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"Taming the beast": Exploring the lived experience of relapsing remitting multiple sclerosis using a life history approach

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CHAPTER 3: THE LITERATURE REVIEW

Format and scope of the literature review

There is a plethora of articles, studies and descriptive reviews reporting on the lived experience of MS. However, the great majority of this literature focuses on a certain *aspect* of living with the disease, whether it is a symptomatic aspect such as fatigue, bladder dysfunction or gait disturbance or a psychological aspect such as coping with diagnosis or change in symptoms. Others have reported on physical aspects such as falls, social aspects such as negotiating a changed world after diagnosis, or an adaptive aspect of MS, such as adapting to disability. This expansive array of literature also covers all phenotypes of the disease, CIS, RRMS, SPMS and PPMS, sometimes together and sometimes separately. Interestingly, however, there is a paucity of literature published on the overall, whole life experience of living with the phenotype of RRMS. In terms of life history research specifically, there is one article reporting on one particular symptom in MS, fatigue, but no in depth life history studies reported. The reasons for this could include the fact that the subject is so broad and heterogeneous that it is simply too vast a topic and needs to be broken down into smaller components; or it could be the time consuming nature of analysing qualitative work, especially when using the ethnographic life history approach.

This chapter, the literature review, will concentrate only on literature which addresses the experience of living with RRMS or MS in general, not reporting on specific characteristics or features of the disease, such as MS symptoms or disability. Later in the thesis, the findings and discussion chapters (6, 7 and 8) will integrate particular literature relating *specifically* to the study themes, reflecting on how the themes relate to what is already known. This approach has been recommended by others (Silverman 2010., Wolcott, 1994) as a process of conducting the bulk of the literature review in and around the specific data collection and data analysis rather than generally before the study commences. As supported by Silverman (2010), one of my study aims was to be innovative and creative in relating the literature to the study findings. *To develop*, rather than just present, ideas and concepts related to the themes advanced directly from the data. As Wolcott (1994) suggests, this calls for introducing related research towards the end of the study rather than at the beginning,

in an attempt to keep the literature more connected with the data analysis. Therefore, this traditional literature review in chapter 3 presents a critique and synthesis of the most significant research in the last three decades; research that is most akin to the current research study. In the subsequent chapters, I will present literature more directly connected with the study findings, some of which are presented in this chapter, but some literature the result of more specialised research in particular domains of MS.

The literature review in this chapter will be approached chronologically to show the progression of research and to demonstrate how new findings have built on what was already known. This approach also reflects how biomedical knowledge about MS developed and strengthened over time, allowing access to new and exciting areas of MS, supplementing the extensive quantitative and scientific work with qualitative, people-focused research.

Several search databases for academic literature were used in this literature review, in addition to medical, nursing and science search databases. These included CINAHL (nursing and allied health), PubMed (biomedical and life sciences), Scopus (scientific, medical, nursing and social sciences), PsycINFO (psychological), Embase (biomedical), Web of Life, Australian Bureau of Statistics, Libraries Australia, Joanna Briggs Institute, Cochrane library, Google Scholar, the Notre Dame University search engine "Summon" and Trove for dissertations/theses retrieval. Keywords used to search for the relevant literature were: life history, life experience, life story, lived experience, life journey, living with, battling, feelings in, meaning of, needs in, paediatric, aged, elderly, qualitative inquiry, qualitative methods, qualitative research, Multiple Sclerosis, Relapsing Remitting Multiple Sclerosis, Progressive Multiple Sclerosis, MS, RRMS, SPMS, PPMS, chronic illness, neurological disease, neurodegenerative disease, acquired disability and disability were used in a combination of ways to acquire the literature search.

The Literature Review

As already outlined in chapter 2, medications used to effectively treat RRMS did not become clinically available until just over twenty years ago, with the first treatment

for relapsing MS (Beta interferon-1b) approved by the American Food and Drug Administration in 1995 (Jacobs et al., 1996). Until that time, a considerable proportion of the qualitative MS literature was centred on coping and adjustment to the disease (Miller, 1997) with only a few descriptive reports, mostly centring on individual experiences living with MS (Robinson, 1990; Toombs 1988, 1990, 1992), but no studies offering explanations or latent meaning to the experiences.

Just under three decades ago, a study investigated the burden of disease and adaptation in 211 adults with rheumatoid arthritis (RA), hypertension and MS (Pollock, Christian, & Sands, 1990). Findings relevant to MS demonstrated that the MS group suffered more psychological stress than the other groups, even though the RA group were more physically burdened by disease than the MS participants (Pollock et al., 1990).

