How and why does social connectedness change in families living with the effects of behavioural variant frontotemporal dementia?

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Abstract

There is a paucity of research studies exploring the changes to social connectedness of people diagnosed with bvFTD and their families, from the premorbid to late stages of the condition. The purpose of this exploratory study was to establish how and why social connectedness changed for the study families over the progression of bvFTD.

Social citizenship was selected as the conceptual framework and case study research as the methodology. Five families living with the effects of bvFTD volunteered for the research, and each person with the diagnosis was in the middle/late stages of the condition. Data was collected from the case study families and contacts, using a range of data collection tools (semi structured interviews; document review; diaries and participant observations). Thematic analysis was selected to examine the data collected from each family (within case analysis) and then the findings were compared and contrasted with the other families studied (cross case analysis). Six themes were identified from the collected data: i)."Why are they behaving like this?:*Finding a reason*; ii)."What is happening to our family? *Changing family relationships.;* iii)."Other people are noticing"; iv)."Getting a diagnosis"; v)."Now we know what it is." and vi). "Grief and loss".

Findings indicated that relationships and social connectedness deteriorated in the pre diagnosis stage, and families stated it was largely due to the behavior, personality and social functioning changes of their family member, and the belief that these changes were intentional. By the time a diagnosis was given, the relationships and social connectedness had broken down to such a degree that they could not be rebuilt. The families described how they were no longer invited to social events in the same way as before and their social network reduced significantly. The families also stated that they tended to withdraw from social networks and events due to the embarrassment caused by their family member's changed behavior.

In conclusion, if bvFTD was recognized, diagnosed and specialist advice and support, given earlier by practitioners it could enable families to understand the effects of bvFTD on their family member's behaviour. This could enable the family to retain relationships and connectedness, develop coping strategies, build resilience and prevent crises.
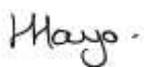
Recommendations for clinical practice: i) increased training for medical practitioners, health and social care staff regarding early recognition and diagnosis of bvFTD; ii) specialist post diagnostic support offered immediately after diagnosis, and iii) awareness raising through campaigns which could include workplace awareness training.

Declaration

This thesis records research undertaken in partial fulfilment of the requirements for the Doctorate of Professional Practice at the University of Northampton. The work presented here is the author's own, except where due reference is made. The work has not been submitted elsewhere for the award of a higher degree.

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Preface: A Personal Reflection

In qualitative research it is important for the researcher to recognise that their beliefs, background, and social identity could have an impact on the research process (Lathlean, 2010). In order to clarify my own position in relation to the research topic, participants and setting, I have written a personal reflection of my background and motivation for conducting this research.

My personal journey:

I commenced my nursing career in 1980, and during my nurse training, I had placements in wards specialising in older people with dementia. Some wards had a high standard of nursing care, despite the shortages of staff, and others were frankly appalling with poor standards of care, institutionalisation and a lack of any kind of stimulation for the patients. From my childhood I was always told to "treat others as you wish to be treated yourself", and have followed that in all parts of my life. What I saw challenged this as I saw people being treated as less than human and I was not prepared to work in this way. I reported what I saw on two different wards to the senior management but no changes were made, and as a result I was known as a "troublemaker" which had implications for me on other wards that I had placements on.

Before my experience as a student nurse, the majority of older people I had met were very fit and healthy so I was shocked by some of the sights, sounds, smells, and ways people were being treated, and it left an indelible effect on me. From this experience, I always swore that I would never be one of those

nurses who gave the appearance of not caring, and who treated older people like packages to be moved around, not spoken to, and as one more task to be completed.

My career developed over the next few years and I became a Sister on a ward specialising in dementia and I now had the chance to change the clinical practice. It was a hard process as old cultures were difficult to change, but with the right staff we did so. From this point in my career, I knew I wanted to specialise in working with people living with dementia as I knew I could make a difference to their experience.

As my career progressed, I moved into nurse education as I thought I could influence how nurses of the future would work with people living with dementia. I realised that although we taught the theory of how to work effectively with people living with dementia, when the students went to their placements in the clinical area they experienced "reality shock", and either, adapted to the work practices on the ward (even if they were of a poor standard), changed job or challenged the staff.

Throughout all of my time in higher education, I maintained clinical involvement, as I strongly believed that in order to retain knowledge and skills, a practitioner should remain grounded in their area of expertise and actively influence the clinical care that is commissioned and delivered to people living with dementia.

During this time I was approached by a Consultant Psychiatrist, who knew about my background as a dual registered nurse (mental health and general nursing), and my experience in working with dementia. She asked me to see a family who had been referred to her with young onset dementia. This was my first experience of working with a family with Pick's Disease (now known as behavioural variant frontotemporal dementia). I was fascinated by his presentation and how it affected not just him, but also his whole family. This family was slowly breaking apart, relationships which used to be very close and caring had now become fractured and punctuated by arguments, shouting, tears, accusations and at times, verbal and physical aggression. I managed to work with the family to support them to understand what was happening to them, suggested and modelled alternative ways of coping, arranged packages of care and respite, and provided the family with time and space for them to talk about their feelings and fears. A number of other Consultants started to refer younger people living with dementia to me, and whilst this was very complex work, I found I had the knowledge, skills and attitude to achieve successful results with these families.

A commissioner heard about the work I was doing and as Northants did not have any specialist provision for younger people with dementia (YPD), a working group was set up and they developed a Nurse Specialist post for YPD. I was successful in the interview and from 2004 to 2010 I set up and managed a very successful nurse led assessment and diagnostic service for younger people living with dementia. Whilst in this post I met many families living with all

types of young onset dementia and developed a special interest, knowledge and skills in working with families living with behavioural variant frontotemporal dementia (bvFTD).

From 2010 to 2013 I worked in senior management posts in the NHS, due to my leadership and management skills, and progressed into a Director of Nursing post. I believed that in this post I would be able to advance clinical practice, not only for people living with dementia, but also across a range of other specialities. However the reality of senior management posts in the NHS is more about politics, meetings and bureaucracy and less about advancing clinical practice. I decided that senior management posts in the NHS did not suit my range of knowledge, skills, values, attitudes, clinical expertise or personal priorities, and started to look for another career.

In 2013 I saw a post advertised within the charitable sector and when I read the job description and specifications it could have been written especially for me! I had heard of Admiral Nurses and the work they did with families, and in fact I had used this approach in my clinical practice. I applied and became the CEO/Chief Admiral Nurse of Dementia UK which fulfilled a long term ambition to work with an organisation that has national influence regarding improving post diagnostic care for families living with dementia and works with families in a relationship centred and solution focused way to build resilience and prevent crises.

I am now in my "dream job", an incredibly privileged position, where I am able to actively influence the advice and support offered to families living with dementia in the UK. This post has strengthened my resolve to conduct research with families whose relationships and social connections deteriorate significantly, both before and after diagnosis with behavioural variant frontotemporal dementia (bvFTD). From this research I plan to identify, and make recommendations for clinical practice and further research.

In my career, I have met families whose relationships and social connections both within and external to the family had deteriorated significantly and when I tried to find research to enhance my clinical practice, I noticed it was very sparse. I decided I would like to research this area and at the same time, was approached to consider enrolling on the Doctorate of Professional Practice. Consequently this research has been the culmination of my personal ambition, professional expertise and research activities.

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List of Abbreviations

bvFTD   Behavioural variant frontotemporal dementia

DH   Department of Health

FTD   Frontotemporal dementia

GP   General practitioner

PLWD   People or Person living with dementia

UK   United Kingdom

N.B: When discussing the author's contribution to the research, the personal pronoun "I" will be used as it provides a level of reflexivity and engagement within the research.

From chapter 1 the thesis will be written in the past tense to reflect the fact that this research took place over the time period 2013-2015.

Chapter 1: Dementia and Social Connectedness

Dementia is defined as a group of syndromes, which are characterised by a progressive decline in cognitive processes, causing significant changes in activities of living, and eventually leading to the person diagnosed losing the ability to self-care (Stephan & Brayne, 2014). Over 200 subtypes of dementia have been identified, but research studies tend to focus on the most prevalent types of dementia, namely Alzheimer's disease, vascular dementia and mixed dementia.

1.1   Statement of the problem

What research there has been on dementia over the last 30 years has tended to focus on the possible cause and identification of a cure or treatment (Bartlett & O'Connor, 2010). There has been very little research on quality of life issues, or the lived experience of a younger person diagnosed with dementia and the effects on the individual and their family (Lockeridge & Simpson, 2012). Although dementia is not just a condition of older age, there has been a tendency to focus on this age group, and the most common forms of dementia which are typically seen in the older population i.e. Alzheimer's and vascular dementia (Cabote *et al*, 2015). As there is a finite amount of money available for dementia research, it could be surmised that this focus could reduce the amount of funding available for studies into the more unusual forms of dementia, such as frontotemporal dementias.

In my clinical practice, I noted that people living with a form of young onset dementia, namely behavioural variant frontotemporal dementia (bvFTD), tended to have problems with social connectedness[1] at a relatively early stage in their condition. This is thought to be due to the clinical changes caused by frontal lobe damage affecting their behaviour, personality and communication skills (Kipps *et al*, 2007). I also observed social connectedness was reduced in the families of the person diagnosed with bvFTD, and there was a paucity of research in this area to identify the possible reasons how and why this happened. I have selected this topic for my research as it is under researched and inadequately understood from the perspective of the families living with the experience (Johannessen & Moller, 2011; Daly *et al*, 2013; Oyebode *et al*, 2013).

1.2   Prevalence and projections

The identification of new cases of dementia over a given period of time (incidence), and the total number of people with dementia in a population (prevalence), provides data in order to generate awareness, and strategically plan health and social policy, and associated infrastructure. This data can also enable the formulation of strategies and treatment to delay, or prevent some forms of dementia (Stephan & Brayne, 2014).

The incidence rates for dementia are highest in over 65 year olds, and this age group is growing at an accelerated rate worldwide (Stephan & Brayne, 2014). It

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[1] "An attribute of the self that reflects cognitions of enduring interpersonal closeness with the social world *in toto.*" (Lee, Draper & Lee, 2001. p310)

is predicted there will be a consequent rise in the numbers of people living with dementia (PLWD) worldwide, from 36 million to 66 million by 2030 and over 115 million by 2050 (ADI, 2010).

In the UK, it is estimated that there are 850,000 PLWD and the number is predicted to increase to over 1 million by 2025 and over 2 million by 2051 (Alzheimer's Society, 2014). There are national concerns that this significant growth in the numbers of PLWD will have negative effects on the UK economy and families (DH, 2015).

Although dementia is generally thought of as a condition of old age, it can affect people under the age of 65 years. It is estimated that 42,325 of PLWD are under 65 years of age but it is cautioned that this figure maybe an underrepresentation of the actual numbers due to diagnostic and reporting difficulties (Alzheimer's Society, 2014).

Table 1.1: Most common types of dementia in the over 65yr group (Alzheimer's Society, 2014) and the under 65yr group (Rossor *et al*, 2010)		
Type of dementia	Proportion of different types of dementia (%) in the over 65yr age group	Proportion of different types of dementia (%) in the under 65yr age group
Alzheimer's disease	62%	34%
Vascular dementia	17%	18%
Mixed dementia	10%	Figures not included in the study.
Dementia with Lewy bodies	4%	7%
Frontotemporal dementia	2%	12%
Alcohol related brain impairment	Figures not included in the study.	10%
Other	5%	19%

Table 1.1 lists percentage figures for the most common types of dementia in the over and under 65yr age group. This clearly indicates that there are differences

in the types of dementia diagnosed in these age groups, with fewer cases of Alzheimer's disease diagnosed in the under 65yr age group, and significantly greater numbers of people diagnosed with frontotemporal conditions and rarer dementias. Table 1.2 details the main symptoms experienced by each of the common types of dementia.

Table 1.2: Main symptoms of the most common types of dementia in the over 65yr group and the under 65yr group (Stephan & Brayne, 2014)	
Type of dementia	**Main symptoms**
Alzheimer's disease	Concentration and memory problems. Language, visuospatial and functional disability.
Vascular dementia	Poor concentration, memory and communication. Disabilities related to area of brain affected.
Mixed dementia	A combination of the symptoms of Alzheimer's disease and vascular dementia.
Dementia with Lewy bodies	Fluctuating concentration and memory problems. Spatial disorientation. Visual hallucinations.
Frontotemporal dementia	Changes in: personality; behaviour; emotional & social functioning; insight; decision-making and problem solving. Language difficulty if temporal lobe is affected.
Alcohol related brain impairment	Impairments in: memory; planning; organising; judgement; social skills and balance.
Other	Varies depending on the type of dementia.

From the comparison of symptoms for each of the common types of dementia in Table 1.2, it can be concluded that people diagnosed with frontotemporal dementia (more prevalent in the under 65yr age group, see Table 1.1) are more likely to have changes in their personality, behaviour, social and emotional functioning, than other types of commonly occurring dementia. It is these changes that families could find more challenging and distressing. In addition, the changes noted in a person diagnosed with frontotemporal dementia could

also affect their social functioning and hence their social connectedness within and external to the family.

1.3   Policy context

Over the last 10 years, there has been an unprecedented focus on dementia by world governments, policy makers, health and social care agencies and the voluntary sector (DH, 2013). This is partly due to the growth in the prevalence rates of dementia and the anticipated effects on the economy (DH, 2015). There is also the recognition that care and support for PLWD needs to improve. This has led to a change in the way dementia care and support is delivered in the UK, but not necessarily the way the public views dementia and the people diagnosed with it.

In order to explore how and why social connectedness changes for people living with dementia and their families, I believe it is important to examine the policy context. In this section, I will be focusing on key policy documents that have had, or are having, an influence on the way dementia is viewed by society (National Institute for Health and Clinical Excellence and the Social Care Institute for Excellence (NICE/SCIE), 2006; Department of Health (DH), 2009; 2012; 2013; 2015). These papers (see Table 1.3) also recommend evidence-based approaches, interventions and support that health and social care professionals and the voluntary sector should implement in order to improve dementia care in the UK.

Table 1.3: Key policy documents that have influenced dementia care, interventions and treatment in the UK	
Document	**Clinical recommendations included:**
NICE/SCIE (2006) Guideline on supporting people with dementia and their carers in health and social care.	- Identification, assessment and diagnosis of dementia. - Treatment and care of PLWD and their carers. - Agencies and services working collaboratively to provide best practice using a bio-psycho-social approach.
The National Dementia Strategy (2009) Living well with dementia.	- Provide advice and guidance and support for health and social care commissioners and providers in the planning, development and monitoring of services. - Provide a guide to the content of high quality services for dementia. - Setting up of memory assessment services. - Reduction in the prescription of antipsychotics for PLWD. - Dementia advisors and peer support services piloted.

In 2006 the NICE/SCIE guideline on supporting people with dementia and their carers in health and social care was published (see Table 1.3). This paper is important, as it also highlighted the needs of younger people living with dementia for specialist assessment, services and support. This guidance cautioned services about age discrimination whilst also recognising that younger people living with dementia had rarer dementias and they, and their families, had different challenges and needs when compared to people living with dementia over the age of 65 years. Although the guideline was influential in setting the standards for dementia care in the UK, it would have had more influence had it set mandatory minimum standards for dementia care and support, and allocated additional funding, in order to make the recommendations and guidance a reality in practice.

In 2009 (DH, 2009) the national dementia strategy, *Living Well with Dementia,* was launched (see Table 1.3). The dementia strategy was one of the first in the world and it earned international recognition. *"Living well with dementia"* set a strategic framework within which local services were expected to perform in order to deliver quality improvements to services and address health inequalities regarding dementia. Additional money was allocated by the government to commissioners of services to action the recommendations in the strategy. However, because this funding was not restricted, two thirds of Primary Care Trusts were unable to demonstrate how they spent their National Dementia Strategy funding (House of Commons All Party Parliamentary Group on Dementia, 2011) with the implication that it was used to offset a shortfall in funding elsewhere.

In 2010, the National Dementia Declaration (Alliance, D.A. (2010) was launched (see Box 1.1, overleaf) which was created by people living with dementia, family carers and organisations committed to seeking radical change in how society responds to dementia. This consisted of 7 outcomes that families living with dementia would like to see in their lives, regardless of age or type of dementia.

