.

Adaptive tasks and changes over time

Planning activities or scaling down activities

Participants reported adjusting their holiday destinations and plans, to take into account their limitations, without changing their previous lifestyle too much. Planning ahead for future deterioration (e.g. financially, house modifications) and planning activities and days out meticulously and well in advanced, helped people feel in control. Sometimes, across the two interviews participants appeared to overestimate their limitations and underestimate their

ability to cope in the future at the first interview compared with what people reported at the second interview. This discrepancy is illustrated below by the quotes of the same participant in the first and second interview.

> *I don't go too far now, I can only cope with about three and half hours on the plane at the most. I can't go long distance or anything like that, you know long haul anymore, it's usually the Canary Islands or somewhere like that, somewhere nice and close (female, 54, T1)*

> *I am still determined that I am going to travel and see as many places as I want. We went away for six weeks over January/February, we went to New Zealand and Australia (female, 55, T2)*

However, at times, participants found it difficult to plan or modify their activities and instead scaled back, especially when the condition progressed rapidly, and almost all participants who had mentioned difficulties finding alternative activities in the first interview had the same difficulties in the second interview.

> *I can't read the same that I used to; I can't go on the computer; I can't really watch TV; gardening, the high impact exercises, you know, the long walk so everything's just had to be tailored down and you know brought into a shorter time period. The human is pretty good at finding ways around things…so yeah you just adapt um it's a struggle you know there's no two ways about it but it is that life is definitely a struggle but again you get used to that really as well (female, 59, T2)*

Emotional regulation

Strategies to manage emotional difficulties included distraction such as playing video games,

reading books, going out for a meal, or avoiding thinking and talking about the future, avoiding MS group meetings and spending the day sleeping. Some people tried to be positive by repeating 'things could have been worse'. Further, partners played an important role in emotionally supporting the participants, by helping them adjust to new MS challenges or by being available to discuss participants' worries. People reported using the same strategies between the two interviews. A few participants talked about how they would have liked access to psychological services.

> *It would be nice to talk about how you feel about it, would be it'd be nice if…do you know it would be nice if just to have somebody there saying you're doing a grand job [Laugh] with how you're doing. Because you are isolated (female, 53, T2)*

However, two participants said that they were offered psychological support, but they thought this was unnecessary.

Being social, fitting in and being of use

For most of the participants transitioning to SPMS also meant increased mobility problems and use of a wheelchair. Use of a wheelchair also posed accessibility issues leading to more carefully planned outings. Even though going out came with more difficulties, and at times people felt it was easier to stay in, most talked about the importance of being with friends and family and retaining relationships and also meeting new people. Participants described getting involved in charity work, MS research and helping others. They reported that this gave them a new purpose, they learned new things and met people. Participants who talked about the importance of being social or helping others in the first interview also talked about it in their second interview, as illustrated below by the same participant at the two time points.

> …going out with friends, going out for dinner, going and mixing with other people, which I would of quite enjoyed before and not thought anything about it, but now I think, "oh do I really want to go?" but then I force myself, you have got to go, because then when I do go, I do enjoy it and I do feel better because your mood's lifted and once you are moving around, things are easier. (female, 59, T1)

> We've got the local MS branch, we meet twice a month, first and third Tuesdays of the month. I find that very helpful. I do find that helpful to talk to other people that have got the same sorts of problems that you've got, gone through the same things. And again there's somebody there that, it's not perhaps a close friend, but has become or is becoming more of a close friend that we will see one another outside of the MS branch. (female, 59, T2)

Discussion

Reflecting on the SPMS diagnosis, people overall did not express trauma or distress, only on the occasions, the diagnosis came as a surprise. People who have recently transitioned to SPMS reported facing a worsening of the condition alongside increased emotional challenges. In the second interview, most people described the increased severity of their symptoms and more emotional difficulties. Participants reported detailed planning, distractions and remaining as active and as involved in the community as possible helped to cope with the condition. Some participants overestimated their limitations and underestimated their capacity to cope between the first and the second interview but overall most reported using similar coping strategies between the two time points. A common theme across all interviews was a sense of abandonment from the health care services and the sense that nothing can be done at this stage of their condition. Therefore, providing appropriate support after the

transition diagnosis is vital.

A qualitative study of people recently diagnosed with MS showed that psychological well-being was described as precariously contingent on reasonable current and future health status [31], for example, people were not sure whether they would be able to cope if they lost their job or had to use a wheelchair. The current study examines what happens after people have moved to SPMS. Participants in our study talked about increased disability since they have transitioned to SPMS or between the first and second interview, but despite these increased difficulties, people felt that health care professionals had nothing more to offer and there was nothing more they could do to control their emotional or physical symptoms. Focusing on well-being is often on diagnosable depression and anxiety. However, participants in the present study described having to deal with challenging emotions which may not reflect in these clinical diagnoses.

An essential finding of this study is that people who described some challenges with self-management in the first interview described the same challenges in the second interview, a year later. This can be linked to their perception of SPMS as the stage where nothing further can be done to manage things. According to the Common Sense Model [32]. Being diagnosed with an illness that is perceived as more controllable will lead to less distress and more control efforts such as seeking advice on symptom management. Participants interviewed here described SPMS as a condition they and their health care team did not have any control over. As people move from RRMS to SPMS, the capacity of personal and treatment control declines, and creates a significant challenge in adapting to the change by identifying areas that they can still have control while letting go of unattainable goals [33].

Participants in the current study reported that there is nothing they or health care

professionals can do to manage their increasing limitations. Even though self-management interventions in progressive MS are scarce, there is some preliminary evidence of interventions that could help with symptom management. For example, endurance training can improve walking [34]; progressive resistance training may improve lower limb strength [35]; bodyweight-supported treadmill can reduce pain [36], and mindfulness training can reduce anxiety, depression and impact of MS [37]. Where available, health care professionals should signpost patients to appropriate services that can help with MS symptoms but more needs to be done to find ways of developing these services, so they are more readily available.

The present study has certain significant strengths. First, to our knowledge, it is the first study attempting to investigate challenges and adaptive tasks shortly after people have transitioned to SPMS. The particular advantage of the present study was its longitudinal approach that allowed the identification of patterns over time. This showed that whereas more severe emotional and physical symptoms were reported in the second interview, these did not result in different or more challenges. While this may suggest participants feeling more comfortable in opening up at time two having built a rapport with the interviewer, it may also reflect the lack of support people were receiving. Finally, a strength of this study also lies in the specificity of the sample selected, who were all people who had transitioned to SPMS within the previous five years.

The study has certain limitations. Firstly, the qualitative exploratory nature of the study does not allow for causal relationships to be established. Second, getting a SPMS diagnosis may take years, in part due to substantial diagnostic uncertainty as a result of the subtlety of signs of early progressive disease [6]. Therefore, it is hard to determine whether the challenges described are linked to having received an official diagnosis or to the challenges faced by

increased disability, as we did not interview people just before being diagnosed and straight after. Furthermore, participants self-reported transitioning to SPMS, although they were asked to confirmed that they have received the transitioning diagnosis officially from an MS health professional, we did not verify this information by cross-checking their medical records. Also, their recollections of the time of the diagnosis might not be accurately represented during the interviews that took place up to five years later. Finally, telephone interviews might be considered inferior to face-to-face, since rapport can be harder to establish. However, research indicates both telephone and in-person interviews can be used productively in qualitative research[38], with the former potentially allowing participants to feel more relaxed and willing to disclose sensitive information[39], enhancing the data's trustworthiness.