Moving forward through the early 1990's, a study by Hainsworth (1994) used a mixed methods approach to determine the presence of chronic sorrow in people living with MS; unfortunately the MS phenotypes were not disclosed nor levels of disability of the participants reported. Ten participants (nine female, one male) were interviewed individually and completed the Burke/NCRCS chronic sorrow questionnaire. The age of the study participants was 28-55 years and the length of time from diagnosis one to 22 years. Data was analysed using content analysis. Participants reported feelings of sadness, anger, frustration, fear and helplessness, with one participant describing their feelings when diagnosed with MS as "I felt a stabbing in my heart" and another as "it was a feeling of helplessness". The study also reported on strategies used by the participants to cope with chronic sorrow, including continuing to work, gaining support from family and friends, gathering information and thinking positively. Study participants also reported that empathic nursing support was helpful to them and an important part of their coping strategy. Even though the MS phenotypes were not disclosed, the presence of eight of the ten study participants reporting chronic sorrow as part of the MS journey possibly indicated the likelihood that many of the participants suffered from progressive forms of MS rather than RRMS, as this has not been a feature of the recent RRMS literature.

The first study using phenomenological methodology to examine the lived experience of people with RRMS was performed by Miller (1997) an MS Nurse in the United States, as part of her doctoral dissertation. This study is the most alike to the current study of all studies published in the literature in terms of the research question and the study sample. The significant difference however, is the data analysis method, which will be discussed in later chapters. The research question "What is it like for you living with MS?" was asked of 10 PwRRMS (seven female, three male) with an average age of 49 years and average duration of RRMS diagnosis of 20 years. Patients were recruited by convenience sampling to the study as they presented for neurological appointments at two MS clinics in New York over a period of about six months. To be eligible for the study participants needed to have an EDSS score of less than 3.5; indicating that they had mild-moderate physical impairment from MS and were able to mobilise independently. Semi-structured interviews were conducted on site, audio-taped, transcribed and analysed concurrently. Hermeneutic phenomenology was used to analyse the transcripts.

Miller (1997) abstracted a total of 12 themes from the study data. These themes covered many domains and included social networks (spouse, family, HCPs, can be both positive and negative), coping with the disease (changes to life because of MS, independence, resting to avoid fatigue), hope/hopelessness (initially "doom and gloom", later hope after seeing the neurologist, hope for a brighter future), issues relating to control (difficulty with unpredictability, maintain independence) and conflict (conflict with HCPs, employers, family), uncertainty (unpredictable day-to-day), issues associated with loss (of independence, employment, relationships, abilities), fear (before diagnosis of brain tumour, relapses, losing support networks) and reveal/conceal issues (concealing from others who don't understand, HCPs keeping MS suspicions to themselves, concealing from family to avoid too much concern). More positive themes centred on a feeling of relief with diagnosis (many feared a worse diagnosis) and gaining control getting to know MS (resources, observing others with MS). Importantly, the themes identified were supported with direct quotes from the participants to strengthen the theme development.

The themes from this study provided the first understanding of what it was like for people diagnosed with RRMS navigating the rapidly changing world around them,

with the first treatment for the disease on the horizon. The themes captured many aspects of life and living with illness, highlighting both the positives and the negatives. Miller (1997) did not find that chronic sorrow was a part of the experience in patients recruited for this study, as previous work had claimed (Hainsworth, 1994), this may have been a reflection of Miller's study focusing only on people with RRMS and possibly living with lower levels of disability than participants of Hainsworth's (1994) earlier study where the MS phenotype was not disclosed.

Miller (1997) identified that people with RRMS appeared to thrive on a sense of hope for, and control of, their MS and a strong drive to really know their disease. This was the first study to document these findings. Incredibly, until this time there had been arguments at length on whether clinicians should even inform patients of their MS diagnosis, due to the unpredictable course of disease, uncertainty of diagnosis, concern for stress triggering symptoms and potential emotional devastation (Sencer 1988). Miller (1997) argued the need for health care professionals (HCPs) to teach families to be supportive rather than stifling in their interactions, the role of independence and normalcy for people with RRMS, the need for nurses to explain the difference between relapsing and progressive forms of MS and the need for patients to be given information about the disease. To this day, these findings form the basis of many nursing interactions with newly diagnosed patients with RRMS and have influenced nursing practice in the specialty of MS considerably.

A major strength of this study was that Miller (1997) presented the themes back to patients in order to verify that the themes were congruent with their own experience. Additionally men with RRMS were also recruited and reflected the natural preponderance of MS incidence of 3:1/female:male (Compston & Coles, 2008), men had not been included in many of the qualitative studies up until this time. Limitations of the research primarily relate to the geographically limiting nature of a rural region in the USA and the fact that all patients interviewed were under the care of an MS clinic (and presumably receiving best practice care), potentially biasing the scope of the findings. There was also no reference to whether themes were saturated at this point, or if new themes were still emerging with the final participants enrolled in the study. However, this seminal study was one of the most important qualitative

studies in MS at this time, as the first phenomenological nursing study to examine the lived experience of RRMS. It is important to revisit the lived experience of RRMS in current times, with a completely different treatment paradigm, different management strategies, exciting biomedical and scientific MS discoveries. This will be discussed in further detail in chapter 8, after the current study findings have been reported.