At the same time the Dementia Action Alliance (DAA) was launched in order to change the experience of living with dementia in England. DAA encourages and supports communities and organisations across England to commit to the Dementia Declaration, and to take practical actions in order to help people live

well with dementia and to prevent crises. The DAA lobbies senior decision makers on issues related to dementia, disseminates research findings and best practice for all types of dementia, and for all ages.

Box 1.1: The National Dementia Declaration (2010).
Families living with dementia want a society where they are able to say:
1.   I have personal choice and control or influence over decisions about me.
2.   I know that services are designed around me and my needs.
3.   I have support that helps me live my life.
4.   I have the knowledge and know-how to get what I need.
5.   I live in an enabling and supportive environment where I feel valued and understood.
6.   I have a sense of belonging and of being a valued part of family, community and civic life.
7.   I know there is research going on which delivers a better life for me now, and hope for the future.

The Prime Minister's Challenge on Dementia (2012) set out a programme of work designed to make a difference in dementia care and research by 2015. The Challenge recognised that not only was dementia care and research a government issue, but it was one for society in general. In order to address this it was identified that there needed to be a growth in awareness, understanding and support of people living with dementia in local communities. The concept of dementia friends and dementia friendly communities was introduced.

The Prime Minister's Challenge (2012) focused on 3 key areas:

- Driving improvements in health and social care

- Creating dementia friendly communities that understand how to help

- Better research

Targets were set for each of these areas and included: improvement of the diagnosis rates of dementia; public health focus on risk reduction and measures to prevent dementia; having 20 dementia friendly communities and 1 million dementia friends by 2015; increase in the number of local DAA groups and additional funding for research.

In order to identify the future priorities for dementia research, the Alzheimer's Society and the James Lind Alliance set up a Dementia Priority Setting Partnership (Alzheimer's Society, 2013). Through this they sought active participation from people living with dementia, their family carers, organisations that represent people affected by dementia, health and social care practitioners. The participants were asked to reach a consensus agreement on what should be the top ten priorities for research (see Box 1.2). These covered the themes of prevention, diagnosis, treatment and care of dementia.

Box 1.2: Top 10 priorities for dementia research (Alzheimer's Society, 2013)

1	What are the most effective components of care that keep a person with dementia as independent as they can be at all stages of the disease in all care settings?
2	How can the best ways to care for people with dementia, including results from research findings, be effectively disseminated and implemented into care practice?
3	What is the impact of an early diagnosis of dementia and how can primary care support a more effective route to diagnosis?
4	What non-pharmacological and/or pharmacological (drug) interventions are most effective for managing challenging behaviour in people with dementia?
5	What is the best way to care for people with dementia in a hospital setting when they have acute health care needs?
6	What are the most effective ways to encourage people with dementia to eat, drink and maintain nutritional intake?
7	What are the most effective ways of supporting carers of people with dementia living at home?
8	What is the best way to care for people with advanced dementia (with or without other illnesses) at the end of life?
9	When is the optimal time to move a person with dementia into a care home setting and how can the standard of care be improved?
10	What are the most effective design features for producing dementia friendly environments at both the housing and neighbourhood levels?

In 2013 a State of the Nation Report on dementia care and support in England was published by the Department of Health to highlight where improvements had been made since 2012 and areas that were still in need of development. The areas identified for rapid improvement were: earlier diagnosis; improved assessment and treatment; more dementia research and better management and care of people with dementia. Unfortunately, the identified areas for rapid improvement were highlighted at a time of unprecedented pressures on health and social care budgets and a consequent reduction in available resources (Appleby *et al*, 2014). This caused significant pressures on health and social care providers, who had to make the choice of reducing services in other areas,

such as community services, respite and carer support, to meet the set targets for diagnosis in their area. This resulted in people getting a diagnosis, but with little post diagnostic support offered beyond this.

In December 2013, the UK hosted the first G8 Dementia Summit, which was held in order to seek agreement from the G8 countries (UK; USA; Japan; Canada; France; Germany; Italy; Russia) on actions to address the issues that arise from the prevalence of dementia. The G8 countries focused on improvements in: funding and innovation in dementia research, the prevention and treatment of dementia and quality of life for PLWD. The G8 ministers (now G7 since Russia exited the group) agreed 7 key actions (see Box 1.3) that their countries would invest in, and implement in order to address the issues faced by a growing number of families living with dementia.

Box 1.3: The G8 ministers agreed key actions (DH, 2013)
1. Set an ambition to identify a cure, or a disease- modifying therapy, for dementia by 2025.
2. Significantly increase the amount spent on dementia research.
3. Increase the number of people involved in clinical trials and studies on dementia.
4. Establish a new global envoy for dementia innovation, following in the footsteps of global envoys on HIV and Aids and on climate change.
5. Develop an international action plan for research.
6. Share information and data from dementia research studies across the G8 countries to work together and get the best return on investment in research.
7. Encourage open access to all publicly- funded dementia research to make data and results available for further research as quickly as possible.

Of the 7 identified key actions, the only one that specifically mentions involvement of PLWD is action 3. This is research focused and concerned with recruitment to research studies, rather than any attempt to understand or improve services and support from an individual's or family perspective. The focus on increasing investment in research was widely welcomed, but dementia research still receives 8 times less funding than cancer research (Alzheimer's Society, 2014). This could be because dementia is still considered to be a normal part of ageing (Bartlett & O'Connor, 2010). In addition, there has been a stigma attached to dementia, so people do not tend to talk about it and therefore it is not to the fore in public consciousness (Alzheimer's Research, 2015).

The focus of policy on a cure or disease modifying therapy raises the following two issues, which of the 200 subtypes of dementia would this be for? As most of these subtypes have different causes it would not appear feasible to have a single cure or disease modifying therapy. Secondly, this focus suggests that most of the research money will be directed to biomedical research and drug testing, which is most likely to be quantitative research with randomised trials. This could lead to less research money being available for qualitative research and studies focused on the lived experience of dementia and post diagnostic support issues. This focus on the biomedical model neglects the important role psychosocial, socio-political and cultural aspects have on the experience of dementia (Bartlett & O'Connor, 2010). Higgins (2012) states it is important to involve PLWD in research to explore their experience of living with dementia and their views on services and support and how they could be improved.

In 2015 the Prime Minister's Challenge on dementia 2020 (DH, 2015) was published. This Challenge set out 18 key aspirations that should be achieved by 2020 (see Box 1.4). It highlighted what the government wants to see in place by 2020 in order for England to be the:

- Best country in the world for dementia care and support and for people with dementia, their carers and families to live

- Best place in the world to undertake research into dementia and other neurodegenerative diseases.

Box 1.4: The main aspirations of the Prime Minister's Challenge on Dementia 2020 (DH, 2015)
1. Improved public awareness and understanding of risk factors and how to reduce them
2. Equal access to diagnosis & reduce wait for initial assessment to 6 weeks
3. GP's to play a leading role in coordination and continuity of care
4. All people with dementia to have meaningful care post diagnosis, which supports them and their family carer.
5. All health and social care staff to receive training on dementia appropriate to their role.
6. All hospitals and care homes to become dementia friendly.
7. Increase from 1 million to 3 million dementia friends in England.
8. At least 50% of people with dementia should be living in Dementia Friendly Communities.
9. All businesses encouraged and supported to become dementia friendly. All business to include dementia awareness in their induction programmes.
10. All national and local government departments becoming dementia friendly and having DAA involvement.
11. Dementia research seen as a career opportunity.
12. Funding for dementia research on target to double by 2025.
13. An international dementia institute established in England.
14. Increased investment in research from pharmaceutical, biotech and diagnostics sectors, small to medium enterprises, universities, research charities, NHS and private sector.
15. Cures or disease modifying therapies on track to exist by 2025.
16. More research made readily available to inform practice and pathways.
17. Open access to all publicly funded research.
18. Increased numbers of PLWD participating in research (with 25% registered to Join Dementia Research and 10% in research (4.5% currently).

Although all of the aspirations are very laudable, and if achieved will lead to some very positive changes for people living with dementia, without adequate funding and support they will not be able to progress to reality. Of the 18 aspirations: 6 are focused on awareness raising of communities, organisations and businesses; one is focused on diagnosis; another on prevention and risk reduction; 8 on research and only 2 on post diagnostic care and support. This is surprising as approximately 45% of PLWD are in the middle to late stages of dementia where they will need post diagnostic support and assistance from their family, local community or services to be able to live as well as possible with dementia (Alzheimer's Society, 2014). These families need help now as they are already living with the experience, it has already reached a stage where there will not be a cure or modifying treatment for them. A survey by the Alzheimer's Society (2014) revealed 43% of family carers and 52% of people living with dementia did not believe they received enough support.

In the NICE guidance on dementia, disability and frailty (2015) it is suggested that individual changes in behaviour and lifestyle could reduce the prevalence of dementia in later life. However it should also be noted that some risk factors cannot be prevented e.g. gender; genetic predisposition and age, so we are unlikely to envision a time when dementia can be significantly reduced or eradicated. In addition, changes in society are still needed to reduce the social inequalities experienced by some families, which could lead to preventable health issues later in life, including dementia. Studies of risk factors and preventative strategies should receive equal funding to that spent on the search for cure and treatment, as the money invested in prevention would not only

benefit a person's cognitive health but also their physical health, reducing the potential costs to the individual and society in general (NICE, 2015).

In order to understand how society views and treats people living with dementia, it was necessary to examine the policy context of dementia in the UK. Although there has been government focus on dementia over the last 10 years, it has produced guidance, recommendations and aspirations with little extra funding and support in order to put the plans in action. The focus on building awareness, reducing stigma and increasing research has been long overdue. However, families living with the complexities that can present in the middle to late stages of dementia have received less support in the community due to spending cuts in the NHS and local authorities. What extra money is available is being allocated to early diagnosis, dementia friendly communities and research rather than effective post diagnostic support.

1.4   Younger people living with dementia

As dementia is more prevalent in the older population there has been a tendency for services and support to be developed to meet their needs (Beattie *et al*, 2004). This has resulted in a system that does not adequately address the specialist needs of families living with young onset dementia (Armari *et al*, 2013). The issues faced by younger people living with young onset dementia are listed in Box 1.5.

Box 1.5: Particular issues faced by younger people living with dementia (adapted from Alzheimer's Society 2014a)

1. Have a rare form of dementia (Rossor *et al*, 2010)

2. Are more likely to have a difficulty receiving an accurate early diagnosis (van Vliet *et al*, 2010)

3. Are more likely to be misdiagnosed (Werner *et al*, 2009)

4. Be in work at the time of diagnosis or have recently lost their job (Rose & Yu, 2010)

5. Have heavy financial commitments including: mortgage and/or children at university (Jefferies & Agrawal, 2009)

6. Have dependent children (Beattie *et al*, 2004)

7. Have additional caring responsibility for parents (Arai *et al*, 2007)

8. Have a partner who still works (Rose & Yu, 2010; Allen *et al*, 2009)

9. Are more likely to be physically fit and active (Armstrong, 2003)

10. Are more likely to have a dementia affecting behaviour and social functioning in the early stages (Rossor *et al*, 2010)

11. Family members are more likely to report significantly higher psychological and physical morbidity (Rosness *et al*, 2011)

The causes of young onset dementia include diagnoses that psychiatrists rarely encounter, such as prion disease and multisystem failure (Baldwin & Murray, 2005) and as a result dementia in young adults can be misdiagnosed (Werner *et al*, 2009). Armari *et al* (2013) conducted research into the needs of people with young onset dementia and found early recognition and prompt referral was the principal area of improvement identified for services. Caregivers in this study also identified a lack of post diagnosis support as a significant issue. There is lack of specialist post diagnostic advice, support and services for younger families who have a member who has been diagnosed with bvFTD which may lead to increased family distress and social isolation (Lockeridge & Simpson, 2012).

One of the most common forms of young onset dementia is frontotemporal dementia (FTD). There are 3 main clinical variants of FTD (Hodges, 2007):

- Frontal or behavioural variant frontotemporal dementia (bvFTD) (also known as Pick's Disease)

- Semantic dementia

- Progressive non-fluent aphasia.

People diagnosed with FTD are more likely to display behaviour symptoms including disinhibition and apathy. Families find these symptoms difficult to cope with leading to feelings of stress and burden (de Vugt *et al*, 2006; Riedjik *et al*, 2006). The delay in receiving the correct diagnosis adds to the feelings of stress and burden (Luscombe *et al*, 1998).

Behavioural variant frontotemporal dementia (bvFTD) is characterized by changes in behavior and personality and the symptoms can include: inappropriate behavior; apathy; loss of empathy; obsessive or repetitive behavior; problems with concentration, decision making, planning and problem solving and appetite change (Rascovsky *et al*, 2011). In addition, about 1/3rd of the cases of bvFTD can be genetic. The progression of bvFTD can range from 2-20yrs, with people who are younger at age of onset having the fastest progression. In comparison with other types of dementia (see Table 1.2, section 1.2), the main symptoms of bvFTD appear to cause more distress in families (Mioshi *et al*, 2009, 2013; Nicolaou *et al*, 2010). However, there is a lack of research exploring how, why and when this occurs in the progression of bvFTD.

The majority of articles published about bvFTD tend to be biomedical, with a focus on: genetics (e.g. Liu *et al*, 2013; Ghosh & Lippa, 2015); biomarkers (e.g. Fernandez-Matarrubia *et al*, 2015; Galimberti & Scarpini, 2012); diagnosis (e.g. Rascovsky *et al*, 2011; Kaiser *et al*, 2013); pathology (e.g. Hornberger *et al*, 2012); biochemistry and treatment (e.g. Fernandez-Matarrubia *et al*, 2014; Riedl *et al*, 2014; Ghosh & Lippa, 2015). There is very little research into the experience of the person living with bvFTD and the effect on the family and social relationships (Werner *et al*, 2009; Johannessen & Moller, 2011; Daly *et al*, 2013; Oyebode *et al*, 2013). There is also a lack of research into a family's experience of the progression of bvFTD from pre diagnosis to the middle and late stages, and how social connectedness changes over this time. Research into the everyday experience of PLWD, and their families, could identify how, why and when social relationships and connectedness start to change. If this is identified, targeted interventions and support could be implemented at an early stage, to prevent family distress, and ensure the family lives as well as possible with bvFTD.

1.5 Family relationships

There is a paucity of research into the effects bvFTD on the family relationships of people diagnosed with dementia (Oyebode *et al*, 2013). The available research highlights the grief, loss, depression and burden families can experience when one of their family is diagnosed with a frontotemporal dementia, including bvFTD (Diehl-Schmid *et al*, 2013; Walters *et al*, 2010; Hellstrom *et al*, 2007; Evans & Lee, 2014; Kaiser & Panegyres, 2007).

Participants in Diehl-Schmid *et al* (2013) study stated the worst aspect of frontotemporal dementia was the loss of the loved one and the feelings of helplessness as dementia progressed, and the person became less like they were before. Some spouses in a study by Evans & Lee (2014) stated that they felt they had lost a partner and their spouse with bvFTD was a "stranger" but still present, a "loss of the person within". However, emotional disconnection from a spouse with bvFTD, could also be due to a form of self-protection and emotional detachment (Walters *et al*, 2010). Hellstrom *et al* (2007) described a process of gradual withdrawal or "moving on" as the last stage of couple-hood when a partner of someone with dementia starts to live a separate life as an "I" to have a new beginning.

Spouses of people with FTD can feel more burdened than spouses with other forms of dementia (e.g. Alzheimer's disease), and a possible reason for this could be the lack of support offered or received from services, family or the community (Riedijk *et al*, 2006). Also families who have a member diagnosed with dementia under the age of 65yrs could face additional stressors, such as: employment issues; financial pressures; dependent children and a rarer form of dementia (see Box 1.5). The studies by Diehl-Schmid *et al* (2013) and Kaiser & Panegyres (2007), identified burdensome behaviours of the person with bvFTD that caused the family members distress and depressive feelings, including: aggression; need for supervision; reduced manners; personality changes; misdemeanour; dependency; inflexibility; egocentric behaviour; irritability and reduced empathy. These behaviours also lead to family members feeling embarrassed in social situations, and so there is a tendency to avoid these,

leading to a reduction of social connections for the whole family (Hutchinson *et al*, 2014; Lockeridge & Simpson, 2012).