Regardless of the above limitations, the findings presenting here underline the additional physical and emotional challenges people face when transitioned to SPMS and that people try to adapt by planning, finding distractions and remaining social but they reported there was nothing further to do regarding symptom management. Identifying physical and emotional challenges at the time of the diagnosis to SPMS will help referral to appropriate services within a multidisciplinary clinical team. Offering people options for continuing symptoms management at the time of SPMS diagnosis may help with feelings of helplessness accompany the diagnosis.

Acknowledgments

The authors would like to thank the people with MS who took part in this study.

Conflict of interest

We have no conflict of interest to declare.

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Author statement

AB and RMM designed the study. AB conducted the interviews, led the data analysis and drafted the manuscript. MM contributed to the development of the topic guide, oversaw data collection and analysis and contributed to drafts of the paper. RMM advised on the conduct of the research, contributed towards refining themes, interpretations of the findings, and drafts of the paper.

Data sharing statement

Paper trail and NVivo coding files available from the corresponding author

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Table 1. Participants' characteristics

Participants' characteristics (T1)	*Number (%)*
Age (Mean, SD)	*57.33 (9.13)*
Gender-female	*16 (72.7%)*
Marital status (N, %):	
Married/cohabiting	*15 (71.4%)*
Single	*3 (14.3%)*
Divorced/separated	*3 (14.3%)*
Time since initial diagnosis:	
0-5 years	*5 (23.8 %)*
6-10 years	*3 (14.3%)*
11-15 years	*3(14.3%)*
16-20 years	*2 (9.5%)*
21years +	*8 (38.1%)*
Time since SPMS diagnosis:	
1 year	*4 (18.2%)*
2 years	*3 (13.6%)*
3 years	*3 (13.6%)*
4 years	*4 (18.2%)*
5 years	*7 (31.8%)*
Walking ability (with aid):	
0-5 meters (EDSS 7-8)	*9 (42.8%)*
20 meters (EDSS 6.5)	*3 (14.3%)*
100 meters (EDSS 6)	*3 (14.3%)*
200 meters (EDSS 5)	*4 (18.2%)*
500 meters (EDSS $\leq$4)	*1 (4.8%)*
unknown	*1 (4.8%)*

Transition to secondary progressive MS

533 Table 2. Topic guide

Questions	Prompts
Can you start by telling me all about what you thought and felt when you were first diagnosed with MS?	Main issues, explore concerns, feelings, (physical, psychological, family issues) what did he/she do about each problem that was identified?
Can you tell me about the time you found out you have moved to secondary progressive type of MS?	Explore how did they find out, period before receiving the diagnosis, thoughts and feelings after receiving the secondary diagnosis.
Can you tell me about what you think and feel about having MS now?	Examples of issues identified?
Can you tell me about all the things you found (un)helpful when dealing with challenges of MS?	Feelings and thoughts on support interventions they were offered or sought/ how did they apply the advice given (if given), support they would like to see in the future
Are there any other relevant issues we haven't covered that you would like to mention?	

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Transition to secondary progressive MS

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537 Table 3. Coding tree of time 1 and time 2 interviews

Themes	Sub-themes	Parent codes	Associated child codes
Transitioning to SPMS and challenges	Initial reaction to the transition	SPMS diagnosis	Time of diagnosis It's easy to say SPMS Before diagnosis Before knowing Now I know Finding out (tests, hospitals, MRI, lumber puncture) Being informed
		I knew I had SPMS	You know you're on secondary MS is part of me
		It was a shock	Shock of diagnosis
		I can't do things I enjoy	It's not a good disease Cannot do things Cannot do things I used to enjoy Can't walk Can't wear normal clothes
		Progression is slow	Progression: creeps up on you Progression is slow
	Worsening of symptoms and emotional shifts	Symptoms getting worse	Various symptoms Symptoms Progression is scary MS is serious
		Depression before diagnosis	I was diagnosed with depression before
		Depression after diagnosis	Tormented Depression diagnosis Thinking of nothing else Depressed It's hard
		I'm afraid/embarrassed	Embarrassing Scared Disfigurement Weight gain is depressing Fear of the unknown I'm not as frightened anymore

		I feel I'm a burden	Annoyed Cry
	Arriving at the point of no help	Contact with MS healthcare professions	General hospitals don't understand MS Dealing with medics
		Neurologists' appointments	MS nurses Neurologists Physiotherapy
		Nothing to offer	Getting medical help Medication
		Support services: currently offered and future	There is nothing for SPMS Psychological support would have been good at diagnosis I'd like to just talk to someone who understands MS
Adaptive tasks and changes overtime	Planning activities or scaling down activities	Holidays	Planning holidays
		Planning in advance	Cannot plan in advance Need to plan meticulously Use of diaries/ reminders
		If you can't do it, do it differently	Partner showed me wheelchair is good Small things but meaningful It's silly to rebel
		Can't do much	Bad days: do nothing Resting Can't exercise Life is dull Knowing my limits
	Emotional regulation	Distractions	Sleeping Day-time TV Video-games
		Help from partner/ family	Partner helps emotionally Being outdoors Couldn't cope without my partner Harder if you're on your own Partner looks after me Partner control Partner gets angry

Transition to secondary progressive MS

		Not thinking/talking about the future	Reading about MS I don't like to talk about it
		"things could have been worse"	A frame of mind Men cope better Young people cope better
	Being social, fitting in and being of use	Accessibility problems	Disrespectful people Accessibility issues
		MS group meetings	Exchanging info Comparing with other people Comparing with other patients Partner organised support group
		Family visits	Grandchildren don't understand We're trying to be normal We do things for the family Family life
		Friendships	Revealing MS I don't look ill I have very good friends
		Voluntary work	Helping other with MS I wish I could help

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Transition to secondary progressive MS

Table 4. Examples of SPMS transition and challenges over time

Table 5. Examples of self-management and changes over time

Figure 1. Data analysis flow

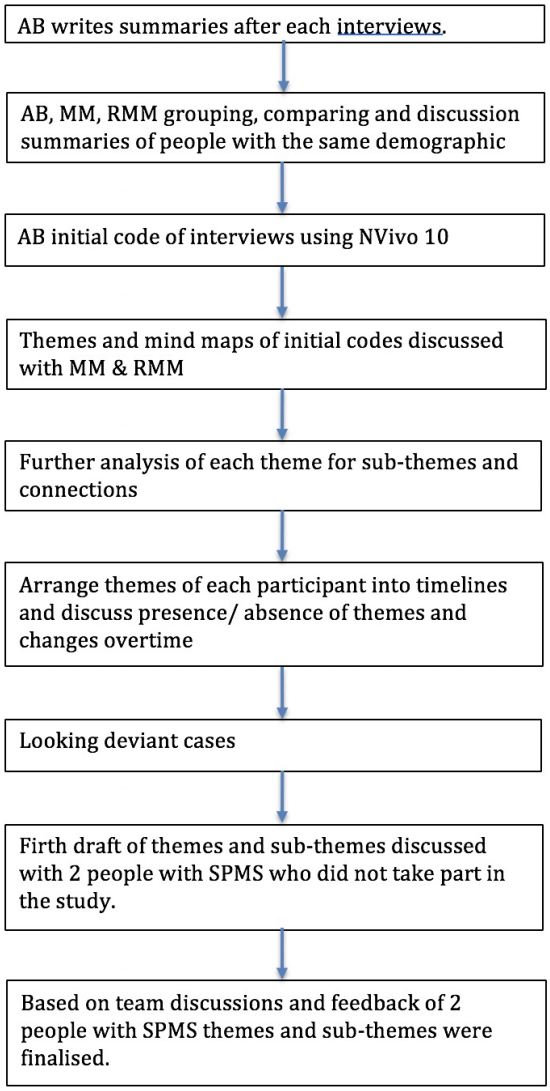

Figure 1. Data analysis flow

Figure 1. Data analysis flow

336x523mm (72 x 72 DPI)