Another seminal paper from Koopman and Schweitzer (1999) followed shortly after Miller's study (1997), and consisted of a qualitative project exploring the journey to MS of five participants, three females and two males with a mixture of disease courses of RRMS and PPMS. Participants were all diagnosed with MS within the previous 12 months, were aged 29-40 years and had experienced symptom onset with MS for between six months and 22 years. Although concentrating on the period up to and around diagnosis, the study aimed to discover the "essence of the lived experience". Individual semi-structured interviews were performed starting with the question "What was it like to have symptoms of an illness and then to be told you have MS?". Data analysis was not discussed in depth, but was reported as being performed using "common threads and patterns", one would imagine this being an early form of thematic analysis. Four major themes were identified from the data and were conceptualised as Whispered Beginnings (onset of symptoms, creating possible answers), Echoes of Silence (intermittent symptoms, come and go, worry and wait), The Spoken Words (unexpected diagnosis, pain, have to tell others, shock, numbness) and Recreating Voice (claiming the diagnosis, losses and gains, refocusing and changing life plans)

Koopman and Schweitzer (1999) concluded that the diagnosis of MS led to uncertainty and variability and participants needed to develop strategies to cope with the challenges of a disease with no cure. Although the themes provided new insights into understanding the experience, a deeper discussion about the methods of data analysis and separating the results for the different phenotypes of the participants would have led to readers being able to make informed decisions about generalisability to their particular situations and contexts. Similar to Miller (1997), this study identified worry, fear and feelings of loss in the participants as major themes living with MS.

A growing body of literature began to emerge exploring how people with MS were making sense of their world, their lived experience. Differences between MS as a "disease" and as an "illness" were seen as important to continuing the dialogue. Clair (2003) suggested that disease is seen as an objective measure of pathological change within the body and illness as the "human experience" of the disease. An exploratory study of 16 women diagnosed with MS (phenotypes not disclosed) initially used narrative data collection from focus group interviews with six participants, to generate an agenda of topics to explore during the second phase of the study with ten semi-structured interviews at a later date (Clair, 2003). Participants were recruited through a local New Zealand MS Society and purposive sampling was used to select women from a variety of settings and situations for the interviews. Data was analysed using a computer based software package to assist coding and analysis and symbolic interactionism was used to build a theoretical foundation for interpreting the illness. Symbolic interactionism has been described as looking for meaning by human beings, with the meaning derived from social interactions they encounter and subsequently process (Blumer, 1969).

Findings suggested that to live in a world of MS is to live in a world with uncertainty and little to no control. Essentially, two separate datasets were evident: women striving to identify the cause of their illness and find meaning, and women who transformed themselves and their illness, reclaiming control and finding peace and harmony (Clair, 2003). Intuitively, the women in the study engaged in story telling to make sense of their illness and regain control over their lives. Employing an in-depth interview method enabled the women to tell their stories, often for the first time, to convey their lived experience with MS. Clair (2003) surmised that MS can be seen in one of four ways, firstly MS as an aggressor (fight it head on, or let it run its course), or MS as a saviour (delivered to save the person from the person they were or were becoming), MS as a guest (but find ways to minimise the intrusiveness) or MS as an adversary (inspiring to overcome obstacles). These findings echoed some of the earlier findings from Miller (1997) and Koopman and Schweitzer (1999) in terms of seeing MS as an adversary and the feelings of participants losing control living with uncertainty. Coping with the disease, previously reported by Miller (1997) is aligned somewhat with Clair's (2003) viewing of MS as an aggressor and fighting the

disease, or MS as a guest and finding ways to reduce the disease impact. However, the findings of MS as a saviour were novel and interesting to consider for HCPs caring for PwMS.

Limitations to the study were due to only women being included and that the participants interviewed were located in a regional area of New Zealand (NZ). Unfortunately the phenotypes of MS of the study participants were not disclosed, it would be interesting to know if the themes predominantly reflected a relapsing form of MS or if the participants were living with progressive MS. A major strength of the study included a purposive sampling method to obtain maximum variation in order to reach participants across a wide range of demographics. This improves generalisability of the study findings to others. It is also worth noting that although published in 2003, the study took place in 1996, before DMTs were available to people with MS living in NZ, rendering a replication of the study worthwhile.