Families indicate that the behaviour changes and lack of empathy displayed by the person with bvFTD, even before diagnosis, can be particularly difficult to accept and cope with. These changes are frequently associated with a breakdown in the family relationship (Kipps *et al*, 2009; Hsieh *et al*, 2013; Massimo *et al*, 2013). Recognition of emotions on another person's face, and the ability to interpret cues in social situations, are compromised in bvFTD (Kipps *et al*, 2009). It is thought this might explain why families think their family member lacks empathy.

A study by Hsieh *et al* (2013) indicated that the behavioural symptoms of bvFTD (e.g. socially disinhibited behaviour, obsessive behaviour, lack of empathy) could lead to an increase in feelings of burden experienced by the spouse. The apparent lack of empathy and the behavioural symptoms in combination could explain why a marital relationship can break down when a spouse has been diagnosed with bvFTD.

Most relationships are characterised by a degree of reciprocity and as the family member starts to exhibit changes due to bvFTD, this reciprocity starts to become imbalanced (Massimo *et al*, 2013). As bvFTD progresses, the person with the diagnosis becomes more dependent on their family to meet their activities of daily living, and gradually the roles and relationships change within

the family (Oyebode *et al*, 2013; Walters *et al*, 2010; Hellstrom *et al*, 2007; Evans & Lee, 2014; Massimo *et al*, 2013; Harris, 2009; Lockeridge & Simpson, 2012). Evans & Lee's (2014) study identified that companionship and reciprocity reduced as dementia progressed, and changed the nature of the relationship from spousal to a parent-child one. However, Harris (2009) found that as well as role loss, changing social roles and role reversal there could be some positive changes in the marital relationship in some cases such as: personal growth; an appreciation of the spouse; appreciation of the "small things" of life; renewed commitment and a better understanding of their relationship.

Families develop strategies for coping with the family member diagnosed with bvFTD (Oyebode *et al*, 2013; Harris, 2009; Lockeridge & Simpson, 2012). In Oyebode *et al*'s (2013) study, it was identified that families managed and coped with the behaviours of their family member by: the use of humour; working around the lack of awareness; promoting quality of life; living in the present; recognising that behaviours were caused by bvFTD and not intentional; explaining and defending their family member when in difficulty. Families indicate that they need information, and psychosocial support (provided by trained personnel), to help them to understand and cope with the changes that they experience when a family member is diagnosed with dementia (Diehl-Schmid *et al*, 2013). In addition, counselling and specialist advice and support could enable the spouse to manage the changes they experience due to their partner's bvFTD (Kaiser & Panegyres, 2007). Increasing public and professional awareness of young onset dementia could reduce the lack of

understanding and stigma associated with dementia (Lockeridge & Simpson, 2012).

Relationships can give people support, happiness, contentment and a sense of belonging (Allan & Killick, 2008). From my experience of working with families living with young onset dementia and from the available literature, relationships reduce and deteriorate over the progression of dementia, so any positive benefits (e.g. fun and enjoyment; mutual interest; support and belongingness), tend to be reduced or lost whilst the negative aspects (e.g. discrimination, stigma and negative judgements), can be amplified. Specific education and training for primary care professionals could increase their ability to recognise the early signs of young onset dementia, and lead to a timely referral for specialist assessment and early diagnosis. Once diagnosed the person living with dementia should receive appropriate post diagnostic support, relationship interventions and support to enable the family to live as well as possible with dementia and to retain their social connectedness.

1.6   Social Connectedness

"Social connectedness refers to the degree to which a person experiences belongingness, attachment, relatedness, togetherness, or entrenchment in one's social relationships." (Santini et al, 2015 p.54).

This definition suggests that social connectedness is a subjective experience, and relates to a person's feelings of connectedness with others rather than an

objective measure. An individual could feel socially connected, with a sense of belonging, attachment and togetherness with a relatively small social network. However an observer may conclude that the same person is socially isolated due to a comparatively limited social network. Conversely an individual may have a large network of social contacts and may appear to have social connectedness, but in fact feel isolated and lonely.

Shared interests draw people closer together and lead to a feeling of social connectedness, which can contribute to a person's self- identity, and feelings of belongingness (Lee *et al*, 2001). Social connectedness develops through the lifespan (Baker & Baker, 1987). It begins with the parent-child relationship, which can confer security, attachment to other family members and belongingness. During adolescence peers, group membership and shared interests become more influential in the development of attachment, identification, belongingness and self-identity (Lee *et al*, 2001). In adulthood, the past experience of relationships that have shaped attachment, belongingness and self-identity can lead to a relatively stable sense of social connectedness (Lee & Robbins, 1998). Social connections evolve throughout life and individual's are shaped, and in turn shape, the context in which they live (Ong *et al*, 2013; Bidart & Lavenu, 2005). All of this however presupposes that the social context and environment are positive experiences from childhood throughout adulthood, but unfortunately for some people that will not be the case (Rook, 1984).

According to Baumeister & Leary (1995), people with low social connectedness often experience loneliness, anxiety, depression and low self-esteem. People who have high social connectedness are more likely to be sociable, participate in groups and activities, and generally enjoy the company of others, and are more accepting and tolerant of interpersonal differences within their social group (Lee *et al*, 2001). Williams & Galliher (2006) also suggest that social connectedness could have a protective role against the development of depression.

Research has suggested that a socially active lifestyle in later life could provide a protective factor in preventing dementia (Fratiglioni *et al*, 2004; Crooks *et al*, 2008). Holwerda *et al* (2014) conducted a three-year study, which examined the links between social isolation, feelings of loneliness and dementia. They found that people who identified themselves as lonely were more likely to develop dementia than those who did not. Lack of social connectedness can be problematic as people are often defined by their roles (e.g. parent, spouse, employee, friend or team mate) and the loss of the role can lead to the person living with dementia losing confidence and self-esteem, which can lead to feelings of social isolation and detachment from the world around them (Rose & Yu, 2010). In addition, MacRae (2011) states that people living with a diagnosis of dementia are not always given the opportunity to use their abilities or remain socially engaged due to other people's negative perceptions of dementia.

MacRae (2011) and Phinney *et al* (2007) suggest that families play an important role in enabling the person with dementia to retain their social

involvement and sense of identity. However, studies have indicated that families living with the effects of dementia are more likely to experience social exclusion, isolation and stress (Nay *et al*, 2015). Brodaty & Green (2002) suggests that family carers could also reduce social participation due to the added role and responsibilities of a carer's role. Families can be embarrassed by the behaviour and personality changes in their family member with dementia and may isolate themselves from their social network and activities, due to their reluctance to take their family member out publicly (Drentea *et al*, 2006). Family carers highly value the support from their social networks and contacts but at times they can be dissatisfied by the response from people they previously thought of as friends, citing their lack of understanding of dementia and the effects it has on families (Lilly *et al*, 2003; Hirst, 2005).

Nay *et al* (2015) indicated that a family carer's social participation changed when a family member was diagnosed with dementia and they identified four main themes of adaptation (see Table 1.4, overleaf). Nay *et al* (2015) recommend that family carer's needs for social participation are recognised and carer support is offered. They also state that public awareness of dementia is essential, so the person with the diagnosis, and their family, can remain actively involved in the community.

Table 1.4: Social participation and family carers of people living with dementia (Nay *et al*, 2015).	
Theme	**Effects on social participation**
1. Autonomy to choose when, in what ways and for what reasons to socially participate.	Spontaneity and planning are the key features of social participation and include: sports; interests; holidays; group activities, 1:1 and individual activities.
2. The impact of dementia care giving on social participation.	Reduced participation in activities as carer's role increases. There is reduced time for self. Family and friends may start to avoid the PLWD and carer due to stigma and behaviour of PLWD.
3. Employing strategies to maximise desired social participation.	Some family carers accept support and assistance from family, friends and services to enable them to retain or regain social participation. Some family carers do not receive (or are offered but do not accept) support and assistance and these carers are more likely to experience stress, depression, health issues and feelings of burden.
4. Establishing new meaningful connections, activities, attitudes, skills and knowledge.	Establishment of new activities, connections and social support. Increasing knowledge and skills in managing the challenges of the diagnosis of dementia. These new activities and supports may include: respite, psychosocial interventions, specialist groups, carer and peer support groups.

Nay *et al* (2015) suggest that some family carers do not progress past theme 2 and this can lead to increasing social isolation and distress. Ideally a family carer should establish new meaningful connections, activities, attitudes, skills and knowledge to enable the family to live as well as possible with dementia. In addition, Chien *et al*'s study (2011) states that carer support can have a positive effect on the psychological health of family carers.

1.7 Purpose of the study

I have observed throughout my career how relationships, social contacts, support networks and social connectedness of younger people living with bvFTD, appeared to reduce earlier in the progression of dementia, when compared with other forms of dementia. I also observed the effects that this had not just on the person diagnosed, but also on the rest of the family. By the time a family was referred to me, relationships had typically already broken down, with significant levels of distress displayed by all family members. The person with bvFTD had usually left their employment through redundancy, ill health or performance issues and therefore had lost contact with their work colleagues. In addition, the quantity and quality of friendships, interest and social groups, had changed for the person who was diagnosed and also for the rest of the family. This appeared to lead to isolation, anger, distress and feelings of abandonment. Families expressed that they felt "different", separate from the relationships and groups they used to belong to and estranged from other family members who did not understood the situation or who withdrew support at a time of vulnerability.

Once relationships, social contacts, social network, social support and connectedness reduced or ceased, it appeared that families found it very difficult to rebuild these, or to develop new contacts, networks or supports. In my research, I wanted to explore how and why social connectedness broke down both, within and external to the family, with the intention of finding out how this could be prevented or reduced. My initial search through the literature identified that this was an under researched area for younger people living with

bvFTD and their families (see chapter 2). What literature was available described this process in older people living with different forms of dementia such as Alzheimer's disease and vascular dementia, which do not appear to have the same effects on social connectedness as young onset dementia.

Having supportive relationships, contacts and networks can assist a person living with dementia and their families to maintain: quality of life; communication; roles; self-esteem; confidence and relationships, all of which are crucial to the wellbeing of the family unit. Despite national attempts to improve dementia awareness by the promotion of dementia friends and dementia friendly communities, younger people living with dementia and their families continue to face stigmatisation, discrimination and isolation due to a lack of understanding of young onset dementia (Rose & Yu, 2010). Some families report feeling shame and embarrassment about the diagnosis and the effects it has on behaviour and everyday activities of living (van Vliet *et al*, 2010). The stigma not only affects the spouse, but can also affect the children and young people in the household. They may feel embarrassed by their parent's behaviour, and so do not invite friends round to the house and

potentially isolate themselves from their peer group (Hutchinson *et al*, 2014).

There is a paucity of research into bvFTD and how it affects social connectedness. What is in evidence within the literature, from clinical experience and carer feedback is that the relationships, contacts, networks and connectedness of not just the person living with the diagnosis, but also the

family, considerably reduces. There have been no studies to date where the experiences of the family, the PLWD and external social networks, have been sought from the premorbid to late stages of bvFTD, in order to explore how and why social connectedness changed over time.

The key aim of this research is to provide an exploration of how and why social connectedness changes in families living with bvFTD. It is hoped this research study could add to the body of knowledge about the lived experience of bvFTD and the effects on social connectedness. It will also indicate implications for clinical practice and make recommendations for further research

1.8   Outline of the thesis

This thesis explores how and why social connectedness changes in people living with bvFTD and their families. Firstly, a review of the literature on social connectedness and frontotemporal dementia will be completed and discussed (chapter 2). From the review of the literature, I will analyse the gaps and inconsistencies in the literature and will devise research questions for my study. Social citizenship is the selected conceptual framework, and I will indicate why this is important for people living with dementia, and how it will be applied in my research study (chapter 3).

In chapter 4 I will identify, describe and provide a rationale, for why I selected the methodology, research design, methods and data collection. The ethical considerations will be explored, and best practice in conducting research with

people diagnosed with dementia will be followed. The data analysis methods will be identified along with the limitations and assumptions of the research study.

Chapter 5 outlines the study families' premorbid social connectedness. I analyse and interpret the findings from the data collection regarding social connectedness in chapter 6 (pre diagnosis and at the time of diagnosis) and chapter 7 (after the diagnosis).

In Chapter 8 I will discuss the findings collected in the study in order to answer the research question: *"How and why does social connectedness change in families living with the effects of bvFTD?"* From this I will identify implications for practice and areas for further study.

Chapter 2: Social Connectedness and Frontotemporal Dementia

2.1   Introduction

Studies have indicated that social relationships are a better predictor of the health status of a person than other biological and economic factors (Santini *et al.*, 2015; House *et al.*, 1988; Umberson & Montez, 2010). Holt-Lundstad *et al.* (2010) suggests that poor social relationships could be more harmful than lifestyle choices, such as excessive smoking, drinking and dietary intake or lack of exercise. As detailed in chapter 1, studies have suggested a socially active lifestyle in later life could provide a protective factor in preventing dementia (Fratiglioni *et al.*, 2004; Crooks *et al.*, 2008). However this has not been adequately researched in younger people with dementia.

As detailed in chapter 1 (section 1.6), the change in social relationships, and lack of social connectedness can be problematic for people living with dementia (and their families), as feelings of social isolation and detachment from their family and social network increase (Rose & Yu, 2010). In addition, the family of the person with dementia may be lonely and isolated due to their social network gradually reducing, leading to a reduced opportunity for social activities and support. This chapter discusses the findings from the literature search into social connectedness and frontotemporal dementia. From the review of the literature, research questions and methodology were designed for my research.

2.2   Literature search

An initial literature search was conducted in July 2013 prior to the commencement of the research and a follow up review was conducted in July 2015. Searches were completed through: CINAHL; Medline; PsycINFO; NELSON; Ovid Online; Proquest; SAGE Journals; ScienceDirect and Wiley Online Library. The search terms for social connectedness included: "Social connect*", "Social contact*", "Social relation*" and "Social network*". These terms were then combined with: "Behavioural variant frontotemporal dementia"; "Frontotemporal dementia"; "Frontal* dementia"; "Young* onset dementia"; "Young* person with dementia"; "Young* people with dementia"; "Pick's"; "working age dementia" and "Pre senile dementia". Full details of the literature search can be found in Appendix 1.

Inclusion criteria:

The following criteria were used to select studies/articles on social connectedness for inclusion in the review:

- Articles/study explored: social connectedness (social network*; social contact*; social connect*; social relation*) and young* person with dementia (behavioural variant frontotemporal dementia"; "frontotemporal dementia"; "frontal* dementia"; "young* people with dementia"; "young* onset dementia"; "Pick's"; "working age dementia" and "pre senile dementia").
- Study/article was published in English
- Articles were dated from 2005 (following the introduction of the National Dementia Guidelines and Strategy)

These inclusion criteria were used during the search to assess the abstracts and subject terms of identified studies to determine whether to access the full article/study. Each selected article/study was reviewed as to relevance to my

research, and their reference lists checked for any relevant articles that may have been referred to and where relevant, these were accessed.

Exclusion criteria:

Articles were excluded if they focused solely on other forms of dementia diagnosed in older age, and did not include consideration of social connectedness or citizenship. This decision was made as the findings in these articles may not be applicable to younger people diagnosed with frontotemporal dementia for reasons previously discussed in chapter 1.

The initial search terms yielded 388 articles. When these were accessed and sorted through, 355 of these articles were rejected due to a variety of reasons including a focus on: late onset dementia; diagnostic process; genetics; clinical treatments; epidemiology; clinical manifestations; other disorders; did not address the relationship between the person living with frontotemporal dementia and others (including family) and duplicate articles. Each of the 33 selected articles were retrieved and reviewed concerning the relevance for my research. On further examination 23 of the articles did not meet the inclusion criteria due to: the PLWD being over 65yrs of age (10); a diagnosis other than FTD (2); duplicate articles (4) and a central focus on interventions rather than social connectedness (7). The 10 relevant remaining articles were hand searched to identify any other articles, book chapters and reports referred to, and where relevant, these were accessed to support the review and thesis. Three of these articles were selected for inclusion, despite not clearly indicating the age or diagnosis of the participants, as the content of the article had a

significant focus on social connectedness and citizenship (Daly *et al.,* 2013; Ward *et al.,* 2011; Roland & Chappell, 2015) that was applicable to younger people living with bvFTD.