Consolidated criteria for reporting qualitative studies (COREQ): 32-item checklist

Developed from:
Tong A, Sainsbury P, Craig J. Consolidated criteria for reporting qualitative research (COREQ): a 32-item checklist for interviews and focus groups. *International Journal for Quality in Health Care*. 2007. Volume 19, Number 6: pp. 349 – 357

No. Item	Guide questions/description	Reported on Page #
Domain 1: Research team and reflexivity		
Personal Characteristics		
1. Interviewer/facilitator	Which author/s conducted the inter view or focus group?	Angeliki Bogosian Page 9, line 165
2. Credentials	What were the researcher's credentials? E.g. PhD, MD	Angeliki Bogosian PhD, Senior lecturer Myfanwy Morgan PhD, Emeritus Professor Rona Moss-Morris PhD, Professor Page 1 (title page)
3. Occupation	What was their occupation at the time of the study?	Angeliki Bogosian Research fellow Myfanwy Morgan Professor Rona Moss-Morris Professor Page 1 (title page) & 7 (line 126-127)
4. Gender	Was the researcher male or female?	Female Page 9, line 165
5. Experience and training	What experience or training did the researcher have?	Data analysis Page 10, line 190-198
Relationship with participants		

6. Relationship established	Was a relationship established prior to study commencement?	Interviews Page 9, line 161
7. Participant knowledge of the interviewer	What did the participants know about the researcher? e.g. personal goals, reasons for doing the research	Participants and methods Page 8, lines 133-136
8. Interviewer characteristics	What characteristics were reported about the interviewer/facilitator? e.g. Bias, assumptions, reasons and interests in the research topic	Interviews Page 9, lines164-169

Domain 2: study design		
Theoretical framework		
9. Methodological orientation and Theory	What methodological orientation was stated to underpin the study? e.g. grounded theory, discourse analysis, ethnography, phenomenology, content analysis	Data analysis Page 10, line185
Participant selection		
10. Sampling	How were participants selected? e.g. purposive, convenience, consecutive, snowball	Participants and Methods Page 8, lines 143-144
11. Method of approach	How were participants approached? e.g. face-to-face, telephone, mail, email	Participants and Methods Page 8, lines 129-133
12. Sample size	How many participants were in the study?	Participants and Methods Page 8, line 149
13. Non-participation	How many people refused to participate or dropped out? Reasons?	Participants and Methods Pages 8 (lines 144-145) & 9 (line 159)
Setting		
14. Setting of data collection	Where was the data collected? e.g. home, clinic, workplace	Interviews Pages 9, line 163 & lines 172-173
15. Presence of non-participants	Was anyone else present besides the participants and researchers?	Interviews Page 9, line 173-174
16. Description of sample	What are the important characteristics of the sample? e.g. demographic data, date	Participants and Methods Table 1 Pages 8 (line 151) & 9 (lines 152-155)
Data collection		

17. Interview guide	Were questions, prompts, guides provided by the authors? Was it pilot tested?	Table 2, Patient & involvement, Interviews Pages 7 (line 113) & 10 (lines 179-182)
18. Repeat interviews	Were repeat interviews carried out? If yes, how many?	Participants and methods Pages 9 (lines 157-159)
19. Audio/visual recording	Did the research use audio or visual recording to collect the data?	Interviews Page 10, line 182
20. Field notes	Were field notes made during and/or after the inter view or focus group?	Data analysis Page 11, lines 211-214
21. Duration	What was the duration of the inter views or focus group?	Interviews Page 9, line 164 & line 171
22. Data saturation	Was data saturation discussed?	Participants and Methods Page 8, lines 147-149
23. Transcripts returned	Were transcripts returned to participants for comment and/or correction?	Interviews Page 10, line 183
Domain 3: analysis and findings		
Data analysis		
24. Number of data coders	How many data coders coded the data?	Data analysis Page 10, lines 189-190
25. Description of the coding tree	Did authors provide a description of the coding tree?	Table 3
26. Derivation of themes	Were themes identified in advance or derived from the data?	Data analysis Page 10, lines 185-186
27. Software	What software, if applicable, was used to manage the data?	Data analysis Page 11, line 200
28. Participant checking	Did participants provide feedback on the findings?	Patient and public involvement

		Page 7, lines 123-124
Reporting		
29. Quotations presented	Were participant quotations presented to illustrate the themes/findings? Was each quotation identified? e.g. participant number	Results Pages 12-17, lines221-341
30. Data and findings consistent	Was there consistency between the data presented and the findings?	Results Pages 12-17, lines 221-341
31. Clarity of major themes	Were major themes clearly presented in the findings?	Results Pages 12-17, lines: 216-220, 223-231, 240-244, 246-250, 255-258, 260-261, 264, 274-279, 282-289, 297-300, 308-314, 322-330
32. Clarity of minor themes	Is there a description of diverse cases or discussion of minor themes?	Results Pages 12-17, lines: 231-233, 244-246, 262-263, 269-273, 314-315, 319-320

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Multiple challenges for people after transitioning to secondary progressive multiple sclerosis: a qualitative study

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Multiple challenges for people after transitioning to secondary progressive multiple sclerosis: a qualitative study

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Word count: 3,850

Abstract

Objectives: Transitioning to secondary progressive multiple sclerosis (SPMS) is demanding for both patients and healthcare professionals. The particular challenges and the ways patients cope are poorly understood. The present study examines what challenges people face when diagnosed with SPMS, by exploring experiences of people who have transitioned recently (up to 5 years).

Design: Semi-structured qualitative interviews at two time points a year apart. Interviews were analysed using inductive thematic analysis.

Setting: United Kingdom

Participants: We interviewed twenty-one people at baseline and seventeen participated in the follow-up interviews.

Results: The majority of participants reported expecting to transition to SPMS, and the diagnosis did not make much difference to them. Participants described increasing emotional and physical challenges after transitioning to SPMS and between the first and second interview. Planning, using distractions and maintaining social roles helped participants cope with the increased challenges. The same coping strategies were used between the two interviews. Participants felt there was not much left to do regarding the management of their symptoms. A key theme was the sense of abandonment from health care services after transitioning to SPMS.

Conclusions: After transitioning to SPMS people are faced with multiple challenges. Participants described a lack of directions for symptoms management and lack of support from the healthcare system. An integrated multidisciplinary health care approach is crucial at

the progressive stage of the disease to alleviate feelings of helplessness and promote symptom management.

Strength and limitations of this study

- The present study is the first to use a longitudinal qualitative design to capture peoples' experience after transitioning to secondary progressive multiple sclerosis.
- The focus on people recently transitioned helped to narrow down peoples' experiences early on in diagnosis.
- We cannot determine whether the challenges described in this study are linked to having received an official diagnosis or to the challenges faced by increased disability, as we did not interview people just before being diagnosed and straight after.