The nature of invisible symptoms in MS and the uncertainty of what lies ahead when living with an unpredictable disease provided the inspiration for researchers to explore the lived experience of MS using separate focus groups for men (n=4) and women (n=6) (Courts et al., 2004). Again, unfortunately the MS phenotypes were not disclosed (Courts et al., 2004). The data was analysed by searching for themes, but no supporting information on how this was achieved was outlined in the article. Study findings identified four main themes; Nobody Listened (nobody takes symptoms seriously, a long time to diagnosis), Symptom Devastation (overwhelming symptoms, battle for normalcy, pain and anger), Picking and Choosing (making choices to regain control, using denial) and Fight Your Own Fight (self advocacy, taking charge). Once again, issues of control and conflict with HCPs, as reported by Miller (1997) were also a feature of these study findings. The theme of Fighting Your Own Fight by taking charge was somewhat seen previously in Miller's (1997) themes of getting to know MS and control, but the presentation of the other themes offered new approaches to understanding MS.

Participants in this study described creative solutions to the continual challenges and changes of MS interfering with goals in their lives. At times they described feeling depressed, dejected and desperate. Their most poignant need was for someone to

listen to them and teach them, as they identified knowledge as power (Courts et al., 2004). The authors suggested that nurses should utilise interventions to empower patients and also to teach self-management strategies.

The concept of the devastation experienced with symptom onset was reported by the researchers to be greatly helped by patient knowledge of their disease, which subsequently assists nurses in planning education sessions for patients and this finding is consistent with previous suggestions for practice (Miller, 1997). These themes again build on earlier work, as well as introducing new concepts for further research. The most notable being the importance of people with MS making their own decisions and practicing self-advocacy, which remain as major teaching elements in the MS Nurse repertoire to this day. By examining the lived experience, nurses are encouraged by the authors to build partnerships with patients, listen to patients, teach patients how to navigate the health care system, provide information and to teach self-advocacy and self-management.

Limitations to the study by Courts et al. (2004) included the study being confined to one small geographical area. A description of the data analysis phase of the study was overlooked in the report, which therefore limits readers being able to replicate the study in their own environment. Once again, no MS phenotypes were disclosed, it would have been interesting to note whether there were differences between relapsing or progressive courses. Strengths of the study centred on recruiting both men and women with MS to better represent both genders. Having the three investigators attend all focus groups together, and reaching a consensus on theme identification also strengthened the study results.

The life-world of one person with early onset RRMS was explored in great depth and richness during an unstructured interview at the end of the participant's first year of living with RRMS (Finlay, 2003). This dissertation used a case study method to focus on one individual's MS experience. Phenomenological methodology was used to access and describe life-world experience, however no meanings or abstractions were explained during the course of the research. The interview began with the general question "What is living with multiple sclerosis like for you?" and continued for a period of two hours, with prompts and requests for examples from the

researcher. Analysis of study results took place in two phases. Firstly, a narrative was created using verbatim quotes and secondly analytic methods suggested by Wertz (1983) involved repeated, systematic readings of the transcript, dwelling on the phenomenon through immersion and reflection, then describing the recurring themes that were present (Finlay, 2003).

The rich description of this lived experience illustrates the overwhelming impact of illness on the life-world and introduces the concept of embodiment in MS nursing research (Finlay, 2003). The lived experience in one persons' body is their own, personal experience. By focusing on the impact of physical illness, we are better able to understand that our embodied experiencing cannot be separated from the essence of who we are as a person and our place in the world (Finlay, 2003). It is just that – simply one person's lived experience.

The major limitation to this study is the focus on just this one individual. However, such a rich narrative may provide nurses with greater understanding, which can then be applied to others with MS, in ways deemed appropriate by the nurse to the context. The second possible limitation to this study is that the subject and researcher had been friends for 15 years prior to the study, possibly introducing some bias and assumed knowledge into the interview. However that may also be seen as a particular strength of the study; with strong rapport, trust and openness already existing between researcher and participant. This may have resulted in less censorship and more open responses.

Two years later, the life journey of one study participant with RRMS was also explored by Fawcett and Lucas (2006) exploring the journey from the first disease symptoms of a fit, active female subject aged in her early thirties who was a friend of one researcher. This study revealed the following key points; nurses play a pivotal role in MS care, the beginning of the journey is fraught with questions, GPs may not recognise symptoms as being MS, the impact of the diagnosis of MS should not be underestimated, uncertainty is a challenge, understanding and getting to know MS is an important step and that support from family and friends is valued by PwMS.

Similarly to Finlay's (2003) study, limitations to this work takes account that one researcher and the participant were friends before the study took place. However, as already mentioned, this could have strengthened the study findings based on the trust and rapport they already shared. Nevertheless, this in-depth case study adds to our insights and understanding of RRMS. Identifying the nurse as being in a pivotal role in the MS journey, the challenge of uncertainty, the under-recognition of MS symptoms in the community, the value of family and friend support and the need for resources to get to know MS further supports the previous research of others (Miller, 1997; Clair, 2003).