From my literature search, I identified that there is limited research into the social connectedness of families living with frontotemporal dementia when compared to dementias affecting older people, such as Alzheimer's disease. The studies that are available tend to focus on one perspective of the relationship, e.g. spouse/carer, rather than the family's or identified others (e.g. colleagues, friends). In most of the studies, the participants were in the early to middle stages of dementia and it could be proposed that the experience at this stage of the progression may be very different to the middle to late stages. In the following section, the 10 selected articles will be reviewed, and their study design, data analysis, findings and recommendations will be evaluated. This assisted in the formulation of the research questions, conceptual framework and methodology.

2.3   Social connectedness and frontotemporal dementia

In their review of the literature on how personhood has been conceptualized in dementia, Tolhurst *et al.* (2014) argued that psychosocial, environmental and cultural issues play an important part in the way society and people understand, and respond to the concept of dementia and those diagnosed with it. Personhood concerns a person's experiences, beliefs, actions and social

connections, which influence how a person lives with dementia. Harris & Keady (2009) suggested that when younger people are diagnosed with dementia, this can negatively affect their personhood and self-identity, as their roles as a family member, worker, social being and sexual partner change during the progression of the condition.

There is a tendency in society to identify dementia as a "disease of old age" and as such this can lead to services and support being developed for the older age group rather than younger people with dementia (Oyebode, 2014). There is an assumption by the National Health Service and Local Authority (who provide the majority of services and support) that the needs of younger people with dementia can be adequately met by the services for older people. However, it has been indicated that younger people with dementia and their families need specialist services focusing more on developing social contacts (Beattie *et al.*, 2004; Tolhurst *et al.*, 2014).

In the following subsections each principal aspect of the other 9 identified studies are described, evaluated and the main findings summarized.

2.3.1 Study themes, designs and analyses

Study themes

Table 2.1 provides an overview of the 10 relevant articles: their theme/focus; the participants; design; measures used and how the data was analysed. The

findings, recommendations and critiques can be found in Table 2.2. Only 2 of the 10 identified articles specifically focused on the social behaviour of people living with the diagnosis of bvFTD (Kipps *et al.*, 2009; Mendez *et al.*, 2014). Both were studies of social functioning, comparing the results of people diagnosed with bvFTD to Alzheimer's disease.

Table 2.1: Review of Literature- overview of papers- study design and analysis

Author	Theme/focus	Participants	Design	Measures used & Analysis
Nicolaou, Egan, Gasson & Kane (2010)	Identifying needs, burden, and distress of carers of people with frontotemporal dementia compared to Alzheimer's disease.	Australia 30 FTD carers 30 AD carers	Quantitative. Interviews with carers and rating scales.	Revised Memory and Behaviour Problems Checklist. Camberwell Assessment of Need for the Elderly. Zarit Burden Interview. Depression, Anxiety and Stress Scale. Analysed using SPSS- logistic and standard regression models. Chi square analyses. Mann-Whitney U test.
Tolhurst, Bhattacharyya, Kingston (2012)	Evaluates how emergent age-based factors may impact upon the experience of dementia.	UK Review of selected literature.	Review of selected literature.	Unspecified.
Mioshi, Bristow, Cook, Hodges (2009)	Factors underlying caregiver stress in FTD and Alzheimer's disease.	UK 108 caregivers (79 FTD & 29 AD)	Quantitative. Postal survey	**Patient-** Neuropsychiatric Inventory Questionnaire; Disability Assessment of Dementia. **Caregiver-** Perceived Stress Scale; Center for Epidemiological Studies Depression Scale; Social Network Index. Statistical analysis: independent t tests and logistic regression analyses.
Mioshi. Foxe, Leslie, Savage, Hsieh, Miller, Hodges & Piguet (2013)	The impact of dementia severity on caregiver burden in frontotemporal dementia and Alzheimer's disease	Australia 17 bvFTD carers 20 semantic dementia and 19 AD.	Quantitative. Carers completed rating scales. PLWD were assessed on rating scales by carer and clinician	**Patient-** Addenbrooke's Cognitive Examination Revised; Frontotemporal Dementia Rating Scale; Cambridge Behavioural Inventory Revised. **Caregiver-** Zarit Burden Inventory; Depression, Anxiety and Stress Scale; Social Network Index; Intimate Bond Measure. Statistical analysis: PASW statistics used to analyse the data. Normal distribution plotting using Kolmogorov-Smirnov tests. Analysis of variance. Multiple linear regression analysis.

Johannessen & Moller (2011)	To find out how people experience living with early onset dementia, and to assess the implications for practice and the development of further services.	Sweden 20 participants (12 men)	Qualitative. Grounded theory. Interviews.	Transcriptions analysed by both authors using grounded theory method (Corbin & Strauss, 2008) to identify core and sub categories.
Daly, McCarron, Higgins and McCallion (2013)	Presented theory explaining the processes used by informal carers of PLWD to manage alterations to their relationships.	Ireland 31 participants. 21 carers and 10 formal carers.	Qualitative Grounded theory. Interviews.	Analysis focussed on open coding to identify labels and these were compared and grouped to identify the principal problem "Living on the fringe". Then selective coding used to code actions carers used to address the principle problem. Memos used to enable tracking, development and refinement of the theory.
Ward, Howarth, Wilkinson, Campbell, Keady (2011)	Exploration of friendships of people with dementia in the early stages and the benefits of looking beyond the family as the support network.	UK One person living with dementia. Peer support group for people with dementia.	Qualitative. Illustrative case studies: 1 PLWD & Peer Support Group.	Case study: personal account. Peer support group- observation, discussions & interviews. Method of analysis not explicitly identified.
Roland & Chappell (2015)	Study explored the concept of meaningful activity as defined by caregivers of people living with dementia.	Canada 906 caregivers	Qualitative. Interviews.	Data from the interviews were transcribed into NVivo10 and categories and subcategories identified. Thematic content analysis used to understand how caregivers attribute meaning to activity participation outside of the home for PLWD.

Mendez, Fong, Shapira, Jimenez, Kaiser, Kremen & Tsai (2014)	The systematic observation of social behaviour during routine activities may be one of the best ways to distinguish patients with bvFTD from other different types of dementia.	USA 10 bvFTD 10AD	Quantitative. Observation of videotaped sessions.	1 hour videotaped sessions of a mealtime in the PLWD own home with their family carer. Two raters independently coded the videotaped interactions on a Social Observation Inventory. Data analysed using SPSS to calculate parametric and non-parametric differences between the groups (AD vs bvFTD).
Kipps, Mioshi & Hodges (2009)	Emotional, social functioning and activities of daily living in frontotemporal dementia.	UK 14 bvFTD 14 AD 16 age matched controls	Quantitative. Assessment scales.	Assessment scales. Disability Assessment for Dementia; Cambridge Behavioural Inventory (CBI); Emotion Hexagon; Addenbrooke's Cognitive Examination Revised; Clinical Dementia Rating Scale.

Study designs

Five of the selected studies used a quantitative approach using the following methods:

1.   Assessment rating scales (Mioshi *et al.*, 2009 & 2013; Kipps *et al.*, 2009).
2.   Assessment rating scales and interviews (Nicolaou *et al.*, 2010).
3.   Observation of videotaped sessions (Mendez *et al.*, 2014).

Four studies used a qualitative approach (e.g. Grounded Theory) using the following methods:

a) Interviews- (Johannessen & Moller, 2011; Daly *et al.*, 2013; Roland & Chappell, 2015)
b) Case study, observation, interviews and discussion- (Ward *et al.*, 2011).

Grounded theory is an approach recommended when investigating social problems or situations to which people must adapt (Corbin & Strauss, 2008). It enables the researcher to move from a description of what is happening to an understanding of the process by which it is happening (Corbin & Strauss, 2008). This approach was appropriate for both Johannessen & Moller, (2011) and Daly *et al.*'s (2013) studies, as they wanted to develop a broad theory and explanation of a process.

The studies by: Mioshi *et al.* (2009 & 2013); Kipps *et al.* (2009); Nicolaou *et al.* (2010); Mendez *et al.* (2014); Daly *et al.* (2013) and Roland & Chappell (2015)

were focused on the family carer's perspective and experience of frontotemporal dementia and excluded the "voice" of the PLWD. However, the focus on one person's perspective and experience provides a one-dimensional view of the topic, which lacks depth and limits the understanding of the topic studied. These studies also focused on carer stress and burden issues and excluded a consideration of the effects of psychosocial, environmental and cultural aspects on the whole family living with the effects of dementia. The research studies into carer's stress and burden by Mioshi *et al.* (2009 & 2013) and Nicolaou *et al.* (2010) could have been improved by further exploration of when, why and how carer's stress and burden levels changed in the progression of dementia.

The studies by Johannessen & Moller (2011) and Ward *et al.* (2011) explored the experience of being diagnosed and living with dementia and how the PLWD tried to maintain quality of life. However, as before, these studies provided a one-dimensional perspective on the topic, and could have been improved by including the PLWD family, or identified others, in the research study to increase the depth of understanding.

In Daly *et al.*'s study (2013) no details were given of the age, demographic background of the participants and no diagnosis was given of the person they were caring for. However, I included this study in my research, as it provided useful insights into how informal carers and person with dementia try to maintain their place in society despite stigmatisation and lack of understanding

of the condition. Once the data was collected, the researchers in the 10 identified studies selected various data analysis methods and these will be discussed next.

Study analyses

The quantitative studies by Mioshi *et al.* (2009 & 2013); Kipps *et al.* (2009); Nicolaou *et al.*, 2010); and Mendez *et al.* (2014), used statistical analysis tools to identify the results in each group (frontotemporal dementia (FTD) and Alzheimer's disease (AD). A comparison was made between the groups, to identify similarities and differences. These results were then utilised in the discussion to provide evidence of how the experience of carers differ as regards FTD and AD. However, the data collected was based on one person's perspective (the family carer or researcher), and the data was collected through rating scales, which do not provide contextual information or space for clarification. In order to provide a deeper understanding of the differing experience of dementia, a range of data collection methods and tools could be used and the perspectives of the person living with dementia, their families and friends collected.

The studies by Johannessen & Moller (2011) and Daly *et al.* (2013) analysed transcriptions to identify core and sub categories. Both of the research studies identified how they did this, with Daly *et al.* (2013) using a classic grounded theory approach (Glaser, 1978 & 1998) and Johannessen & Moller using the

reformulated approach by Corbin & Strauss (2008) both studies included details of how this was achieved.

Roland & Chappell's (2015) were transcribed into NVivo10 and identified categories and subcategories using thematic content analysis, to understand how caregivers attribute meaning to activity participation outside of the home for PLWD. They analysed identified themes in terms of content, frequency, co-occurrence (in the same respondent) and dementia severity. This enabled the researchers to identify the benefits of meaningful activity for the PLWD and family carer.

The findings and recommendations from the analyses of the data collected are detailed in Table 2.2 and discussed in section 2.3.2.

Table 2.2: Review of the literature: overview of papers- findings, recommendations and critique.

Author	Findings	Recommendations	Critique of the articles by the thesis author
Nicolaou, Egan, Gasson & Kane (2010)	Needs of carers of people living with FTD were significantly higher than AD. Elevated needs re: age of onset of FTD; financial aspects; symptoms of FTD; lack of access to specialist services, information and support. Female carers rating higher levels of distress than males.	Recommend: -Specific education and support programs - Raising community awareness and understanding - Tailoring domiciliary services and activities for FTD. -Specialist interventions for families living with the effects of bvFTD.	Needs more research into the sociocultural effects of FTD. Could have been improved by examining how, why and when distress and burden changed during the progression of FTD. The research focused on the needs of the carer and did not seek the perspectives of the PLWD or other family members or contacts.
Tolhurst, Bhattacharyya, Kingston (2012)	Very little literature explicitly addresses personhood with reference to young onset dementia. There are separate literatures on both personhood and young onset dementia but these two strands tend not to be drawn together.	Further research should be undertaken that has an explicit focus on personhood and young onset dementia.	More research on social cultural effects is needed. More focus needed on FTD. Literature search criteria not indicated.
Mioshi, Bristow, Cook, Hodges (2009)	Depression was a cardinal feature in FTD caregivers and accounted for more than 58% of stress scores. Both depression and stress was significantly higher than in A.D. Neither the severity of behaviour changes nor functional disability explained caregiver stress. Caregivers of FTD nursing home residents were still exhibiting high levels of stress, suggesting that other factors could account for high stress scores.	Recommend: - Health professionals need to have a better understanding of caregiver burden factors to ensure appropriate interventions are given.	Further identification of causes of caregiver burden and stress. Need longitudinal study to identify when, why and how stress and burden develops. Social cultural effects not explored.
Mioshi, Foxe, Leslie, Savage, Hsieh, Miller, Hodges & Piguet (2013)	Burden increases with disease severity in FTD. This study indicates that caregivers of bvFTD exhibit higher burden than other types of dementia. Study demonstrates that burden worsens with disease progression in FTD.	Recommend: - Interventions targeted at increasing caregivers understanding of changes in bvFTD beneficial as can help them to adjust their expectations and beliefs.	Sociocultural effects missing. Small groups studied. Needs a longitudinal approach to provide an in depth understanding of when, how and why burden increases for carers.

Johannessen & Moller (2011)	Described changes in the PLWD and the process of being diagnosed. Once diagnosed this research highlighted the PLWD's fight for dignity, and how they faced stigmatisation. PLWD in the study also faced a reduction in their self-esteem due to thoughts they were a burden, had lost their role with family, employment and with friends. Many of the participants felt lonely and lost sense of social cohesion.	Recommend: - PLWD opinions should be included in treatment plan. - Services should focus on helping PLWD keeping connected to others and society. - Work with employers to support PLWD in the workplace. - Increase awareness of society about dementia in the under 65 age group.	Specific diagnosis is not indicated so could not identify if FTD. All were in the earlier stages of dementia (4 months to 3 years following diagnosis). Social cultural effects could have been included.
Daly, McCarron, Higgins and McCallion (2013)	"Sustaining Place" theory identifies "place" and refers to a person's social identity and how they believe they are situated in social relationships and society. This can be affected by dementia related stigma and how society and its members understand and frame dementia (e.g. disease of old age; medical disease; debilitation and decline). The theory of "sustaining place" explains the social pattern of actions employed by informal carers to manage these issues on behalf of themselves and the PLWD.	Recommend: - Tailored interventions to address individual needs based on informal carers positions. - Nurses, other formal carers and society should engage in actions to support and enable social connectedness, social inclusion and citizenship for informal carers and PLWD.	Type of dementia and age of carers wasn't indicated. Carers were recruited from Alz. Soc. So carers not receiving support or services excluded from the study.
Ward, Howarth, Wilkinson, Campbell, Keady (2011)	Focus on friendships represents a broader shift in thinking and theorising associated with the condition. The model of service delivery and the role of the practitioner are being rewritten with a growing emphasis on co-productive working and the need to encompass the more diffuse nature of support and resources that exist for PLWD. Creates a sense not only of being supported but also of belonging and inclusion. Evolving role for practitioners and the on-going significance of their expertise as advocates and facilitators.	Recommend: - Co-productive working with PLWD. - PLWD should be actively involved in setting the terms of engagement and exercising greater control over how dementia is understood and responded to.	Replicability? Small sample size.