Introduction

Multiple Sclerosis (MS) affects more than 2.5 million people worldwide, including 127,000 in the United Kingdom [1]. MS is a condition that presents unique challenges for the individuals, as it usually starts with minimal physical and cognitive dysfunction which progresses over time. There are three types of MS. The most common type is relapsing-remitting (RR, 85%-90% of people with MS are diagnosed with this type) which is characterised by unpredictable attacks (relapses) of new or increasing neurological symptoms that are followed by periods of partial or complete recovery (remissions) [2]. The secondary progressive (SP) course develops in approximately 82% of those with RRMS by 20 years of onset [3]. Finally, a minority of people (10%) will be diagnosed with the primary progressive (PP) course that involves a steady disability increase without attacks [4].

The transition to secondary progressive multiple sclerosis (SPMS) is an expected part of the disease trajectory. Epidemiological studies indicate nearly 10% of people with RRMS reach the SPMS stage after five years, which increases to 25% at ten years and 75% at 30 years [5]. A mean of 2.9 $\pm$ 0.8 years is a typical length of time when people are faced with the uncertainty of whether or not they have transitioned to SPMS [6]. The delay in the diagnosis is thought to be due to the subtle nature of early progression and clinicians' reluctance to label a patient as having SPMS, given the absence of evidence-based treatment for SPMS and patients' anxieties regarding its prognosis [6]. People with SPMS are older, are less likely to be employed, and a lower proportion is female when compared to people with RRMS [7]. When people transition to SPMS, they are likely to experience more severe neurological symptoms, more frequent hospitalisation [8], have more frequent and pronounced cognitive deficits [9, 10], and higher levels of fatigue [11] compared to people with RRMS. The onset of

SPMS has also been associated with fear, low mood [12], greater distress [13, 14], lower quality of life [15], and higher levels of depression and anxiety [16] than in both relapsing-remitting and primary progressive types of MS. Only four qualitative studies have been conducted to examine the experiences of people with SPMS. In two of these studies, women with SPMS recognised the impact of the loss of meaningful activities on the sense of self [17, 18]. A qualitative study on patients' experiences while transitioning to SPMS showed that some people learned they had SPMS without any advanced knowledge or understanding of what SPMS meant for them, which sometimes caused confusion and upset [19]. In the fourth study, MS health professionals highlighted the value of adequate information and support around the transition period, while patients talked about the reclassification with SPMS as a turning point [20].

We know little about peoples' experiences after transitioning to SPMS, and psychological and behavioural interventions aimed at people with SPMS are scarce. Further, a recent meta-analysis on the effectiveness of interventions for treating depression in MS identified 13 papers, with 8 of these excluding people with progressive MS from participating [21]. When people with SPMS are included in psychological interventions, they also seem to gain fewer benefits compared to people with RRMS [22]. The psychological challenges that people with SPMS face may, therefore, differ to the ones that people with RRMS face.

In this study, we explored psychological and physiological challenges people experience after transitioning to SPMS and the strategies they use to manage those challenges over time. Knowing the challenges that patients face at this stage of their MS trajectory makes it easier to identify information needs and tailor support interventions.

Further, it is not well understood how transitioning to SPMS contributes to psychological

challenges and adjustment attempts. The present study is the first to use a longitudinal qualitative design to capture peoples' experience shortly (within five years) after transitioning, so recollection of transition is recent but also there is time for people to have developed new coping strategies. The follow-up interviews allowed respondents to reflect on the changes (or lack of them) since the first interview. The focus on people recently transitioned helped to narrow down peoples' experiences early on in diagnosis.

Methods

Patients and public involvement

This research was part of a larger research project, and two individuals with SPMS were part of our user involvement committee, and they have helped with different aspects of the project from the initial conceptualisation of the study, feedback on the grant application and feedback of the topic guide. They were also asked to assess the burden of participation in the study. The two people with SPMS gave us feedback on an initial version of our themes. They inquired about whether the experiences differ based on the age of diagnosis or whether the people who described more psychological challenges had also reported cognitive difficulties. Following this, we checked the emerging themes to identify any patterns or differences in themes of people with different demographic characteristics. The two people with SPMS also suggested exploring further the experience of diagnosis and the importance of receiving the diagnosis of SPMS from a healthcare professional. Finally, they commented on the clarity of description and presentation of the results, for example, they made some suggestions about how themes linked together and asked for a few additional quotes at places to better illustrate themes. Participants of the study were sent the final results of the study but were not asked to provide feedback.

Participants

This research is part of an MS Society UK funded junior research fellowship awarded to the first author with a view to increasing understanding of psychological challenges and adjustment of people with progressive MS. The current study was approved by the West of Scotland Research Ethics Committee (14/WS/0077). We recruited participants through online adverts on the MS Society UK website, adverts after speaking engagements at MS Society events and through study's information sheets provided by research nurses at MS hospital clinics (Northamptonshire Healthcare NHS Foundation Trust and Cumbria Partnership NHS Foundation Trust). At that stage through the participants' information sheet, potential participants learnt the aims of the research project and what the researchers were planning to use the information for, i.e. develop a psychological intervention tailored to the needs of people with progressive MS. Participants who self-reported that they had received a diagnosis of PPMS and SPMS from their neurologist or MS nurse were recruited, but here we focused on the interviews of people with SPMS. Eighty-seven people with PPMS and SPMS expressed interest in participating in the study. Fifteen (17%) did not meet the inclusion criterion of diagnosis within five years of progressive MS. The remaining 72 completed a short screening questionnaire that included demographic questions, as well as questions about MS duration, symptoms and walking ability. We chose to interview people using maximum variation [23], i.e. sampling to represent different demographic characteristics. No one refused participation after they have been invited to the interview. Participants were recruited from a wide range of locations in the UK, including more rural (small towns) and urban areas. Therefore, there was also a variability on the services they were able to access. Interviewing ceased once data saturation was reached, defined as the point at which no new information or themes are

observed in the data [24]. We interviewed 21 participants who were diagnosed with SPMS and had given informed consent to take part in the study.

As shown in Table 1, participants were between 40 and 77 years of age (mean: 57.38 years) at the time of the interview. More than half the sample was unemployed or retired due to MS (n=14, 66.66%) and lived with their partners (n=15, 71.43%). Half of the participants interviewed (n=12, 57.1%) were able to walk for 20 meters or less, which is reflective of the demographics of people with SPMS. Most people with MS accumulate no more than moderate disability (fully ambulatory) during relapsing-remitting phase [25], and the onset of the SP phase represents the key determinant of severe disability accumulation [25-27]. Seventeen participants were interviewed for the second time, four participants (19%) did not respond to the email invitations for a second interview, and no reason was provided.

Interviews

Researchers had not established a relationship with the participants prior to study commencement, with the exception of one participant, who had taken part in a previous study. Participants were given the option of face-to-face or telephone interviews. In the first round, all participants opted for telephone interviews, and the interviews lasted between 26 minutes and 160 minutes (mean=79 minutes). Interviews were conducted by the first author (AB, female). AB has conducted her MSc, PhD and post-doctoral studies in the area of MS and through her contact with people with MS and her knowledge from the literature has concluded that people with progressive MS are underrepresented in the research literature and they feel left out when it comes to treatments and health care services. This sparked the interest in the research topic. The initial interviews were conducted between April 2014 and April 2015. The second set of interviews tended to be slightly shorter as they focused on

changes between interviews and lasted between 36 minutes and 103 minutes (mean=66.12). With one exception, participants in the second round also opted for telephone interviews. There was no one else present during the interview besides the participant and the researcher (AB). The second round of interviews was conducted between July 2015 and April 2016. There was some variation in the intervals between the first and second interview, due to staff changing institute and participants' availability. One interview was conducted 13 months after the first interview, seven interviews were conducted 14 months after the first interview, eight interviews were conducted 15 months after the first interview and one interview was conducted 18 months after the first interview. Table 2 shows the topic guide used for the two interviews. Examples people gave in their first interviews were also used as prompts to assess changes in emotions, thoughts or symptom-management. All interviews were audiotaped with consent and transcribed verbatim. Transcripts were not sent back to participants to comment.