Barker-Collo et al. (2006) performed a qualitative study using semi-structured interviews to obtain narratives from 16 people, half living with RRMS and the other half with progressive MS. The age range for the study was 27-72 years with five males and 11 females. The primary aim of the study was to expand on the earlier work of Koopman and Schweitzer (1999) using a larger sample to explore the experience and subsequent journey at the time of MS diagnosis. The research was analysed using thematic analysis. The strength of this study was the separation and comparison of RRMS and progressive MS in the findings and discussion. A marked difference was found between PwRRMS and progressive MS. RRMS narratives tended to reflect variability and unpredictability for the patient and for their family "it's the not knowing". This contrasts with progressive MS where the prevailing theme was a sense of hopelessness in relation to the inevitability of disease related decline "you have this thing that will eventually make you a burden". Other themes in general, where RRMS and progressive MS results were reported together, included the impact of changes to life roles, the fear of impact from MS on others, the possibility of positive effects on relationships and on lifestyle and health, the potential for isolation from others, the need for appropriate MS related information and the central role of the MS nurse in patient care.

Once again, similar themes to previous work from Miller (1997) and others were identified, however the emphasis on the positive effects from living with MS were a welcome addition to our knowledge about living with MS. The concept that this devastating illness could bring fresh and new insights to PwMS and cause positive change to lifestyles and relationships was innovative information.

Malcomson, Lowe-Strong and Dunwoody (2008) aimed to explore the personal accounts of people living with MS in order to gain insights for HCPs and to identify effective self management strategies for patients. This study used focus groups to interview 13 individuals, nine females and four males, six participants lived with RRMS and the rest of the sample with progressive MS. Five study participants were no longer ambulant. The study results were analysed using thematic analysis, but again unfortunately, findings were not separated according to MS phenotype.

Themes identified included learning something was wrong (distress, uncertainty, fear), getting a name (the diagnosis, unhelpful feelings), getting help (lack of psychosocial support), consequences to lifestyle (interpersonal, changing employment circumstances, challenges), getting on with day to day life (proactivity, perspective and control), providing advice for others (peer support, self management) and providing advice for HCPs (personal needs, guidance and information, peer groups). The concept of patients providing advice for HCPs is particularly enlightening and nowadays is an important part of shared decision making in clinical care. One would imagine it was a surprising finding when it was first identified, before making its way into recommended MS care in recent times (Giovannoni et al, 2016).

A possible limitation to generalising the study to others with MS is that the study participants were invited into the study because they felt "able to cope" with MS and may have thus represented only part of the MS population. The method of focus groups rather than individual interviews may have also limited the information people were prepared to divulge in a group setting in front of strangers, especially in regards to sensitive topics such as sexuality and cognition. The findings however are useful as they provide an in-depth exploration of a very important time of the MS journey (up to, during and just after MS diagnosis) and also considers the insights of both males and females.

A more recent qualitative study used interpretative phenomenological analysis (IPA) to understand the experience of young adults living with RRMS, and was undertaken as part of a nursing dissertation (Beshears, 2010). The research question was "What

is the meaning of the experience of living with RRMS for the young adult?" with the purpose of understanding the lived experience in order to propose education and support for this specific MS population. The cross sectional study used convenience, purposive sampling to recruit six female participants between 20 and 40 years of age. Data collection involved three in depth interviews, which were transcribed, and data analysed using a Miles and Huberman approach and the Heideggerian method. This form of IPA uses the researcher as the data collection instrument, uncovering meanings in the narratives of the participants (Miles & Huberman, 1994). Specifically the study aimed to discover the meaning of changes occurring in the participant since their diagnosis of RRMS, to uncover how the participants made meaning of the expectations of others in their lives and how they learnt new ways of being during MS relapses.

Beshears (2010) presented many ideas from the data analysis, including the participants wishing for opportunities to talk to others about MS, living day by day, of MS being "scary", the importance of planning ahead and the value of listening to one's body. These findings led to the compilation of four broader themes; firstly participants realising that MS involves uncertainty, their discovery that RRMS is frustrating, concepts of being scared and fearful getting to know MS and that RRMS is always in the backdrop of life.

Beshears (2010) hoped to interview both sexes in the study, but no men volunteered, so unfortunately only women were included in this study, again potentially limiting the generalisability of results to men and extending the significant gap in the literature relating to RRMS in males. All participants in the study were married with the spouse being the household wage earner, thereby further hindering generalisability of the results to other social situations. Recruitment of participants was in a rural region of the US, with potential local influences biasing the data in unknown ways, for example local health care systems and the availability of neurology care in the rural area. A major strength of the study is the rich data in the six narratives and a strong focus on intimacy and relationships, which are important developmental milestones in adult development (Busch & Hofer, 2012).