Roland & Chappell (2015)	Carers emphasise the benefits of social connectedness, physical health, and mental stimulation. They highlighted the importance of activity for both caregivers and people with dementia as well as the need to support caregivers to enable PLWD to continue to participate in meaningful activity, so improving the quality of life of both. Activity participation may be effective in improving PWD's sense of self, daily functioning, and the well being of the caregiver.	Recommend: - Meaningful activity participation for PLWD.	Not specifically YPD. What about bvFTD as services/ activities tend to exclude them due to their behaviours. Focused only on the carer's perspective.
Mendez, Fong, Shapira, Jimenez, Kaiser, Kremen & Tsai (2014)	Compared to caregivers and patients with A.D., patients with bvFTD were significantly disturbed in social behaviour. In contrast, patients with A.D. were indistinguishable from their caregivers. The lack of "you" comments and decreased tact and manners distinguished 92.6% of the patients with bvFTD from patients with AD and caregivers.	Recommend: - Clinicians need to identify social interactions outside of the clinic that disrupt lives to aid in diagnosis.	Medical model. Small numbers. Potential observer bias. Focused on diagnostic issues. No analysis of individual experience of family living with the effects of bvFTD.
Kipps, Mioshi & Hodges (2009)	The bvFTD group had significantly greater impairment in emotional recognition in others when compared with AD. BvFTD had a reduced ADL due to reduced motivation. Social dysfunction in bvFTD appears to be due to impairments in emotional processing and insensitivity to social cues and social disapproval. These impairments lead to significant carer burden.	Recommend: - To understand and appreciate the nature of the social dysfunction of bvFTD so the advice and interventions given to families are appropriate and effective.	Small number of patients. No observation of behaviours. Overreliance on rating scales.

2.3.2 Study findings and recommendations

Nine research articles were selected in the literature search and these were detailed in Table 2.1. From the findings in these articles three overarching themes were identified: i) The changes in social behaviour of the person diagnosed with bVFTD (Kipps *et al.*, 2009; Mendez *et al.*, 2014), ii) Caregiver stress and burden (Mioshi *et al.*, 2009, 2013; Nicolaou *et al.*, 2010), and iii) Changes in social support and connectedness for the person living with dementia and their families (Roland & Chappell, 2015; Ward *et al.*, 2011; Daly *et al.*, 2013; Johannessen & Moller, 2011).

i) Changes in social behaviour of the person diagnosed with bvFTD

The results of Mendez *et al.*'s (2014) observation study of the social behaviour of people diagnosed with bvFTD suggested that people with this diagnosis exhibit disturbed social behaviour (e.g. failure to attend to others or recognise their needs, decrease in tact and manners, improper verbal or physical acts), as compared with people with Alzheimer's disease. This study suggested that observation of social behaviours during routine activities, and in the person with bvFTD's own environment, is important for diagnostic purposes and also for understanding how their behaviours may be affecting the relationships and social connections they have with others.

People with bvFTD can be markedly worse at recognising emotion in others, particularly negative emotions (Kipps *et al.*, 2009). Impairment in recognising other people's emotions and a reduced ability to "read" social cues, may lead to

misunderstandings in social situations (Mendez *et al.*, 2014). This could provide a possible reason for the perceived lack of empathy a person with bvFTD displays when they see distress in a family member (Kipps *et al.*, 2009). Families tend to misinterpret this lack of response in emotionally charged situations as intentional, and evidence of lack of concern or affection, which results in distress, and distancing from the person with bvFTD (Kipps *et al.*, 2009).

The above studies have provided some valuable insights into the observed changes in social behaviour of a person diagnosed with bvFTD. However, they tended to focus on biomedical issues, such as rating behaviour and cognitions and the effects of bvFTD on the different brain regions. Due to the methods used in these research studies, there was an exclusion of the psychosocial, environmental and cultural aspects, which could lead to a partial picture of the lived experience of bvFTD.

ii) Caregiver stress and burden

Mioshi *et al.* (2009; 2013) and Nicolaou *et al.* (2010) focused predominantly on caregiver stress and burden rather than social connectedness, but they were included in the current review as they highlighted how the lack of social support and networks could increase feelings of distress, burden and isolation for family members. The researchers in these studies used a quantitative approach, which, it could be argued, does not adequately provide the understanding of the lived experience of the families and PLWD.

Nicolaou *et al.*'s (2010) study suggested that the needs for specialist advice and support by carers of people with bvFTD were significantly higher than for Alzheimer's disease due to: financial pressures; behavioural changes in bvFTD; lack of access to specialist services; inadequate information or support; lack of awareness and understanding of bvFTD. The studies by Mioshi *et al.* (2009 & 2013) to some extent mirror the findings of Nicolaou *et al.* (2010), indicating that family caregivers experience significantly more distress, and burden in their relationship with the family member diagnosed with bvFTD. However, two further interesting points were raised, firstly that family carers continue to feel a high level of stress (compared with family carers of other dementias) when the person with bvFTD is admitted to a care home. Secondly, the burden felt by family carers increases as bvFTD progresses.

The degree of caregiver stress and burden experienced by families living with a person diagnosed with bvFTD appears to be higher than with other more common forms of dementia such as Alzheimer's disease (see chapter 1, section 1.4). A possible reason for this could be, the lack of appropriate advice and support available to enable families to cope with the changes in behaviour and social functioning (see chapter 1, section 1.4).

iii) Changes in social support and connectedness

Families indicate that social support and connectedness changed for their diagnosed family member and themselves, over the progression of dementia

(Roland & Chappell, 2015; Johannessen & Moller, 2011; Ward *et al.*, 2011; Daly *et al.*, 2013). Johannessen & Moller's (2011) research explored the lived experience of young onset dementia, but it did not indicate what the exact diagnosis was. There are many different forms of young onset dementia (see chapter 1,Tables 1.1 & 1.2), and as mentioned earlier, the lived experience can be very different between one form of dementia and another (see chapter 1).

Findings from the Johannessen & Moller (2011) study highlight the changes noticed by the younger person with dementia pre and post diagnosis. The participants in the study identified the changes they noticed pre diagnosis, including memory problems and performance changes. They described how they attempted to find an explanation for the changes experienced, by visiting a medical practitioner. Participants indicated that some health professionals exhibited a lack of recognition of the early signs of young onset dementia and as a result the wrong diagnosis was initially given (e.g. burn out or depression). Once diagnosed with dementia, the younger people in the study reported there was a lack of advice and support to enable them to manage their condition and the feelings it engendered.

The participants stated dementia was a stigmatising and embarrassing disease, which affected their self-image and identity. They thought dementia was more stigmatising when it occurred before the age of 65, and this influenced how and when, they shared the diagnosis with others. Sharing the diagnosis could give the person with the diagnosis and their family the opportunity to retain their

social connectedness, and receive understanding and support from their social network. Participants in this study believed they lost opportunities for engagement in social activities and interests due to embarrassment regarding memory problems, reduced functional ability, and communication skills.

In Daly *et al.*'s study (2013), participants' highlighted dementia related stigma and the changes that accompany dementia progression, which Daly *et al.* (2013) titled "Living on the fringes". Daly *et al.* (2013) developed the theory of "Sustaining place", which describes how carers maintain their and their partner's position in society throughout the progression of dementia. This theory suggests the carer progresses through 4 interconnecting stages (Table 2.3).

Table 2.3: Theory of "Sustaining place" (Daly *et al.*, 2013)	
4 Stages of "Sustaining place".	**Progression through the 4 stages of "Sustaining place".**
"Unsettled place"	A family: • Notices a difference in their family member's usual functioning, e.g. memory, activities of daily living or behaviour changes. • Feels increasingly that their "world" has changed. • Copes with this by "explaining away", or finding possible reasons for these changes e.g. stress, physical reason, age. When this fails to explain what is happening and the changes become progressively worse, the family seeks understanding of why these changes are happening, by seeking a reason or diagnosis from a medical practitioner.
"Threatened place"	Once the diagnosis of dementia is given the family carer's in the study described how the social network and connectedness of the PLWD and themselves began to reduce. One of the reasons given for this was withdrawal from social contact, due to increased caring responsibilities by the family carer. In addition, family carers also reported that their social network withdrew or reduced due to the fear and stigma of dementia, and lack of understanding of what to do or say. Family carers stated that they felt they became a "lesser person", due to changes in their spousal role, becoming a carer, and the behaviour of other people in their social network towards the family.
"Sustaining our place"	In this stage, the family carer and PLWD attempt to maintain their place in society. They do this by "nurturing the PLWD", and trying to uphold their dignity by respecting their personhood and ensuring other people in their network and local community do so to. Family carers recognise their own limitations in caring and face the recognition that at times, they have to prioritise their own needs over the needs of the PLWD, and accept advice and support from others, in order to continue in the caring role. The importance of a positive support network was emphasised and joining peer support group or informal networks of other people in a similar position was found particularly helpful in reducing social isolation, and helped to develop a feeling of identification and belonging.
"Sustained place"	Family carers and people with living with dementia achieve "sustained place"; if they think and feel they have a sense of belonging, social connectedness and citizenship, within their local community and social network. Family carers, who achieved "sustained place", would often become a resource and guide for other families living with dementia. Some participants also advocated and campaigned for improved services and support, and challenged the misconceptions and stigma of dementia.

Whilst Daly *et al.*'s (2013) theory is useful in describing the social agency of the majority of family carers, this would need to be examined in more detail in families living with young onset dementia, and in particular, those families experiencing bvFTD. From my clinical experience, the family carer of someone living with bvFTD often avoids social contact due to the embarrassment or their fear of what their family member will do or say. This can lead the family to be stuck in the "Threatened place", especially if there is not the available specialist services and support to enable them to work through the feelings, and changes, they are experiencing.

Ward *et al.* (2011) explored the role of friendship through a single case study of an older person diagnosed with Alzheimer's disease. They also observed, interviewed and held joint discussions with a peer support group of older people with dementia. Although the sample size was small, and it was not clear whether the peer support group had any members with bvFTD, it made some interesting points about how friendships could be maintained, developed and new friendships made. It indicates that clinical practitioners should be advocates, or co-facilitators of peer support groups, to encourage participatory approaches and direct involvement of people living with dementia. This could increase friendship facilitation to enhance inclusion, belonginess, social connectedness and social citizenship.

Johannessen & Moller (2011) recommend services should focus on helping people living with young onset dementia and their families establish peer group connections. This may reduce the sense of isolation the PLWD and their family feel. It is also important for people living with dementia to maintain their social connections for as long as possible, so they continue to feel a part of society.

Younger people with dementia frequently lose their employment in the early stages of the condition (Johannessen & Moller, 2011). Employers should be encouraged to develop individualised work solutions with the person diagnosed with dementia, so they can continue in employment for as long as possible, with support as required. This could include having a job assessment to establish what changes need to be made in the workplace, or to their conditions of employment, to support the person living with dementia.

Roland & Chappell (2015) explored the concept of meaningful activity as defined by caregivers of people living with dementia by interviewing 906 family caregivers. Although the people diagnosed were older adults with dementia, there were aspects within the study that could be applied to younger families living with dementia. The research revealed 3 main themes (see Table 2.4).

I would concur with the findings in Roland & Chappell's (2015) study as in my clinical experience, often families are concerned by the PLWD's lack of meaningful activity during the daytime, and general lack of interest in things that they previously enjoyed. In addition, some family carers of PLWD seek outside

activities for their family member to enable them to receive some respite from caring, even if the activities are not actually enjoyed by the PLWD.

Table 2.4: Meaningful activity for people with dementia as defined by family caregivers (Roland & Chappell, 2015)	
Benefits for the person living with dementia.	Retaining social connectedness to family, friends and community. Connectedness and involvement in outside interests and meaningful activities are important to provide stimulation, enjoyment and enhance self-esteem of the PLWD. Maintenance of a social life, independent from the caregiver, in a safe environment where people understand dementia and its effects.
Benefits for the caregiver.	As dementia progresses relationship and roles can change within a family with one spouse adopting the carer role and the other the cared for. This can lead to an imbalance in the relationship. The act of being engaged in an activity together as partners rather than the carer and cared for can be enjoyable and provide a sense of fulfilment for both parties. Family carers get some respite from caring and engage in their own interests.
Activity as a negative experience.	For some PLWD leaving the home environment and going to a social situation or activity can be anxiety provoking. The personality changes and reduced motivation that some people with dementia experience, may also lead to a lack of interest in engaging with activities or hobbies they previously enjoyed. This can cause stress for the family carer, as they can feel guilty when encouraging the PLWD to engage in activities they no longer enjoy or resist attending, so they can get some respite time.

The lack of age and interest appropriate "meaningful activities" for younger people diagnosed with dementia, limits their social opportunities, connectedness, stimulation and enjoyment (Harris & Keady, 2004). If the activity or social group is not perceived to be of interest or to benefit the younger person they are less likely to want to attend, so their family carer does not get the respite or time to maintain their interests and social network.

Another specific issue for people living with bvFTD is that they are more likely to be excluded from a service or group, due to socially disinhibited behaviour, lack of awareness of and empathy for, other people's needs or emotions (Werner *et al.*, 2010).

There is limited research into the social connectedness of families living with the effects of frontotemporal dementia. It has been indicated in the literature that the experience of living with bvFTD can be more distressing for families than other forms of dementia, and can lead to negative effects on their social relationships and connectedness. The literature that was accessed through the current study's literature search, did not explore when, how and why social connectedness changes in bvFTD for the family or others.

2.4   Analysis of gaps in the literature

As identified throughout this chapter, there is a paucity of research into the experience of families living with bvFTD especially as to how this affects social connectedness. It could also be important to know why and at what stage social connections and supports change in order to identify, appropriate preventative strategies and supports, to enable families to live as well as possible with bvFTD, and to prevent a breakdown in relationships.

The research available regarding bvFTD (or FTD) and social connectedness, tended to focus on participants in the early to middle stages of dementia (e.g.

Nicolaou *et al.*, 2010; Mendez *et al.*, 2014). More research is needed in the middle to late stages of bvFTD to identify if there are significant changes in the social connectedness and relationships of the family at this stage.

In the identified research, when participants were interviewed, rated or observed, there was a tendency to focus on one time point in the post diagnostic stage rather than conducting longitudinal studies or examining the changes that have happened through the premorbid, pre-diagnosis and post diagnostic stages. It is necessary to have data from each of these stages in order to understand the process the families go through during the progression of bvFTD. From the information gained, appropriate advice, interventions and support can be devised for families effected by bvFTD, in order to prevent crises, build resilience in the family and ensure they can live as well as possible with dementia.

In the majority of the articles, only one person's perspective of social connectedness was explored, either the family caregiver (Nicolaou al010; Mioshi *et al.*, 2009 & 2013; Roland & Chappell, 2015), or the person living with the diagnosis (Johannessen & Moller, 2011; Ward *et al.*, 2011). If other people's perspective regarding bvFTD were also included in the research studies, it could lead to a more in-depth understanding of how and why social connectedness changed over time for people living with bvFTD, their families and identified others (e.g. friends, ex work colleagues and social network). There was also a limited range of methods used in the research studies

examined (see study designs p. 38). In order to increase the depth and breadth of the research, a range of tools could be used.

2.5   Identification of research questions

The literature search and review identified there were gaps in the literature regarding social connectedness and families living with the effects of bvFTD. In addition, the available literature did not fully explore how and why social connectedness changes when a family member has bvFTD. In order to achieve a breadth and depth of understanding of the topic, it was important to seek information from the person diagnosed with bvFTD, their family members and others who were identified as being in the social contacts and networks of the family. Consequently, the central research question in the current thesis was designed to provide information on these areas. The research question:

"How and why does social connectedness change in families living with the effects of bvFTD?"

The sub questions derived from this are:

1. "How and why does social connectedness change for people diagnosed with bvFTD?"

2. "How and why does social connectedness change for family members of a person with a diagnosis of bvFTD?

3. "How and why does social connectedness change for identified others (e.g. work colleagues; siblings; faith group; neighbours;

parents) who have a social relationship with a person living with bvFTD?"

2.6   Identification of methodology

As there has been so little research into how and why social connectedness changes in families living with bvFTD the current research study will be exploratory in nature, as it will be adding to the body of knowledge in order to assist in the development of models and theories as well as proposing areas for further research.