Data analysis

We used inductive thematic analysis [28]. An inductive approach means the themes identified emerge from the data themselves [29]. Therefore, the inductive analysis is the process of coding the data without trying to fit it into a pre-existing coding frame, or the researcher's analytic preconceptions. This form of thematic analysis is, therefore, data-driven [28].

Coding was undertaken by the first author (AB) under the supervision of the second author (MM) with discussions of emerging themes with the third author (RMM). AB has extensive experience analysing qualitative methodologies. She completed qualitative methodologies training as part of the MSc in Health Psychology course (Southampton University) and attended a two-day workshop on longitudinal qualitative research organised by the

Methodological Innovation Network and delivered at Southampton University (Nov 2012). At the time of this study the researcher had completed and published three qualitative studies as the lead researcher (conducting of interviews, analysis and write-up), supervised 2 MSc student and 2 PhD students on their qualitative methodologies, and was teaching a series of qualitative methodologies lectures for MSc and Doctoral students. A variety of techniques were employed to increase familiarisation with the data. Audiotapes of each interview were listened repeatedly, and transcripts were read and reread. We used NVivo 10 for coding.

The analysis of the transcripts was conducted in parallel with data collection and refinement of the themes continued after the end of the data collection, during writing up. First, each coding unit in the first transcript was given a code name, using vocabulary as close as possible to that used by participants themselves [30]. Initial codes were then applied systematically to the entire dataset, and new codes developed and refined as appropriate. We also grouped participants' interviews that shared similar demographic characteristics and explored patterns or commonalities. We examined whether themes differed across sub-groups (e.g. method of recruitment, gender, and family circumstances). Figure 1 shows the steps followed during the analysis. The analysis was an iterative process, and at times we had to go back and forth between steps. The coding tree in table 3, shows how we moved from codes to refined categories. In order to document the analysis process, a detailed paper trail was kept with the notes taken after the interviews, the development of the codes and relationship between the raw data and the refined categories and codes. The quotes presented in the result section were chosen for typicality in illustrating the themes. Pseudonyms are used for the quotes.

Results

Two overarching themes about (1) Transitioning to SPMS and challenges and (2) Adaptive

strategies over time. The first theme illustrated challenges involved in the transition and the second theme related to how participants responded to those challenges. Tables 4 provides examples of SPMS transition and challenges over time and Table 5 provides examples of self-management and changes over time.

Transitioning to SPMS and challenges

Initial reaction to the transition

For the majority of the participants, the transition to SPMS was expected rather than shocking. For some, transitioning did not make much difference because at that point they had already lost essential relationships and activities they valued. For others transitioning to SPMS did not make much difference because the progressive worsening of their symptoms was slow and subtle.

We compared the interviews of people that found about the SPMS diagnosis in different ways, for example, accidentally when visiting the hospital for other issues, or in one of the annual consultations or they suspected they had transitioned and asked the neurologist for confirmation. In contrast to the majority, the five participants, who found out about the transition accidentally described being upset and shocked. These five people had MS between 3 to 21 years (median=12 years).

> *[Neurologist's name] started other MRI's and tests and things and said that I'd progressed to secondary progressive. So, I was shocked and- I was shocked that I got signed off work, and it progressed. I thought he'd just say, "You're signed off for a couple of weeks", not that and, I just thought, you know, "If I sleep then I'll be all right." (female, 45, T1)*

Worsening of symptoms and emotional shifts

Transitioning to SPMS also meant worsening of symptoms, and people described progressive worsening of symptoms including mobility, fatigue, vision, and bladder dysfunction, urinary tract infections and falls. There were further worsening of symptoms reported between the first and second interview. This deterioration also brought more changes in participants' lives.

Transitioning to SPMS also meant worsening of emotional issues. A few participants had a diagnosis of depression before or after the initial diagnosis of MS, which made managing MS more difficult. However, the majority of participants described dealing with other difficult emotions in both interviews, including feeling like a burden, being afraid of having accidents, being trapped or feeling embarrassed. They also expressed low self-esteem and confidence, and some participants described the uncertainty of whether they have the skills to cope with MS, accept the condition and deal with stress.

> *I had a bad fall last week where I split all my head open, so I am feeling a bit lack of confidence in just going out for a little walk up the road and back with my walker on my own.* (female, 54, T2)

Overall, in the second interview participants talked in more depth about their psychological difficulties and challenges in coping over the years and presented a more severe picture than in the first round of interviews. Further, people who described emotional challenges in the first interview described the same challenges in the second interview, a year later.

Arriving at the point of no help

After transitioning to SPMS, most participants reported that they did not feel health services

had anything more to offer since they could no longer have access to disease-modifying drugs. A few participants talked about the benefits of physiotherapy but either the sessions offered in the NHS were too few, or physiotherapy was expensive to access privately. They saw their transition as a point beyond help.

> *I have an MS nurse but quite frankly you know I have come to an end what you can do, I don't feel I am getting any support at all at the moment. I feel I am being left to my own devices. Yes I see him every six months, every twelve months but there is nothing he can offer so you know it is a bit of a waste of time really. (female, 54, T2)*

However, a few participants reported doing their own research and identifying treatments that could potentially help them (e.g. oxygen therapy, incontinence nurse) and then asked their healthcare team to refer them accordingly. Also, a few participants attended self-management courses run by local MS charities or access psychological treatments as part of research projects.

When asked whether there is any support that they would like to receive in the future, one participant said: '*I think there is quite a lot of support when you're first diagnosed but I think it tails off I think people find it more difficult to support you the longer you have the MS I think people become quite cynical about the help that can be got and cynical about what the NHS has to offer and cynical about the help that can be obtained as to whether it's any use or not.*' (female, 65, T2). Most participants shared a similar opinion.

Adaptive tasks and changes over time

Planning activities or scaling down activities

Participants reported adjusting their holiday destinations and plans, to take into account their

limitations, without changing their previous lifestyle too much. Planning for future deterioration (e.g. financially, house modifications) and planning activities and days out meticulously and well in advanced, helped people feel in control. Sometimes, across the two interviews participants appeared to overestimate their limitations and underestimate their ability to cope in the future at the first interview compared with what people reported at the second interview. This discrepancy is illustrated below by the quotes of the same participant in the first and second interview.

I don't go too far now, I can only cope with about three and a half hours on the plane at the most. I can't go long distance or anything like that, you know long haul anymore, it's usually the Canary Islands or somewhere like that, somewhere nice and close (female, 54, T1)

I am still determined that I am going to travel and see as many places as I want. We went away for six weeks over January/February, we went to New Zealand and Australia (female, 55, T2)

However, at times, participants found it difficult to plan or modify their activities and instead scaled back, especially when the condition progressed rapidly, and almost all participants who had mentioned difficulties finding alternative activities in the first interview had the same difficulties in the second interview.

I can't read the same that I used to; I can't go on the computer; I can't really watch TV; gardening, the high impact exercises, you know, the long walk so everything's just had to be tailored down and you know brought into a shorter time period. The human is pretty good at finding ways around things…so yeah you just adapt um it's a struggle you know there's no two ways about it, but it is that life is definitely a struggle but

again you get used to that really as well (female, 59, T2)

Emotional regulation

Strategies to manage emotional difficulties included distraction such as playing video games, reading books, going out for a meal, or avoiding thinking and talking about the future, avoiding MS group meetings and spending the day sleeping. Some people tried to be positive by repeating 'things could have been worse'.