The most recent study to explore the experience of living with MS comes from Jordan in the Middle East (Al-Sharman et al., 2018) and is from the speciality of rehabilitation. A total of 16 participants (equal numbers of male and female) with an average age of 36 years and average time since diagnosis of seven years, participated in a total of four focus groups directed by an interview guide. The study aimed to explore and describe the daily living experiences and challenges of PwMS. The study sample was a convenience sample representing two geographical areas in Jordan and was the first of its kind to be undertaken in the region. Unfortunately, no phenotype was disclosed for the study participants, however the average EDSS was 3.5, indicating mild-moderate disability for most of the participants with one participant having an EDSS of 6.0 (needing a unilateral walking stick to mobilise). The focus groups were split into one gender only to encourage open discussion, which may have been impacted in this region by cultural conditions inhibiting females from discussing important insights in front of males (Metcalfe, 2008).

Two major themes were developed by the researchers: firstly, experiences related to the disease itself (physical decline, psychosocial withdrawal and fear of the future) and secondly, experiences related to the health care system (difficulty of diagnosis, poor communication and rapport with HCPs, lack of awareness about MS and MS rehabilitation)

The researchers recommended greater attention be given to MS in Jordan, with improved awareness of MS for both HCPs and for the general public. In addition, Al-Sharman et al. (2018) also recommended optimising and enhancing rehabilitation services for PwMS. The study is limited by the geographical region and cultural differences to the western world, however the study is strengthened considerably by the inclusion of eight males in the research, a demographic which until now had been greatly under-represented in qualitative MS research.

Life history research and RRMS

There is only one brief piece of research exploring the life of a PwMS using a life history approach (de Chesnay, Rassilyer-Bomers, Webb, & Peil, 2008). This was a

four page narrative presented in a book chapter discussion about performing life history studies where the purpose was to present an abbreviated story, told by the participant and interpreted by the researcher, to teach others about overcoming obstacles in chronic illness (de Chesnay et al., 2008). The author of this particular MS life history narrative within the combined chapter story (Webb) presented a single life history of a 46 year old female who successfully overcame her debilitating MS-related fatigue. The data is limited by the study purpose, dealing with only one aspect of her MS experience. The current study aims to explore all facets of living with RRMS and in many participants.

The lived experience in progressive forms of MS

As the current study focused solely on RRMS, only a very short literature review will be presented on progressive MS to provide some perspective of the overall situation. Several qualitative studies have concentrated on the lived experience of people living with progressive forms of MS (Edmonds, Vivat, Burman, Silber, & Higginson, 2007; Olsson, Skär, & Söderberg, 2010; Strupp et al., 2012). Although these phenotypes are distinctly different from the proposed study group, people living with SPMS did originally start their journey with RRMS, and may have important insights to offer when considering living with RRMS.

The most relevant of these studies to the current study, is a recent dissertation in the field of psychology (O'Loughlin, 2015). The investigator aimed to gain rich insight into the experiences of people with SPMS during the critical, but poorly understood, time of disease transition and change of labelled disease phenotype from RRMS to SPMS. A total of 16 people took part in the research, nine PwSPMS and seven HCPs involved in MS care. Thematic analysis was used to explore the experiences, coping strategies and needs of people during transition from RRMS to SPMS. Four themes were developed by O'Loughlin (2015) in exploring SPMS: is this really happening? (noticing decline, feeling "in limbo"), SPMS becoming a reality (shock, meaning, turning points), feelings of living a life of struggle ("it's all downhill now") and the concept of brushing oneself off and moving on (accepting, making the best in the circumstances).

Recommendations from this study included the need for HCPs to undergo education regarding the psychological impact of SPMS during the transition, the provision of peer support, and exploration of existing resources and coping strategies that PwSPMS may already have. A great strength to the study is the fact that participants were recruited within 12 months of their SPMS transition, possibly enhancing the accuracy of their recall of events.

As a university research Professor, Toombs (1988, 1990, 1992, 1995, 2001) began a series of phenomenological work exploring her own experience of living with MS, whilst navigating the world as a researcher. Toombs (1995, p.12) writes:

> *"For MS, like every other illness, is experienced not just as the breakdown of the body, but as the disruption of the life that is lived in that body. To live with MS is to experience a global sense of disorder; a disorder which incorporates a changed relation with one's body, a transformation in the surrounding world, a threat to self, and a change in one's relation to others".*

Such intimate and descriptive detail adds to the understanding of the individual's lived experience with MS. Toombs (1988, 1990, 1992, 1995, 2001) provided reflections of her personal experience of living with progressive MS over many years to provide a phenomenological account of the human experience of disability. Particularly moving and striking in its simplicity, Toombs (1995) suggested that the loss of upright posture, as disability takes hold in MS, diminishes autonomy for the person and affects the way one is treated by others, disrupting social connections and leading to a loss of dignity. This insight from Toombs profoundly changed the way I viewed patients as I consulted in the MS clinic. In particular, it led to many changes in how I considered visits and appointments for patients, paying greater attention to positioning and clinic set-up to try and restore as much dignity as possible to the lives of PwMS visiting the clinic.