In order to address the current research question, I selected case study methodology for my research, as it is used to explore how and why questions, it requires no control over behaviours and it focuses on real life situations in contemporary settings (Yin, 2009). I needed to collect data from a number of families to explore similarities and differences in their experience of living with bvFTD, and to find out how and why their social connectedness changed over time. It was important that I gathered information from a variety of participants to ensure that I had as many perspectives on these changes, to build as comprehensive a picture of the family's experience as possible. In view of that, I selected the following methods for data collection: semi structured interviews; documentary review; diaries and participant observation to increase the depth of understanding of the topic.

2.7   Summary and transition

In summary, this chapter has indicated how I performed the search and review of the literature in order to identify articles on social connectedness and frontotemporal dementia.

The focus of my research is: *"How and why does social connectedness change in families living with the effects of bvFTD?"*

From the review of the literature, I identified gaps in the research (section 2.5) which are addressed in my research study via:

a)   The identification of families with a person diagnosed with bvFTD in the middle to late stages, to identify if there are significant changes in the social connectedness in these stages.

b)   An exploration of the changes to social connectedness that occurred through the premorbid, pre-diagnosis and post diagnostic stages of bvFTD.

c)   Utilisation of a case study approach, using a range of data collection methods to explore the perspectives of as many people as possible who are socially connected to the families living with bvFTD.

In chapter 3 (Conceptual Framework) I will introduce social citizenship as my conceptual framework, indicating why this is the most appropriate framework to use for my study.

Chapter 3: Conceptual Framework

"A conceptual framework explains either graphically, or in narrative form, the main things to be studied – the key factors, concepts or variables and the presumed relationship among them" (Miles & Huberman, 1994 p.18).

3.1 Introduction

A conceptual framework indicates why the research issue studied matters, and why the approach and methods used to study it are most appropriate and rigorous (Ravitch & Riggan, 2012). The framework links the current literature with the study's objectives to guide and structure the research process.

People living with dementia have the right to experience freedom from discrimination and have the opportunity to grow and fully participate in life (Bartlett & O' Connor, 2010). Having supportive social networks can assist a person living with dementia, and their families, to maintain: quality of life; communication; roles; self-esteem; confidence and relationships all of which are crucial to the wellbeing of the family unit. Despite national attempts to improve dementia awareness by the promotion of dementia friends and dementia friendly communities, younger people living with dementia and their families can still face stigmatisation, discrimination and isolation due to a lack of understanding of young onset dementia (Rose & Yu, 2010).

The majority of articles published about bvFTD tend to be biomedical, with a focus on: genetics (e.g. Liu *et al.*, 2013; Ghosh & Lippa, 2015); biomarkers (e.g. Fernandez-Matarrubia *et al.*, 2015; Galimberti & Scarpini, 2012); diagnosis (e.g. Rascovsky *et al.*, 2011; Kaiser *et al.*, 2013); pathology (e.g. Hornberger *et al.*, 2012); biochemistry and treatment (e.g. Fernandez-Matarrubia *et al.*, 2014; Riedl *et al.*, 2014; Ghosh & Lippa, 2015). This leads to a tendency to highlight deficits, disabilities or dependency, rather than explore strengths and capabilities, this can further strengthen the public's perception of dementia as a "disease of old age" and "something to be feared".

I considered using the notion of person centred care (Kitwood, 1997) as the conceptual framework to underpin this research study due to its widespread use as a guiding principle in dementia care. Kitwood (1997 p.7) defined personhood as:

"A standing or status that is bestowed upon one human being by others in the context of particular social relationships and institutional arrangements. It implies recognition, respect and trust."

Kitwood (1997) believed that stigma, and it's depersonalising and dehumanising effects, is the greatest threat to living well with dementia. He argued that dementia care should move from the medical model to a more holistic approach that recognises the person for who and what they are. In addition, there should be recognition of the PLWD relationships with others in society, as it is these that develop a person's self-identity and sense of

wellbeing. Kitwood's (1997) approach to dementia care identified that PLWD have a need for comfort, identity, occupation, inclusion, attachment and love.

Whilst I agree with Kitwood's (1997) approach, I rejected person centred care as my conceptual framework since Kitwood's theory was based on the study of older people who were diagnosed with severe cognitive impairment who, due to their severe impairment and dependence on others, were living in care facilities. The participants in my study were to be younger people, who were in the middle to late stages of bvFTD, the majority of whom would be community living. Secondly, Kitwood did not focus on sub-types of dementia, as he observed people with severe global impairment in care settings. Grouping all people with dementia together can lead to the belief that all are the same and issues such as age, socioeconomic status, gender, cultural background and sexual orientation may be overlooked (Downs, 2000). The danger of homogenising dementia as one experience, could lead to overlooking the complexities that some people living with dementia have as a result of inequalities, discrimination and lack of understanding in society. Finally, it has been proposed that person centred care overlooks the potential discrimination, and the lack of power over the external environment and society which families living with dementia can experience (Bartlett & O'Connor, 2010).

I selected social citizenship as an alternative framework, to underpin this research study as this provided a socio-political perspective for understanding dementia in a societal context. It also provided deeper insights into how the

person with dementia, and their family's, social connectedness and status changed both pre and post diagnosis. Social citizenship considers the rights, responsibilities, and status of people living with dementia and the power imbalances and discrimination that can be experienced. These are important issues for families, and have important implications for society as a whole. Government, academic, health and social care settings, emphasize that people diagnosed with dementia and their families should be involved as participants in planning and developing policy and services (Patient Participation and Involvement). However, there is a danger that this can become tokenistic with participation taken as people being present, rather than being actively involved in all stages of the process (Morrison & Dearden, 2013).

When considering an appropriate conceptual framework, the researcher needs to be aware that the conceptual framework selected could be influenced by their experience and knowledge. Once developed, the framework selected could influence the researcher's thinking throughout the study and may result in some findings being given prominence and others being ignored. In order to understand how my choice of conceptual framework influences the construction of the data, I engaged in reflexive analysis of my own positioning within the research (see Appendix 9). In this chapter, I discuss social citizenship as a conceptual framework, moving from the theory to how it underpinned the research study to explore how and why social connectedness changed in families living with bvFTD.

3.2　What is social citizenship?

Marshall (1950) wrote about citizenship, social class and the evolution of rights in England, and divided this into three stages in order to understand how it developed over time in the "West" (see Table 3.1 below).

Table 3.1: Development of citizenship	
18th century - civil rights	Right to justice; freedom of speech, thought and faith; liberty; property ownership.
19th century - political rights	Voting; right to participate in the exercise of political power.
20th century - social rights	Welfare and security; right to live according to a societies standards.

Marshall (1950) believed that citizens within a society have an image of what constitutes an ideal citizen. The citizens of that society measure other people according to that ideal and accord them the right of citizenship. In return, a society or community confers certain rights and responsibilities that can be expected by their citizens. His view of citizenship assumes that in order to be given the status of citizen, a person should be a full member of the community in which they live. However, it could be argued that Marshall (1950) was writing this from the viewpoint and experience of white working men in England, and the account is therefore not representative of all societies.

Bartlett & O'Connor (2010) state that Marshall's view of citizenship appears to exclude people living with dementia as they may not meet the expectations of an "ideal citizen" who, for example: votes; makes decisions, works and behaves in a manner considered "acceptable" by their community. This view of

citizenship also fails to recognise how an individual's expectations, wants and needs can differ from the rest of their community or to indicate how this can be accommodated (Faulks, 2000). Marshall's view of citizenship overlooks the impact of citizen driven campaigns for social change, as it suggests that the wider society sets the image of an ideal citizen rather than individuals or small groups of people. For example, campaigns by special interest groups have radically altered how society views people living with disability (Barnes & Mercer, 2010). The work by groups such as Dementia Engagement and Empowerment Project (DEEP) and Young Dementia UK, challenge societies prevailing culture, beliefs and behaviour regarding dementia.

The postmodernist approach to citizenship recognises and embraces, difference and personal experience. This approach indicates that the complexity of a person's life experience, the multiple identities and social roles they have (or had) is what influences how they are as a citizen (Faulks, 2000). Building on this, a working definition of social citizenship for dementia was written by Bartlett & O'Connor (2010, p.37):

"Social citizenship can be defined as a relationship, practice or status, in which a person with dementia is entitled to experience freedom from discrimination, and to have opportunities to grow and participate in life to the fullest extent possible. It involves justice, recognition of social positions and the upholding of personhood, rights and a fluid degree of responsibility for shaping events at a personal and societal level".

From personal and clinical experience, I have observed how a person can go into a medical appointment as a full citizen, with all of the consequent rights, responsibilities and respect, and come out after the diagnosis with a label, which reduces them to the inferior status as a "patient", and "sufferer". This label can change their experience of being a citizen, and a valued member of society, which can affect their self-identity and esteem (Burgener *et al.*, 2013). This can also affect the way they are perceived and treated by others (Zeilig, 2014). In addition, it can also alter the way other people perceive the family of the person living with the diagnosis, as a unit to be pitied, and set apart from the community due to their change in circumstance (Iliffe *et al.*, 2003).

3.3   Why promote social citizenship for people living with dementia?

The medicalization of dementia has led to a focus on dementia as a disease, on diagnosis, treatment and more recently, cure (Bartlett & O'Connor, 2007). Research on dementia over the last 30 years has tended to focus on the possible cause of dementia and identification of a cure or treatment. Much less research attention has been expended on quality of life issues, or the lived experience of the person with the diagnosis or their family (Bartlett & O'Connor, 2010). This may be because the large drug companies and research institutes have the most money to invest in research and as they tend to have a medical bias they are more likely to fund research into a cure or disease modifying treatment (Ridley *et al.*, 2014). It could be argued that these institutions also have a vested interest in finding a cure or treatment, not only to increase their

international profile, but especially in the case of drug companies, to find a marketable "solution" to the "problem of dementia".

The media, government and charities tend to focus on the negatives of dementia, propagating negative perceptions of people living with dementia being a drain on resources, and stoking concerns about the potential for a costly "dementia time bomb". This negative language used by the media can lead to the public having misconceptions of what dementia is and how it affects everyday life, and has contributed to the public fearing dementia more than other conditions such as cancer (Alzheimer's Society, 2014). These fears and misconceptions can lead to the stigmatisation and discrimination of the person living with the diagnosis (Burgener *et al.*, 2013). The common viewpoint of society is that everyone with dementia is the same, when in fact there are many different types and causes of dementia and many other factors that influence how a person lives with dementia e.g. life experience; social class; family cohesiveness and relationships (Bartlett & O'Connor, 2010).

Kitwood (1997) disputed that behaviour change was entirely due to neuropathological changes. Instead, he claimed that a person's experience of dementia is formed by the relationship between neuropathological changes, and social and psychological factors. He focused his work on the person living with dementia and wrote about personhood, and the need to understand whom the person was and where they come from to understand them now. Kitwood (1997) spoke about the importance of the environment and its effects on people

living with dementia, and how malignant social psychology can be present in society and institutions. Malignant social psychology can be defined as ways of interacting with people living with dementia that dehumanises or demeans them, which can be an intentional or unintentional interaction or behaviour. Whether intentional or not, this can lead to a negative outcome for the person living with dementia.

Critics of Kitwood (e.g. Downs, 2000; Phinney, 2008; Bartlett & O'Connor, 2010) suggest that his model locates people with dementia as recipients of welfare, and as passive within the care process, although it should be noted that he stressed the need for people with dementia to have a sense of agency. Phinney (2008) claims that Kitwood (1997) neglects the needs of employers, friends, and family in his theory of personhood, a theory that places dementia at an individual level, rather than a societal issue. On the other hand, Kitwood's theory, with its explicit emphasis on the damaging effects of malignant social psychology, is essentially an interactional and relationship-based approach. As Kitwood's model focused on older people who had severe cognitive impairment, the different presentation and experience of people in the earlier stages of different types of dementia were not clearly articulated. As discussed in chapters 1 and 2 of this thesis, dementia can present in very different ways depending on the type of dementia and how it affects the brain. There are also significant differences between the presentation and effects of dementia, depending on the age at diagnosis (see chapter 1, Box 1.5).

In response to the above criticisms of Kitwood's model, Bartlett & O'Connor (2010) devised a conceptual framework for social citizenship in order to extend understanding of the experience of dementia in a sociocultural context, and in interpersonal and social relationships. In their framework social citizenship consists of six components (see Table 3.2). There is a shift from the traditional viewpoint of the rights and responsibilities of citizenship, to one where the individuality of a person's experience, the context, multiple roles and identities are recognised and valued. As a result the person living with dementia is treated by society as a valued citizen.

Table 3.2: Six components of citizenship	
Growth	Hopes; aspirations; development.
Social positions	How multiple identities are maintained and developed; perceived status.
Purpose	Meaning in life and purpose.
Participation	Active involvement; decision-making; input valued.
Solidarity	Uniting with others to make a difference; social connections.
Freedom from discrimination	People with dementia can be denied access to certain services and benefits due to the diagnosis as others may assume they are not able to speak for themselves or make decisions.

According to Barnes *et al.* (2004) social citizenship can be measured in society, by the language that is used and the actions towards people who have a disability. In the UK, although there has been a focus on dementia awareness as part of the Prime Minister's Challenge on Dementia (DH, 2012; 2015), there

is still a lot of work to be done to change society's perception of people living with dementia, to prevent discrimination and loss of social connectedness.

In order to reduce negative stereotypes, stigmatisation and discrimination there needs to be a restructuring of the ways that people living with dementia are portrayed, or written about in the media, that avoid the use of negative terms such as "sufferers" or "patients". Our society has a tendency to think of people living with dementia as needing to be taken care of, and whilst that will indeed be true for some people with the diagnosis, it is by no means true for all. There are many examples of people living positively with the diagnosis, lobbying and working to dispel discrimination, stereotypes and myths, and improving services and support.

More recently there has been an increasing focus on awareness raising, and building a community spirit regarding dementia with the growth of dementia friends, dementia friendly communities and dementia pressure groups (DH, 2012; 2015). It is hoped that this will remove the stigma and discrimination that people living with dementia can face. This could also lead to an increased awareness by the general public and businesses in how they can support people living with dementia in their community.

Seddon *et al.* (2001) indicates that people living with cognitive impairment, such as dementia, may be seen as having a lower status than other physical disabilities when competing for funding and support. Adams & Bartlett (2003)

suggests this may be the case because, in general, the person with a physical disability is more likely to be younger when compared with someone living with dementia. This implies that we live in an inherently ageist society. The suggestion is that society needs to change, and that power imbalance and discrimination should be addressed.

3.4   How is social citizenship applied in this research?

Social citizenship was built on the work of Brooker (2004) concerning person centred care and Davies & Nolan's (2008) work on relationship based care. Bartlett & O'Connor (2010) included a socio-political context focusing on issues of power, personal agency, discriminatory practice and disempowerment. They identified 4 principles of social citizenship as applied to people living with dementia (see Table 3.3). These principles focus on the active involvement of people living with dementia in raising awareness, in order to challenge and change society's attitude and behaviour towards dementia.

Table 3.3: Four principles of social citizenship	
Maximising and valuing participation	Active involvement; people with dementia speaking for themselves; anti-oppressive practices; no tokenism; participatory research.
Facilitating growth and creativity in the dementia experience	Looking for the positives; strengths focused; relationship changes; developing interest.
Connecting personal experience to broader socio-political and cultural context	Exploring individual stories; interventions at a societal level; changing society's attitudes and behaviour to people with dementia.
Promoting solidarity by constructing a "we" community	Building a supportive community by developing political awareness; challenging society's stereotypes; campaigning and reinterpreting problems as shared experiences.

I used these principles in my study by: actively involving families living with dementia throughout all stages of the research; focusing on any strengths, or ways of adapting to the diagnosis the family used; exploring the lived experience of the families and their social connectedness, and recommending ways to build supportive communities based on the suggestions from the families in the study.

3.5   My reflections on social citizenship:

I selected social citizenship, as the conceptual framework as it was the best fit for the identified questions of my research. From my experience many of the issues families have living with dementia are due to: a lack of understanding; the behaviour of others; fear; stigmatisation and discrimination rather than simply the symptoms of dementia (Alzheimer's Disease International, 2012).