Further, partners played an important role in emotionally supporting the participants, by helping them adjust to new MS challenges or by being available to discuss participants' worries. People reported using the same strategies between the two interviews. A few participants talked about how they would have liked access to psychological services.

It would be nice to talk about how you feel about it, would be it'd be nice if…do you know it would be nice if just to have somebody there saying you're doing a grand job [Laugh] with how you're doing. Because you are isolated (female, 53, T2)

However, two participants said that they were offered psychological support, but they thought this was unnecessary.

Being social, fitting in and being of use

For most of the participants transitioning to SPMS also meant increased mobility problems and use of a wheelchair. Use of a wheelchair also posed accessibility issues leading to more carefully planned outings. Even though going out came with more difficulties, and at times people felt it was easier to stay in, most talked about the importance of being with friends and family and retaining relationships and also meeting new people. Participants described getting involved in charity work, MS research and helping others. They reported that this gave

them a new purpose, they learned new things and met people. Participants who talked about the importance of being social or helping others in the first interview also talked about it in their second interview, as illustrated below by the same participant at the two time points.

> …going out with friends, going out for dinner, going and mixing with other people, which I would have quite enjoyed before and not thought anything about it, but now I think, "oh do I really want to go?" but then I force myself, you have got to go, because then when I do go, I do enjoy it and I do feel better because your mood's lifted and once you are moving around, things are easier. (female, 59, T1)

> We've got the local MS branch, we meet twice a month, first and third Tuesdays of the month. I find that very helpful. I do find that helpful to talk to other people that have got the same sorts of problems that you've got, gone through the same things. And again, there's somebody there that, it's not perhaps a close friend, but has become or is becoming more of a close friend that we will see one another outside of the MS branch. (female, 59, T2)

Discussion

Reflecting on the SPMS diagnosis, people overall did not express trauma or distress, only on the occasions, the diagnosis came as a surprise. People who have recently transitioned to SPMS reported facing a worsening of the condition alongside increased emotional challenges. In the second interview, most people described the increased severity of their symptoms and more emotional difficulties. Participants reported detailed planning, distractions and remaining as active and as involved in the community as possible helped to cope with the condition. Some participants overestimated their limitations and underestimated their capacity to cope between the first and the second interview but overall most reported using

similar coping strategies between the two time points. A common theme across all interviews was a sense of abandonment from the health care services and the sense that nothing can be done at this stage of their condition. Therefore, providing appropriate support after the transition diagnosis is vital.

A qualitative study of people recently diagnosed with MS showed that psychological well-being was described as precariously contingent on reasonable current and future health status [31], for example, people were not sure whether they would be able to cope if they lost their job or had to use a wheelchair. The current study examines what happens after people have moved to SPMS. Participants in our study talked about increased disability since they have transitioned to SPMS or between the first and second interview, but despite these increased difficulties, people felt that health care professionals had nothing more to offer and there was nothing more they could do to control their emotional or physical symptoms. Focusing on well-being is often on diagnosable depression and anxiety. However, participants in the present study described having to deal with challenging emotions which may not reflect in these clinical diagnoses.

An essential finding of this study is that people who described some challenges with self-management in the first interview described the same challenges in the second interview, a year later. This can be linked to their perception of SPMS as the stage where nothing further can be done to manage things. According to the Common Sense Model [32]. Being diagnosed with an illness that is perceived as more controllable will lead to less distress and more control efforts such as seeking advice on symptom management. Participants interviewed here described SPMS as a condition they and their health care team did not have any control over. As people move from RRMS to SPMS, the capacity of personal and treatment control declines,

and creates a significant challenge in adapting to the change by identifying areas that they can still have control while letting go of unattainable goals [33].

Participants in the current study reported that there is nothing they or health care professionals can do to manage their increasing limitations. Even though self-management interventions in progressive MS are scarce, there is some preliminary evidence of interventions that could help with symptom management. For example, endurance training can improve walking [34]; progressive resistance training may improve lower limb strength [35]; bodyweight-supported treadmill can reduce pain [36], and mindfulness training can reduce anxiety, depression and impact of MS [37]. Where available, health care professionals should signpost patients to appropriate services that can help with MS symptoms but more needs to be done to find ways of developing these services, so they are more readily available.

The present study has certain significant strengths. First, to our knowledge, it is the first study attempting to investigate challenges and adaptive tasks shortly after people have transitioned to SPMS. The particular advantage of the present study was its longitudinal approach that allowed the identification of patterns over time. This showed that whereas more severe emotional and physical symptoms were reported in the second interview, these did not result in different or more challenges. While this may suggest participants feeling more comfortable in opening up at time two, having built a rapport with the interviewer, it may also reflect the lack of support people were receiving. Finally, a strength of this study also lies in the specificity of the sample selected, who were all people who had transitioned to SPMS within the previous five years.

The study has certain limitations. Firstly, the qualitative exploratory nature of the study does not allow for causal relationships to be established. Second, getting an SPMS diagnosis may

take years, in part due to substantial diagnostic uncertainty as a result of the subtlety of signs of early progressive disease [6]. Therefore, it is hard to determine whether the challenges described are linked to having received an official diagnosis or to the challenges faced by increased disability, as we did not interview people just before being diagnosed and straight after. Furthermore, participants self-reported transitioning to SPMS, although they were asked to confirm that they have received the transitioning diagnosis officially from an MS health professional, we did not verify this information by cross-checking their medical records. Also, their recollections of the time of the diagnosis might not be accurately represented during the interviews that took place up to five years later. Finally, telephone interviews might be considered inferior to face-to-face, since rapport can be harder to establish. However, research indicates both telephone and in-person interviews can be used productively in qualitative research[38], with the former potentially allowing participants to feel more relaxed and willing to disclose sensitive information[39], enhancing the data's trustworthiness.

Regardless of the above limitations, the findings presenting here underline the additional physical and emotional challenges people face when transitioned to SPMS and that people try to adapt by planning, finding distractions and remaining social but they reported there was nothing further to do regarding symptom management. Identifying physical and emotional challenges at the time of the diagnosis to SPMS will help referral to appropriate services within a multidisciplinary clinical team. Offering people options for continuing symptoms management at the time of SPMS diagnosis may help with feelings of helplessness that accompany the diagnosis.

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Conflict of interest

We have no conflict of interest to declare.

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Author statement

AB and RMM designed the study. AB conducted the interviews, led the data analysis and drafted the manuscript. MM contributed to the development of the topic guide, oversaw data collection and analysis and contributed to drafts of the paper. RMM advised on the conduct of the research, contributed towards refining themes, interpretations of the findings, and drafts of the paper.