The lived experience of children and adolescents diagnosed with RRMS

Significantly fewer studies have focused on the lived experience of children and adolescents with MS, mainly due to the rarity of this condition in children younger than 18 years. An explorative qualitative study used a phenomenological approach to

explore experiences of children living with MS (Boyd & MacMillan, 2005). Patients from a children's hospital in Canada (n=12) were interviewed using a semi-structured format. A software package was utilised to sort data and then narrative analysis was used to generate themes and subthemes. The investigators found that the children described similar themes to their adult counterparts described earlier in this chapter; that is, worrying about burdens on families, fear of disclosure, hiding symptoms, dealing with uncertainty and fear of rejection from their peers (Boyd & MacMillan, 2005). A major difference was that the children and adolescents in this study reported conflict with their parents and lack of understanding from teachers, rather than the experience reported with adults who described this same conflict, but with the medical professionals (Miller, 1997). The great strength of the study is that it fills an enormous gap in knowledge regarding paediatric MS lived experiences and provides an excellent starting point for further research, possibly involving more centres and greater numbers.

A further research study in 2009 employed grounded theory to develop a theoretical model to understand psychosocial experiences of paediatrics with MS (Thannhauser, 2009). The study looked at the interplay between grief and peer relationships, which adds to the knowledge about paediatric MS, but not the lived experience. It has been suggested by others that there are significant gaps in qualitative literature in paediatric neurological conditions (Audulv, Packer, & Versnel, 2014).

Other aspects of MS and the lived experience

At the other end of the life trajectory, there is a paucity of research investigating the lived experience of the aged population with MS. Currently a quarter of people living with MS are aged over 65 years of age and experiencing a decreased health related quality of life (Buhse, 2015). This demographic of MS research demonstrates a significant gap in the literature and also needs immediate attention.

There have also been several studies investigating the lived experience of caregivers and partners of people with MS, but they have not been individually reviewed here as it is not aligned with the research question focused on people living themselves with RRMS (Aoun, McConigley, Abernethy, & Currow, 2010; Bjorgvinsdottir &

Halldorsdottir, 2014; Buhse, 2008; Buhse, Dela Ratta, Galiczewski, & Eckhardt, 2015; Cheung & Hocking, 2004; Corry & While, 2009; Pakenham & Samios, 2013; Strickland, Worth, & Kennedy, 2015).

The lived experience of other chronic illnesses

There are many illness states which have overlapping symptoms or share certain features with MS, which may also provide MS Nurses and HCPs with insights to help them understand what it is like to live with chronic illness. These lived experience studies include those in motor neuron disease (Brown & Addington-Hall, 2008; O'Brien, Whitehead, Jack & Mitchell, 2012), in stroke (Burton, 2000), in spinal cord injuries (Desanto-Madeya, 2006), in Parkinson's disease (Soundy, Stubbs, & Roskell, 2014), in medically unexplained symptoms (Nettleton, 2006), in fibromyaligia (Lempp, Hatch, Carville, & Choy, 2009), in the rare illness lymphangioleiomyomatosis (Haylen, 2015) and in lupus (Mendelson, 2006; Beckerman, 2011). There are some commonalities in the findings between these chronic conditions and RRMS, most notably feelings of being unheard and voiceless (Nettleton, 2006) of uncertainty (Haylen, 2015) and of invisible symptoms (Beckerman, 2015).

The significant gaps in the literature

A significant amount of literature regarding MS has been published in the last decade, as important advances in earlier diagnosis of the disease and the advent of more efficacious drug treatments have transformed MS care. A search in Google Scholar reveals 210,000 articles in MS since 2011. However, the great majority of this research has been biological, scientific and medical in nature. Qualitative research in particular has been lacking in this specialty.

This literature review did not reveal any other studies using life history methodology to explore the experience of people living with MS (besides a short four page narrative). Why is this so? Using life history in researching chronic illness reflects the complexity of the human experience it is examining (de Chesnay, 2014),

presenting an ideal methodology to gain insights and understanding. However, there are many challenges inherent in using this methodology, including deeply personal narratives which may affect the researcher emotionally and the fact that the interviews and follow-up are time consuming and lengthy. Perhaps it is because life history is an under-recognised methodology in nursing. These aspects will be discussed further under methodology in the next chapter, together with the reasons why life history is an ideal approach to explore the entire life experience of living with RRMS.