However social citizenship, as defined and described by Bartlett & O'Connor (2010), focuses on the person living with the diagnosis and does not fully consider the social citizenship of the family, and how dementia impacts on their lives and social connectedness. From my clinical experience, it is not just the person with the diagnosis who is affected, it is the whole family. Living with the diagnosis can have major implications on employment, finances, education, relationships, physical and psychological wellbeing and social connectedness.

Social citizenship describes very well how people living with the diagnosis in the early to middle stages of dementia can be maintained as citizens. However, some people with the diagnosis are very dependent on other people for all of their activity of living needs, and in addition may display some very complex behaviours due to the way dementia is affecting their brain, and so cannot engage with their local community or society in general.

3.6   Summary and transition

In this Chapter, I justified why social citizenship is a useful conceptual framework to underpin my research. In Section 3.1, I defined conceptual framework and how one can be used to underpin research. Section 3.2 explored citizenship, and the 6 components of social citizenship for people living with dementia. In Section 3.3, I examined how society's views of dementia have changed over time and the work that is still to do to increase public awareness, address the power imbalance and prevent discrimination. The application of social citizenship principles to my research is described in Section 3.4.

In Chapter 4 (Methodology) I will present the research purpose and questions, and identify why I have elected to use multiple case research to explore how and why social connectedness changed in families living with bvFTD.

Chapter 4: Methodology

4.1   Introduction and statement of the problem

Chapter 1 (sections 1.1 & 1.6) indicated why I decided to study social connectedness changes in families living with bvFTD. In chapter 2 I conducted a review of the literature, which indicated that there is limited research into the social connectedness of families, and how and why this changes during the progression of bvFTD. In order to establish a breadth and depth of understanding of the topic it is important to collect information from the perspectives of multiple stakeholders (Yin, 2009). This would include the person with bvFTD, their family members and others who were identified as being social contacts and in the social networks of the family. In addition, I wanted to collect the data in a naturalistic context e.g. social or home environment. The review of the literature pointed to important research questions, which I used to guide this study (see chapter 2, section 2.5).

In this chapter, I justify the research approach, the methods and data collection tools used (sections 4.2 & 4.3). In section 4.4 I outline the pilot study to be used in order to refine the case study protocol and data collection tools. The population sample and setting is identified, including the challenges of involving people with dementia in research (section 4.5) and the ethical considerations (section 4.7). The data analysis process followed in the research to generate findings, themes and assertions is discussed in section 4.6.

4.2 Research approach

During the design of the research, six ex-carers of people who had bvFTD and who attended a carers support group, were contacted by email to request a 1:1 meeting, to assist me to form my research focus. I asked the ex carers to highlight some of the issues they experienced with social connectedness when their spouse became symptomatic with bvFTD. Their responses helped to define the research purpose and questions, the methodology and the data collection tools used in the research.

4.2.1 Qualitative inquiry

The purpose of this research is to understand how and why social connectedness changes in families living with bvFTD. In order to study this in detail, a qualitative research approach was used due to its main features of: primacy of data; contextualisation; immersion in the setting; exploration of the "insiders" perspective (emic); detailed portrayal of the participant's experience; close relationship based on equality, mutual trust and researcher critical reflection (reflexivity) (Holloway & Wheeler, 2010).

Qualitative research methods are used when a researcher wishes to explore, understand and describe the way people make sense of the world and their experiences within it (Mertens, 2014). It is also used when the researcher wants to empower people to tell their story, whilst recognizing and trying to limit the power imbalance that can occur in quantitative research (Creswell, 2013). Whilst power imbalances can occur in qualitative research, the requirement of control over variables

that the researcher needs to have when conducting quantitative research increases the likelihood of imbalance.

Creswell (2013) states that there are 4 philosophical assumptions made by researchers when undertaking qualitative research: ontological (nature of reality); epistemological (what counts as knowledge and how it is justified); axiological (role of values in research) and methodological (process of research). These assumptions assist the researcher in identifying the problem, formulating questions and the methods used to seek answers to the research questions. For this study, I used the transformative paradigm because it aims to bring about social transformation and focuses on the lives and experiences of diverse groups that are marginalised, are facing inequities of power and explores how this is linked to political and social issues within a society (Mertens, 2014). This paradigm fits well with the social citizenship conceptual framework used to underpin this study, as it explores the everyday experience of bvFTD and addresses the potential discrimination, stereotyping, lack of understanding and stigma existing within a community and society in general (O'Sullivan *et al.*, 2014).

In the transformative paradigm, multiple realities are recognised (ontological assumption) and the acceptance of the different viewpoints as equally legitimate, is emphasised. However it also stresses that realities which seem "real" may in fact only appear so due to the influence of historical or cultural situations or beliefs (epistemological assumption). The values (axiological assumption) of the transformative paradigm emphasize the importance of

human rights, equality, social justice and ethical practice. In the current

research, transparency and reciprocity were important values practiced

throughout the research with the aim of sharing findings to add to the *"voice"*

for change and to increase social justice. I considered using a

phenomenological approach to examine the lived experience of the identified

families but I decided against this, as I wanted to explore social connectedness

in individual families, and across families, to explore differences and similarities

within and between them. In order to examine the multiple realities and the

different viewpoints of how and why social connectedness changes for people

living with bvFTD and their families, I selected case study research as my

investigative approach.

4.2.2   Case study research

"Case study is an in-depth exploration from multiple perspectives of the
complexity and uniqueness of a particular project, policy, institution, programme
or system in a "real life" context. It is research-based, inclusive of different
methods and is evidence-led. The primary purpose is to generate in-depth
understanding of a specific topic (as in a thesis), programme, policy, institution
or system to generate knowledge and/or inform policy development,
professional practice and civil or community action." (Simons, 2009 p.21).

Case studies are used to explore 'how' and 'why' questions, they require no

control over behaviours and focus on real life in contemporary settings (Yin,

2009). A case study approach was selected for the exploration of how and why

social connectedness changes in families living with bvFTD, because this

approach enabled me to perceive situations and events through the

perspectives of the participants. As case studies gather rich in-depth information about a person's lived experience within a real life context, it enabled me to gain an analytical insight into the topic studied (Stake, 2006).

In order to structure my research, a case study protocol was completed (see appendix 2). Thinking in detail about the data and how it would be interpreted, encouraged me to consider the different outcomes I might find, and to specify the potential causes for these. This led me to refine my data collection procedures so I could properly distinguish between the different reasons for the changes in social connectedness.

Thomas (2011) suggests that there are 3 main reasons for choosing a particular subject for case studies: key case (a classic case); outlier case (different to the norm) and local knowledge case (something within personal experience which you want to find out more). My research would be an example of a local knowledge case as I have experience of working with families living with bvFTD, but would like to explore how and why social connectedness changes throughout the progression of the condition.

As there has been so little research into social connectedness and bvFTD, this case study will be exploratory in nature, as it will be adding to the body of knowledge in order to assist in the development of hypotheses, models and theories as well as proposing areas of further research.

Case study research benefits from having a small sample size, which enables rich data to be gathered using a range of methods (Yin, 2009). The number of families selected for my study had to be limited due to the time and resource limitations of the study, and also because bvFTD is a relatively rare type of dementia, so there would be fewer families available who would be interested in taking part in research. I decided that I would recruit 5 families as this would be a manageable number, given the time and resources, and would provide the relevant data to address the set research question (Creswell, 2013). An explorative case-by-case analysis was conducted before cross case comparisons were made for differences or similarities. Each family was carefully selected so that they all had direct experience of a family member diagnosed with bvFTD, and all were at the middle to late stage in their progression with the condition (Yin, 2009).

According to Creswell (2013) there are 4 defining features of case studies: identification; intention; in depth understanding; data analysis and case description and identification of themes (see Table 4.1).

Table 4.1: Defining features of case studies (Creswell, 2013)	
1. Identification of a case.	In my study, a case is a family living with the effects of bvFTD and their social contacts (identified others).
2. Intention of conducting the case study.	In my study, there was an exploration of how and why social connectedness changed in families living with the effects of bvFTD.
3. In-depth understanding of the case by collecting a range of data by a variety of methods.	The methods used in my case study were interviews, observation, diaries and documentary analysis.
4. Data analysis.	In my study, 5 families living with the effects of bvFTD were selected and thematic analysis was used to analyse the data for patterns and themes.
5. Case description and identification of themes.	Themes were identified from each study family and then analysed across cases to explore similarities and differences. From the cross case analysis, patterns and explanations were devised.

Hodkinson & Hodkinson (2001) identified that a potential limitation of case study research is that the results are easy to dismiss by those who do not agree with the messages that they contain, stating that the sample was too small, not representative or the researchers were biased. Hodkinson & Hodkinson (2001) also indicated that case studies are not generalizable in the conventional sense and this can lead other researchers to make the claim that case studies have little value in research. Yin (2009) states that a single case study can expand or generate theory, to lead to "analytical generalization". A strength of using case study methods is the analytic generalizability achieved in electing to study multiple cases in-depth (Yin, 2009). Thomas (2011) argues that researchers do not always need to address population generalizability and cite that some of the most inspirational research is a result of case study research (Gomm *et al.*, 2000).

There is a limited literature (see chapter 2) suggesting that families living with bvFTD experience a reduced social network, and this research study seeks to explore this further. This research studied 5 families' experience of bvFTD and its effects on social connectedness in order to explore their similarities and differences with the available literature. It was proposed that the findings from the study would strengthen the claims for analytical generalisation.

As suggested by Yin (2009), the potential difficulties of case study research have been limited by carefully organizing the research by the use of a case study protocol (see appendix 2) to ensure the focus on the topic is maintained throughout the research, and with each case study family. In order to address the questions posed for the research, a range of methods were selected to collect as much in-depth data as possible including: semi structured interviews; diaries, documents and participant observation.

4.3   Methods & data collection

I collected data throughout the research study using a variety of tools. In the first meeting with the person living with dementia and their family, I conducted the semi-structured interview, planned the observation visits and explained how to use the research diaries (family carer only). In addition, I examined any relevant documentation regarding the PLWD or family. At the first meeting, the PLWD and family also identified other people (e.g. social contacts, other family members, ex work colleagues) who could be interested in being involved with the research (identified other).

4.3.1 *Semi structured interviews*

As this research is exploring how and why social connectedness changes for families living with bvFTD it is important to find out what participants think and feel about their experiences. One way of doing this was to conduct semi-structured interviews, which can provide in-depth data from a person's perspective (King & Horrocks, 2010). Interviews can be targeted directly onto the case study topic, but Yin (2009) suggests it is important that interviews should be guided conversations rather than structured queries, and this can be achieved by the use of a list of issues rather than specific questions. This list then acts as a reminder of the issues and reveals potential questions or probes (Thomas, 2011). The role of an interviewer in case study research is to collect data to answer the questions posed in the case study protocol, whilst at the same time conducting the interview in a neutral manner (Yin, 2009).

I decided to use semi-structured interviews for the families who volunteered for the research and key figures in the person's life outside of the family and originally 1 hour was allocated per person for each interview. A prompt sheet was designed in order to ensure that the research questions were addressed (see appendix 3). The semi structured interview prompt sheet was designed around the issues that were identified as important by families living with bvFTD and from the literature reviewed (Nicolaou *et al.*, 2010; Johannessen & Moller, 2011; Daly *et al.*, 2013). The topics in the interview prompt sheet examined the changes that occurred pre and post diagnosis that affected social

connectedness and included recognition of change, experience of bvFTD, social networks and support (Daly *et al.*, 2013; Kipps *et al.*, 2009).

This prompt sheet was piloted (see section 4.4) and minor changes made to the format. The use of semi-structured interviews enabled me to be flexible and responsive to what the interviewee said about their experience, and follow issues raised by them while enabling some control and direction for the interviewee (Tod, 2010). Once all participants gave informed consent, interviews were conducted, either in the person's own home, or in a setting of their choice. The choice of setting was important not only to maximize the response from participants, but also to select an environment that helped the PLWD feel safe and relaxed (Clarke & Keady, 2002). The interviews were digitally recorded and transcribed by the researcher as soon as possible after the interview, and in addition to this, field notes were written in a timely way after leaving the interview, to supplement the data recorded (Creswell, 2013).

Family members and the person living with bvFTD were interviewed separately so the participant being interviewed could say what they thought and felt without fear of upsetting other family member(s). This also ensured there was less chance of interruption or contradiction. Each person with the diagnosis of bvFTD was given the option of having an advocate sitting in the interview with him or her but none chose to exercise this.

Originally, I planned to allocate about 1 hour for each interview, in fact all of the interviews with the families took much longer than this (ranging from 1- 3hrs). Each family said it was the first time they could talk about everything that had happened to them with someone who would actually listen. Interviews with people living with the diagnosis tended to be shorter (10mins-50mins) in duration and were more difficult to conduct as the conversations had a tendency to be much shorter or to go off at a tangent to talk about something else in the environment, rather than the topic of the interview. I addressed this by talking about what interested the person and then gently guiding back to the research questions.

4.3.2 *Document review*

Documents typically consist of autobiographies, biographies, official documents and reports and can be useful in case study research as they give additional information that cannot be easily investigated by direct observation or questioning (Holloway & Wheeler, 2010). Primary source documents are written by people involved in the experience, action or event such as diaries. Secondary source documents are written about events at a later stage e.g. letters, minutes and plans. The data accessed through documents in the current study covered both primary and secondary data (Holloway & Wheeler, 2010).

Stake (2006) states that the usefulness of documents should be estimated in advance and time allocated for their review, as frequently not enough time or

attention is given to their inclusion. When I met with the families, the type of documents requested for additional information included medical letters; assessments; letters and notes. Permission was granted to use these documents as long as the content was anonymized. I examined the documents for themes to supplement the interview and observation data.

4.3.3 *Diaries*

It is important to capture the day-to-day experience of families living with dementia (Valimaki *et al.*, 2007). One way of capturing day-to-day experience involves the use of diaries. However diaries have rarely been used as a research tool in studying dementia and its effects (Valimaki *et al.*, 2007). This could be due to researcher's lack of understanding of their usefulness in researching day-to-day experience of dementia. Another issue could be that the method requires a degree of motivation from the diarist to continue writing the diary over a given period of time. This could be difficult for PLWD due to the progressive nature of dementia and the potential effects on concentration, motivation, word finding and working memory. According to Valimaki *et al.* (2007) diaries can be divided into five groups: i). Primary data collection; ii). Memory jogger; iii). A combined method of diaries and interviews; iv). Part of the triangulation process; v). Used to compare retrospective and prospective reports. In this study, 3 families kept diaries for a period ranging from 1 month to 1 year as part of the research study and they used them as a memory jogger, whilst I used them as a combined method of data collection and part of the triangulation process. The diaries were written by the spouse of the person

living with bvFTD in two of the families and the mother of a person living with bvFTD in the other. The families that did not write diaries started to do so but stopped writing in them as they found it to upsetting to write down what was happening to their family. Each family was given a template they could use to guide their diary writing or recording (see appendix 4) in order to structure their writing, but were told they could use any approach that worked for them as the template was just for initial guidance.

The diary enabled me to collect insights into the diarist's day-to-day experience,

and the inner thoughts and feelings about their lived experience. Diaries were useful to collect information that may have been forgotten in interview. The use of the diary also provided an opportunity for reflection on what had been written. This method is relatively unobtrusive and was completed when it was convenient to do so.

4.3.4 *Participant observation*

In addition to semi structured interviews, diaries and documents it was decided to conduct observation in this research study, as the purpose of the research was to understand how and why social connectedness changes in families living with bvFTD. However, after the first interview with the person living with bvFTD and their family it became rapidly apparent, in all five cases, that the social network was already significantly reduced for the family and especially

for the person living with the diagnosis. This would therefore limit the amount of observation studies that could take place outside of the family home.

Four types of researcher involvement regarding observation research have been identified (Hesse-Biber & Leavy, 2006): complete participant (being part of the setting and a member of the group studied e.g. covert observation); participant-as-observer (participant observation); observer-as-participant (an observer who only participates in so much as they are in the same location) and complete observer ("fly on the wall approach" whereby the observer is not a participant). I decided to collect participant observation data both as "participant-as-observer" and "observer-as-participant". This allowed me to observe the family, including the person with the diagnosis, in a social context to provide insight into interpersonal behaviour. The process and research purpose was discussed fully with all participants to gain informed consent.