Data sharing statement

Paper trail and NVivo coding files available from the corresponding author

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Table 1. Participants' characteristics

Participants' characteristics (T1)	*Number (%)*
Age (Mean, SD)	*57.33 (9.13)*
Gender-female	*16 (72.7%)*
Marital status (N, %):	
Married/cohabiting	*15 (71.4%)*
Single	*3 (14.3%)*
Divorced/separated	*3 (14.3%)*
Time since initial diagnosis:	
0-5 years	*5 (23.8 %)*
6-10 years	*3 (14.3%)*
11-15 years	*3(14.3%)*
16-20 years	*2 (9.5%)*
21years +	*8 (38.1%)*
Time since SPMS diagnosis:	
1 year	*4 (18.2%)*
2 years	*3 (13.6%)*
3 years	*3 (13.6%)*
4 years	*4 (18.2%)*
5 years	*7 (31.8%)*
Walking ability (with aid):	
0-5 meters (EDSS 7-8)	*9 (42.8%)*
20 meters (EDSS 6.5)	*3 (14.3%)*
100 meters (EDSS 6)	*3 (14.3%)*
200 meters (EDSS 5)	*4 (18.2%)*
500 meters (EDSS $\leq$4)	*1 (4.8%)*
unknown	*1 (4.8%)*

Transition to secondary progressive MS

535 Table 2. Topic guide

Questions	Prompts
Can you start by telling me all about what you thought and felt when you were first diagnosed with MS?	Main issues, explore concerns, feelings, (physical, psychological, family issues) what did he/she do about each problem that was identified?
Can you tell me about the time you found out you have moved to secondary progressive type of MS?	Explore how did they find out, period before receiving the diagnosis, thoughts and feelings after receiving the secondary diagnosis.
Can you tell me about what you think and feel about having MS now?	Examples of issues identified?
Can you tell me about all the things you found (un)helpful when dealing with challenges of MS?	Feelings and thoughts on support interventions they were offered or sought/ how did they apply the advice given (if given), support they would like to see in the future
Are there any other relevant issues we haven't covered that you would like to mention?	

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Transition to secondary progressive MS

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539 Table 3. Coding tree of time 1 and time 2 interviews

Themes	Sub-themes	Parent codes	Associated child codes
Transitioning to SPMS and challenges	Initial reaction to the transition	SPMS diagnosis	Time of diagnosis It's easy to say SPMS Before diagnosis Before knowing Now I know Finding out (tests, hospitals, MRI, lumber puncture) Being informed
		I knew I had SPMS	You know you're on secondary MS is part of me
		It was a shock	Shock of diagnosis
		I can't do things I enjoy	It's not a good disease Cannot do things Cannot do things I used to enjoy Can't walk Can't wear normal clothes
		Progression is slow	Progression: creeps up on you Progression is slow
	Worsening of symptoms and emotional shifts	Symptoms getting worse	Various symptoms Progression is scary MS is serious
		Depression before diagnosis	I was diagnosed with depression before
		Depression after diagnosis	Tormented Depression diagnosis Thinking of nothing else Depressed It's hard
		I'm afraid/embarrassed	Embarrassing Scared Disfigurement Weight gain is depressing Fear of the unknown I'm not as frightened anymore

		I feel I'm a burden	Annoyed Cry
	Arriving at the point of no help	Contact with MS healthcare professions	General hospitals don't understand MS Dealing with medics
		Neurologists' appointments	MS nurses Neurologists Physiotherapy
		Nothing to offer	Getting medical help Medication
		Support services: currently offered and future	There is nothing for SPMS Psychological support would have been good at diagnosis I'd like to just talk to someone who understands MS
Adaptive tasks and changes overtime	Planning activities or scaling down activities	Holidays	Planning holidays
		Planning in advance	Cannot plan in advance Need to plan meticulously Use of diaries/ reminders
		If you can't do it, do it differently	Partner showed me a wheelchair is good Small things but meaningful It's silly to rebel
		Can't do much	Bad days: do nothing Resting Can't exercise Life is dull Knowing my limits
	Emotional regulation	Distractions	Sleeping Day-time TV Video-games
		Help from partner/ family	Partner helps emotionally Being outdoors Couldn't cope without my partner Harder if you're on your own Partner looks after me Partner control Partner gets angry

Transition to secondary progressive MS

		Not thinking/talking about the future	Reading about MS I don't like to talk about it
		"things could have been worse"	A frame of mind Men cope better Young people cope better
	Being social, fitting in and being of use	Accessibility problems	Disrespectful people Accessibility issues
		MS group meetings	Exchanging info Comparing with other people Comparing with other patients Partner organised support group
		Family visits	Grandchildren don't understand We're trying to be normal We do things for the family Family life
		Friendships	Revealing MS I don't look ill I have very good friends
		Voluntary work	Helping others with MS I wish I could help

Table 4. Examples of SPMS transition and challenges over time

Interview	P15	P19	P12
Pre-transitioning Context	Participant transitioned to SPMS 14 years after initial diagnosis. Before the MS diagnosis, she was diagnosed with depression. She needed support after the diagnosis, but none was offered.	Participant transitioned to SPMS 7 years after initial diagnosis. Unable to work due to MS and he faced financial difficulties after the diagnosis of MS.	Participant transitioned to SPMS 21 years after initial diagnosis. Before the MS diagnosis, she had a lot of close friends and family members that she saw regularly.
First interview	She expected to have transitioned to SPMS; it was not a surprise, but her neurologist was reluctant to diagnose transition to SPMS. Her condition has progressed a lot after the transition, especially her mobility, pain and fatigue. She was not sure whether anything could be done with SPMS. She had difficulties coping with the loss of control and uncertainty.	The SPMS diagnosis had not a significant impact on him as he had already lost everything he valued after his initial diagnosis. He reported a lack of sensations, trouble with thinking, very severe pain, tremor and diagnosis of depression. He knew that stress made his symptoms worse but could not do anything to control stress.	She did not expect to transition to SPMS and was shocked. She thought she had a relapse. She reported difficulties with mobility, fatigue and pain. She did yoga and meditation to manage her pain and has learned how to pace to manage her fatigue through workshops organised by her MS nurse. She had a positive outlook that helped her cope with MS.
Second interview	Her pain and fatigue more severe compared to her first interview but mobility remained the same. She was diagnosed with another autoimmune condition and with depression. She felt low and isolated. She was worried about her other condition and was disappointed with the lack of support from health professionals on how to manage her conditions.	His fatigue and pain were more severe compared to his first interview, and he lost bladder control. Depression was still present. Feeling isolated and completely depleted of energy. He did not think there was any ~~legal~~ way to manage fatigue or pain or that anyone could help him.	Her fatigue and pain were worse compared to her first interview, and she was not able to exercise anymore. She was diagnosed with another long-term condition. Her health care team always had good suggestions on how to manage her symptoms for both her conditions.

Table 5. Examples of self-management and changes over time

Interview	P01	P05	P21
Pre-transitioning Context	She was diagnosed with SPMS from the start. Before the MS diagnosis, she had a higher managerial role, which she left after her diagnosis.	He transitioned to SPMS 6 years after initial diagnosis. Before the MS diagnosis, his job taught him how to anticipate risks and plan accordingly.	She transitioned to SPMS 23 years after initial diagnosis. Before the SPMS diagnosis, she was diagnosed with depression and other long-term conditions.
First interview	She reduced her activities, but she was OK with that. She had to change her goals to simpler more achievable ones. She did not need to see a psychologist to help her adjust. She would have liked to volunteer, but she could not commit to it.	He planned meticulously so he can carry on his usual activities and attended MS self-management workshops. Keeping busy and planning helped him to deal with worries about the future. He was involved with MS charities and MS research to help others and also learn more about MS.	She tried to plan everything but sometimes planning is too much and it is not worth the trouble. She wanted to exercise and see her friends more, but she was too busy with her health care appointments. She believed that you have to do things for yourself not wait for the scientists to find the cure.
Second interview	Her MS symptoms and severity progressed a lot compared to her first interview. She reduced her activities even further. Her neurologist referred her to see a psychologist, but she did not want to see one. She avoided seeing friends as they were shocked by her deterioration. She did not want to see health care professionals as it was a waste of hers and their time.	His MS symptoms and severity did not change compared to his first interview. He managed to do most of the things he used to do with some alterations or at a slower pace. He was still very happy with the support from his health care team and how effectively they referred him to appropriate specialists. He still enjoyed volunteering at the local MS centre and taking part in MS-related research.	Her MS symptoms and severity progressed little compared to her first interview. She had to do her research and find out what support is available and how to manage her symptoms and then she would seek the advice from the most appropriate health care professional. The help was not forthcoming. She was not feeling low because of MS but frustrated she could not do certain things. She reported very little social activities.