From this literature review examining the experiences of people living with MS, three things are abundantly clear. Firstly, there is an enormous gap in recent literature surrounding what it is like to live with and experience RRMS. There were several pivotal studies in the early to mid 1990's, most likely they arose at this point because drug treatments were not yet available and major breakthroughs in genetics and understanding scientific aspects of the disease were still several years away. In this setting most of the nursing research centred on trying to understand the patient experience, in order to inform patient education and symptom management strategies. A seminal paper by Miller (1997) outlined the lived experience of people with RRMS using a phenomenological approach and was the first of its kind to examine how it felt to be the person at the centre of the MS experience (Miller, 1997). This informed health professionals to become aware of the patient experience, that patients wanted to learn more about their disease, to take control and to be in charge, to teach families to be supportive and not stifling in assisting with their loved one's care and for nurses to be at the forefront of educating patients about the different phenotypes of MS. Unbelievably, up until shortly before this time, it was still regularly debated in medicine whether patients should actually be told their diagnosis, for fear of causing emotional devastation in a disease with no available treatments (Elian & Dean, 1985; Sencer, 1988). Miller's (1997) findings set up important conceptual frameworks for MS nursing practice to understand the concerns of people living with MS.

Several studies in the last two decades have continued to explore the experiences of people living with RRMS, but recent studies are lacking. This gap in qualitative literature is of prime importance as the landscape of treatments for MS, the potential

side effect profiles of these pharmacological treatments and the emergence of even more new therapies continues to grow and become even more complicated for people with RRMS. How do patients feel? What is it like to be living with MS today? It is imperative that qualitative research answers these questions so that nursing practice and intervention reflects the needs of patients and that nursing theory and frameworks take these needs into consideration and develop as our understanding of the disease matures. Many of the studies presented above either did not discuss the phenotype of the participants, or only partly included PwRRMS in the research. Many of the themes identified in progressive MS research are vastly different to RRMS and therefore cannot be extrapolated. If study findings refer to both groups of participants, there should be a discussion (where possible) outlining any differences between the two groups.

The second area identified with significant gaps in the literature is research involving males living with RRMS. Many of the studies described here selectively, or coincidentally, recruited only or mainly women. Although the preponderance towards women developing RRMS at a rate of 3:1 is widely known, this still leaves a significant portion of men in the community whose experience living with RRMS is under-researched and unknown. Recruiting men into the current study is an attempt to ensure men are adequately represented and by doing so will enhance our understanding of the current experience of *both* genders living with RRMS.

The third areas of significant literature gap in this field involves research into the experience of both ends of the lifespan spectrum; children and adolescents, as well as the aged population living with MS. In regards to paediatrics and adolescents living with MS, this is an acknowledged area of deficit (Audulv et al., 2014). However, this will not be the focus of the research question in this proposal due to the specialty of the area and lack of experience and skills of the researcher in paediatrics, experience which would be essential to undertake such a study. The current study unfortunately did not see any PwRRMS over the age of 65 years volunteer to participate.

Summary

The current study aims to fill the significant gaps in the qualitative literature exploring the experience of adults living with RRMS. To achieve this goal, the study sample has targeted both males and females to add to the body of knowledge in the area of RRMS. A body of knowledge which is striking for its burgeoning scientific and biomedical research, but scarcity of contemporary lived experience literature. The findings and discussion following in chapters five to seven will bring into the conversation literature relevant to each theme and subtheme as they are explored, to provide further knowledge, background and linkage with previous research and theory.

There are no studies reported in the literature that have been performed using the same methodology and methods as the current study. In many ways, this was daunting at the beginning of the study, but as the study progressed and my faith in the methodology and methods strengthened with each interview, there was a quite confidence I developed in the operational framework of the study as the life histories from the participants were so strong with relevant data. These concepts will be discussed in the next chapter and will support how the life history methodology, underused in MS research (and illness research in general), provides a dynamic and useful tool to learn as much as possible about the experience of living with RRMS. The final chapter will connect the studies discussed in this chapter's literature review with the findings from the current study, with additional concentration on the research from Miller (1997), Clair (2003) Courts et al. (2004) and Beshears (2010) as the most alike to the current study, either in study aims or study sample. However, the data analysis for these studies used hermeneutic phenomenology, symbolic interactionism, searching for themes and interpretative phenomenological analysis respectively in their data analysis to uncover meaning rather than the current study method, which used thematic analysis (Braun & Clarke, 2006, 2013).

The following chapter, Methodology and Methods, will outline the ontological and epistemological viewpoints of the study, and the study process and procedures. Chapter 4 will also explain why the selection of the life history approach is an ideal methodology to answer the research question "What is the experience of living with

RRMS?" in a novel and insightful way, to add to the body of knowledge on the experience of living with RRMS. The current research approach differs from previous research in that it is contemporary, takes into consideration recent advances in MS care and provides a unique viewpoint looking at the whole life experience and incorporating previous life events into how RRMS is experienced.