Figure 1. Data analysis flow

Transition to secondary progressive MS

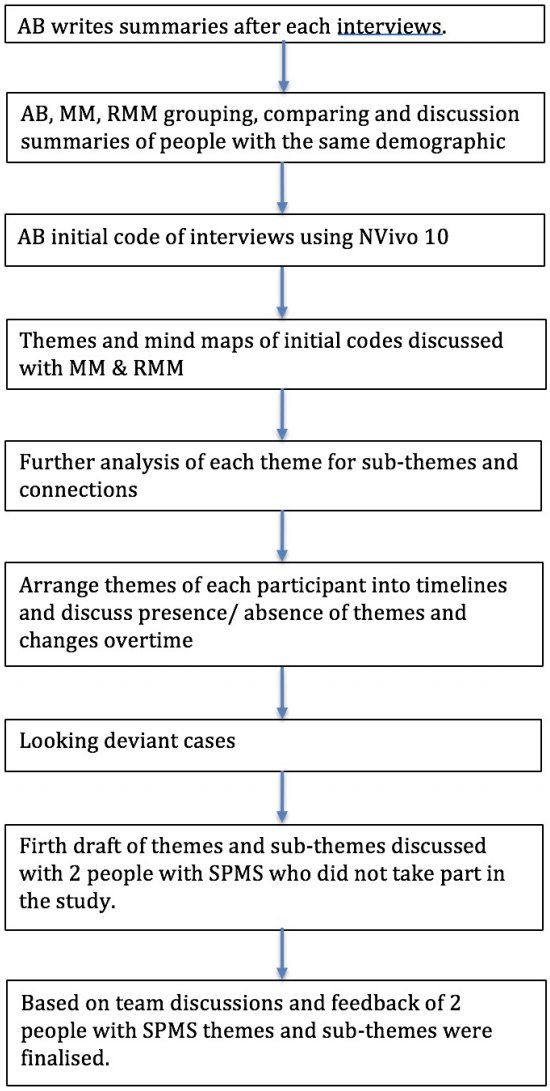

Figure 1. Data analysis flow

Figure 1. Data analysis flow

336x523mm (72 x 72 DPI)

Consolidated criteria for reporting qualitative studies (COREQ): 32-item checklist

Developed from:
Tong A, Sainsbury P, Craig J. Consolidated criteria for reporting qualitative research (COREQ): a 32-item checklist for interviews and focus groups. *International Journal for Quality in Health Care*. 2007. Volume 19, Number 6: pp. 349 – 357

No. Item	Guide questions/description	Reported on Page #
Domain 1: Research team and reflexivity		
Personal Characteristics		
1. Interviewer/facilitator	Which author/s conducted the inter view or focus group?	Angeliki Bogosian Page 9, line 165
2. Credentials	What were the researcher's credentials? E.g. PhD, MD	Angeliki Bogosian PhD, Senior lecturer Myfanwy Morgan PhD, Emeritus Professor Rona Moss-Morris PhD, Professor Page 1 (title page)
3. Occupation	What was their occupation at the time of the study?	Angeliki Bogosian Research fellow Myfanwy Morgan Professor Rona Moss-Morris Professor Page 1 (title page) & 7 (line 126-127)
4. Gender	Was the researcher male or female?	Female Page 9, line 165
5. Experience and training	What experience or training did the researcher have?	Data analysis Page 10, line 190-198
Relationship with participants		

6. Relationship established	Was a relationship established prior to study commencement?	Interviews Page 9, line 161
7. Participant knowledge of the interviewer	What did the participants know about the researcher? e.g. personal goals, reasons for doing the research	Participants and methods Page 8, lines 133-136
8. Interviewer characteristics	What characteristics were reported about the interviewer/facilitator? e.g. Bias, assumptions, reasons and interests in the research topic	Interviews Page 9, lines164-169

Domain 2: study design		
Theoretical framework		
9. Methodological orientation and Theory	What methodological orientation was stated to underpin the study? e.g. grounded theory, discourse analysis, ethnography, phenomenology, content analysis	Data analysis Page 10, line185
Participant selection		
10. Sampling	How were participants selected? e.g. purposive, convenience, consecutive, snowball	Participants and Methods Page 8, lines 143-144
11. Method of approach	How were participants approached? e.g. face-to-face, telephone, mail, email	Participants and Methods Page 8, lines 129-133
12. Sample size	How many participants were in the study?	Participants and Methods Page 8, line 149
13. Non-participation	How many people refused to participate or dropped out? Reasons?	Participants and Methods Pages 8 (lines 144-145) & 9 (line 159)
Setting		
14. Setting of data collection	Where was the data collected? e.g. home, clinic, workplace	Interviews Pages 9, line 163 & lines 172-173
15. Presence of non-participants	Was anyone else present besides the participants and researchers?	Interviews Page 9, line 173-174
16. Description of sample	What are the important characteristics of the sample? e.g. demographic data, date	Participants and Methods Table 1 Pages 8 (line 151) & 9 (lines 152-155)
Data collection		

17. Interview guide	Were questions, prompts, guides provided by the authors? Was it pilot tested?	Table 2, Patient & involvement, Interviews Pages 7 (line 113) & 10 (lines 179-182)
18. Repeat interviews	Were repeat interviews carried out? If yes, how many?	Participants and methods Pages 9 (lines 157-159)
19. Audio/visual recording	Did the research use audio or visual recording to collect the data?	Interviews Page 10, line 182
20. Field notes	Were field notes made during and/or after the inter view or focus group?	Data analysis Page 11, lines 211-214
21. Duration	What was the duration of the inter views or focus group?	Interviews Page 9, line 164 & line 171
22. Data saturation	Was data saturation discussed?	Participants and Methods Page 8, lines 147-149
23. Transcripts returned	Were transcripts returned to participants for comment and/or correction?	Interviews Page 10, line 183
Domain 3: analysis and findings		
Data analysis		
24. Number of data coders	How many data coders coded the data?	Data analysis Page 10, lines 189-190
25. Description of the coding tree	Did authors provide a description of the coding tree?	Table 3
26. Derivation of themes	Were themes identified in advance or derived from the data?	Data analysis Page 10, lines 185-186
27. Software	What software, if applicable, was used to manage the data?	Data analysis Page 11, line 200
28. Participant checking	Did participants provide feedback on the findings?	Patient and public involvement

		Page 7, lines 123-124
Reporting		
29. Quotations presented	Were participant quotations presented to illustrate the themes/findings? Was each quotation identified? e.g. participant number	Results Pages 12-17, lines221-341
30. Data and findings consistent	Was there consistency between the data presented and the findings?	Results Pages 12-17, lines 221-341
31. Clarity of major themes	Were major themes clearly presented in the findings?	Results Pages 12-17, lines: 216-220, 223-231, 240-244, 246-250, 255-258, 260-261, 264, 274-279, 282-289, 297-300, 308-314, 322-330
32. Clarity of minor themes	Is there a description of diverse cases or discussion of minor themes?	Results Pages 12-17, lines: 231-233, 244-246, 262-263, 269-273, 314-315, 319-320