Participant observation has its origins in social anthropology and is used to explore, understand and interpret a culture or group from an insider's perspective (Watson *et al.*, 2010). As a participant observer, the researcher's role is known to all study participants when they consent to being observed. It is important that the researcher knows what it is they want to observe so the observation becomes more focused. Patton (2002) suggests the researcher's overall objectives are to: gain an understanding of the setting; look for ways that participants organize themselves into groups and subgroups, observe the

interactions and communication (verbal and non-verbal) patterns; observe what participants do and how they behave. This form of observation was selected as I thought it would be better to actually participate in events rather than simply observe which, as Holloway & Wheeler (2010) suggests, could lead to people feeling uncomfortable or behaving differently. I was very aware not to lead or manipulate situations so I would not influence the outcome of the observed event. I did not use covert observation as I thought this was not ethical in the settings I visited.

I decided not to make notes during the observation session, as I did not want to prevent people from acting in their usual ways in fear of what I would be writing. Therefore it was essential to make notes regarding what I observed immediately after the event, using a document to guide my observations (see appendix 5). I used a strategy of progressive focusing by starting with description, focusing on the key areas that would address the purpose of the research and then selecting very specific issues observed that provided data for the research questions.

Overt observation was used in that I was open about the observation of the social situation and communication. I recorded these observations as soon after the observation event as possible, in detail, making objective notes about what was seen, heard and experienced using an observation template (see appendix 5).

4.4   Pilot study

A pilot study was planned and a pilot site was selected with geographical proximity. The Young Onset Dementia Team was contacted in the Midlands for volunteers to take part in the pilot study. Four families expressed an interest in being involved with the pilot, however, two withdrew due to changes in personal circumstances, and one due to a last minute change of mind. The remaining family stated that they wanted to be involved in formulating the research study as they felt very strongly that this was an under researched area, and they wanted post diagnostic support and awareness to improve for other families living with the effects of bvFTD.

The draft aim of the pilot was: *"An exploration of the effects of bvFTD on social connectedness"*. This was formulated in order to give some focus for the pilot study, provide discussion points and to assist with the formulation of the case study questions, design and data collection tools.

During the pilot study I interviewed the family using the interview prompt sheet in appendix 3, the observation method and template (see appendix 5) and the diary template (see appendix 4). The family was asked to give feedback regarding the purpose of the research, what the research questions should be and what data collection methods could be used to provide data. Following the

pilot study the feedback given by the family helped to formulate the research questions to focus on social connectedness (see chapter 2, section 2.5). The family stated that in their opinion the plan of using a combination of interviews, participant observation, diaries and documents, as utilized in the pilot study, was an effective way of collecting the necessary data to address the research questions, and they suggested no changes to the tools, or the application of these.

4.5   Involving people living with dementia in research

Up to the 1990's, the perspectives of people living with dementia were not actively sought in research studies, due to mistaken beliefs that they could offer nothing to the research process, or could not give reliable account of their experience (Downs, 1997; Murphy *et al.*, 2014; Smebye *et al.*, 2012). Instead, there was an over-reliance on seeking the viewpoints of others (e.g. family carer, spouse, or children) regarding the perspective of the PLWD, which could lead to inaccuracies as the family member could only assume what the PLWD felt or experienced (Downs, 1997; Hellstrom *et al.*, 2007).

The lack of active involvement in research of PLWD could also be argued to lead to exclusion, which further exacerbates their lack of value and regard in society (Dewing, 2002). In order to address this, authors and researchers have called for the active involvement of PLWD in research to improve our understanding of their experiences, and to increase rights to: equality; inclusion;

respect and autonomy (Slaughter *et al.*, 2007; Downs, 1997; Beattie *et al.*, 2004; Murphy *et al.*, 2014).

Hoe *et al.* (2005; 2007; 2009) argued that PLWD can be actively involved in research throughout the progression of the condition to the late stages. However, there may be potential challenges to address when involving PLWD who are in the late stages of dementia. This could include their ability to give informed consent and articulate their views and opinions verbally (Hubbard *et al.*, 2003). These need to be recognised and addressed to ensure that all parties benefit from the research and potential risks are reduced, and preferably eliminated.

Murphy *et al.* (2014) devised 4 key strategies for maximising involvement of PLWD in interviews and these include:

i) Gaining consent: which ideally should be gained face to face and should be a continuous process throughout the research to ensure consent is given (Dewing, 2007).

ii) Maximising responses: selecting an appropriate venue for the PLWD, building rapport, being flexible and selecting an appropriate style of interview can all help to maximise responses and make the experience more pleasurable for the person involved in the research (Hellstrom *et al.*, 2007; Hubbard *et al.*, 2003; Murphy *et al.*, 2014).

iii) Telling the story: Murphy *et al.* (2014) point out that when PLWD participate in research they may give very short, less detailed and unfocused answers so it is important to: include observations in interview data; interview while doing something else; or interviewing with prompts. Whilst additional data can be collected from family members or others it is suggested that care should be taken not to lose the voice of the PLWD (Murphy *et al.*, 2014).

iv) Ending on a high: Hellstrom *et al.* (2007) recommend that PLWD who are involved in research should be left feeling that they contributed positively to the research interview and not feeling they were a failure. The PLWD contribution should be recognised and valued, and they should receive appropriate feedback, both verbally and non-verbally. Time should also be spent with the PLWD after the interview talking about their interests to ensure they are relaxed and happy after their involvement in the research.

All of the above strategies were fully utilised within my research study to maximise engagement and involvement in the research not only with the PLWD, but also with other family members and participants in the study. When meeting the families and identified others who agreed to participate, I spoke about the purpose of my research, what it involved, how the data collected would be stored and used in my thesis (see Appendix 7) and a consent form was signed (see Appendix 8).

4.5.1   Sample

Stake (2006) suggested that between 4-10 cases would be the ideal number for multiple case study research to provide an adequate amount of data for research questions. BvFTD is a rarer form of dementia and people tend to lose insight into the effects it has on their social functioning, behavior and personality much earlier than other more common forms of dementia. In this research, five families was considered to be a realistic sample size to recruit, due to the comparative rarity of the diagnosis when compared with other forms of dementia, such as Alzheimer's disease.

Purposeful sampling was selected as the sampling framework as the research purpose was very specific about the sort of dementia to be studied (bvFTD). The principle inclusion criterion was families living with bvFTD. The principle exclusion criteria was families living with any other form of dementia. Consenting family members of the participant who had a diagnosis of bvFTD were interviewed, as were other consenting social contacts.

Participant invitations were sent out to the 'Younger People with Dementia' team based in the Midlands and they were briefed about the research and their roles in the recruitment process. Unfortunately, they did not have appropriate people living with bvFTD who could engage in the research. The national Frontotemporal Dementia Support Group (FTDSG) was approached and they sent out an email asking for volunteers to participate in the research. From this, five families came forward and after reading the study information, they agreed to participate in the research.

Five families agreed to participate in my research study and to be interviewed, observed and keep a diary during the study. In addition, each family agreed to contact any identified others (e.g. family, friends, contacts), who wanted to be involved in the study. Table 4.2 provides brief details regarding the 5 families' involvement in the research (for further details of the families, see chapter 5, Table 5.3). The PLWD selected for the research were in the middle to late stages of bvFTD. When I met each of the families, it became apparent, that the social connectedness of the person with bvFTD had already broken down, with limited contacts outside of the family home. This resulted in a reduced number of identified others available to participate in the research. Another issue that emerged when conducting the interviews, was the number of children who were undergoing counselling as a direct result of their parent's diagnosis. When invited to take part in the research, the adult children in the case study families declined to be interviewed (F2, F3, F5). The children in the Davies family (F4) were under the age of 16yrs, and as both were undergoing counselling, due to the trauma caused by their mother's condition, I decided not to involve them in the research.

The key strategies for the maximum involvement of the PLWD were followed (Murphy *et al.*, 2014). Despite this, there were communication difficulties with Diana Davies (PLWD, F4) and Brian Brown (PLWD, F2) due to reduced verbal expression, poor concentration levels and deficits in the working memory. These communication difficulties significantly impacted on the ability of the PLWD to maintain social connectedness.

Table 4.2: Brief outline of the 5 research families and people who were actively involved in the data collection (all details anonymised).				
PLWD	**Interviewed**	**Observed**	**Diary**	**Documents**
Andrew Allen **Family 1**	Andrew (PLWD) for 49mins. Ann (spouse) for 1hr 22mins	i) Farm shop café for 1hr ii). Barbers for 25mins iii) Singing for the Brain for 1hr. iv) Day centre for 15mins. v) Within their family home for 2hrs.	Ann wrote a diary for about 3 months.	a) Letters from medical practitioners.
Brian Brown **Family 2**	Brian (PLWD) for 10mins. Betty (spouse) for 1hr 23mins. (Interview arranged with 1 son but then he declined).	i) Within their family home for 2hr.	None.	a) Letters from medical practitioners.
Colin Cooper **Family 3**	Colin (PLWD) for 50mins. Carol (spouse) for 1hr. Family friend for 15mins. Tried to arrange interviews with the children but they declined.	i) Within their family home for 2hr.	None.	a) Letters from medical practitioners. b) Documents from past employment

Diana Davies **Family 4**	Diana (PLWD) non communicative Diana's mother for total of 3hrs . Diana's father for 2hrs. Diana's sister for 1hr 20mins. Darren (spouse) declined an interview.	i) Within care home (PLWD) for 1hr.	Mother wrote a diary for 1 year.	a) Letters from medical practitioners. b) Letters to and from family members. c) Letters to and from health and social care professionals.
Eric Evans **Family 5**	Eric (PLWD) Emma (spouse) for 2hr 35mins. Daughters declined to be interviewed.	i) Within their family home for 3hrs.	Emma wrote a diary for about 1 month.	a) Letters to and from health and social care professionals b) Letters from a family friend.

In the interviews, I established the social network of the family and they were then asked if they could approach the identified social contacts (identified others) to ask if they would like to be involved in the research. They were given information letters (see appendix 6), participation information sheets (see appendix 7) and consent sheets (see appendix 8) to give to identified others with my email address, and phone number so they could contact me directly.

The interviews and observational visits were held during the latter part of 2013 to early in 2015. The families who agreed to participate in the research were very willing to be interviewed, and observed in interaction with the person living with a diagnosis.

4.5.2 Data collection setting

All of the volunteer families elected to use their own home for the interviews. I supported this as I thought it important that the family felt comfortable and in control of the interview situation. I did have some initial concerns that it could prove difficult to conduct interviews if the environment was full of distractions, such as people walking in and out, telephone calls or other interruptions, but this did not prove to be the case in any of the interviews.

Three families agreed to the observation session taking place within the family home, but did not want the observations to take place in the community. Two families consented for participant observation to take place outside of the home environment, in external social settings. The external settings, included: hairdresser; singing for the brain session; care homes, café and local community. In the external settings I introduced myself, explained about the research and offered to answer any questions any of the attendees had. I also explained what I would be doing whilst in the setting, e.g. in singing for the brain and the café, I joined in the activity; in the hairdressers and care homes, I observed the interactions rather than joined in any activity (observer-as-participant).

4.6 Data analysis

According to Creswell (2013), in qualitative research, data analysis consists of preparing and organising the data for analysis, intensifying themes through a

process of coding and condensing, and then presenting the data. The researcher should establish patterns and look for a correspondence between two or more categories (Yin, 2009). The researcher should be seeking similarities and differences across cases.

All interviews and observations were transcribed verbatim, and read through several times whilst making notes in the margin and subjected to thematic analysis to identify:

- Issues that mattered to the participants

- The meaning of those issues

- The ways in which the researcher might characterise the participants stance in relation to the issues

Thematic analysis was used to analyse the data collected, including the diaries and documents, as this approach assisted me to identify patterns and themes in the collected data (Braun & Clarke, 2006). See Table 4.3 for the 6 phases of analysis.

Table 4.3: Six phases of analysis of qualitative research identified by Braun and Clarke (2006):	
Phase 1	Familiarising with your data
Phase 2	Generating initial codes
Phase 3	Searching for themes
Phase 4	Reviewing themes
Phase 5	Defining and naming themes
Phase 6	Producing the report

Atlas ti (data analysis and research software) was used to assist with the coding phase of the analysis. After the themes had been identified in the individual case studies (within case analysis) they were compared and contrasted with what was known from other research, publications or policy. The results of each case studied was also compared and contrasted to identify similarities and differences (cross case analysis).

4.6.1 Multi cultural validity & triangulation

Mertens (2014) states that the concept of validity is usually related to the data collection instruments used, but in the transformative paradigm she advocates that multicultural validity is a more important measure of validity. Kirkhart (2005 p.22) wrote about multicultural validity and states this is: *"Correctness or authenticity of understandings across multiple, intersecting cultural contexts"*.

Mertens (2014) identified 5 justifications for multicultural validity (see Box 4.1) and these have been applied throughout my research.

Box 4.1: Five justifications for multicultural validity (Mertens, 2014)
1. Theoretical stance underpinning the study.
2. Experiential- lived experience of participants.
3. Consequential- social consequences of actions from the study.
4. Interpersonal- quality of the interactions between participants.
5. Methodological- cultural appropriateness of the data collection instruments.

As detailed previously, I selected the transformative paradigm (detailed in section 4.2) and social citizenship as the conceptual framework (chapter 3) as they were the most appropriate theories to underpin the topic selected. To address the identified research questions, it was important to explore the lived experience of all of the participants regarding social connectedness and in order to do this I used multiple sources of evidence e.g. interviews, observation, diaries and documents and involved participants fully in each stage of the research (as detailed in sections 4.4 & 4.5). The data gathered from these were transcribed and then sent by email to the participants to review, make clarifications and comments. A follow up phone call was made following the review, clarifications and comments, and we then agreed the final transcript for analysis. Once the data was analysed and themes identified it was resent to the participants for their further review, clarifications, comment and approval.

Patton (2002) identified 4 types of triangulation in research: data triangulation (data collected from multiple sources); investigator triangulation (among different evaluators): theory triangulation (perspectives of the same data set) and methodological triangulation (methods). For this research, data triangulation was achieved by using a variety of data sources (families living with bvFTD and identified others). I also used a variety of data collection methods (methodological triangulation) including semi-structured interviews, observation, documents and diaries. I ensured that theoretical triangulation was achieved by checking transcripts, data, findings and themes with participants throughout the research and by comparing the findings with other research articles regarding bvFTD.

4.7   Ethical considerations

Ethical approval was requested and approved by the NHS Health Research Authority (NRES Committee- East Midlands (February 2013) and NHS Foundation Trust (April 2013). Following Health Research Authority guidelines (formerly National Research Ethics Service), I designed an information letter (see appendix 6) explaining the purpose and conduct of the research, and a participation information sheet (see appendix 7) for people diagnosed with bvFTD, families or identified others who were considering being involved in the research. A consent form (see appendix 8) was also devised to be completed if a person wished to be involved in the research, to ensure informed consent was given in each case using Health Research Authority guidelines (HRA). When I first met with the PLWD, I sought permission and consent to their involvement in the research and then checked their consent throughout the interview process as suggested by Dewing (2002)[2]. The families stated that at the time of diagnosis the person diagnosed with bvFTD and their family had consented to involvement in research studies. As all of the people living with bvFTD wanted to give their consent to future research studies at the time of their diagnosis, whilst they still had capacity to do so.

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[2] The original research proposal submitted to the Research Ethics Committee had a mental capacity assessment form to be completed with participants with bvFTD. I was advised not to use this routinely to assess capacity but to make a judgment throughout the research process.

These documents were written in terms that could be easily understood. It was explained that the aims of the research were to gather information about their experiences and views. It was made clear that participants could withdraw at any time without any implications and without being required to provide an explanation, as suggested by Walker *et al.* (2005), but none of the participants did so. It was explained that any information gathered during the research was kept confidential at all times and was suitably anonymised, following the code of practice for anonymisation of "rich data" (ICO, 2012). This proved to be challenging due to the small sample size, so special care was taken to keep all names, addresses separate from the collected data so families could not be identified by anyone other than myself. In the write up of the research, I have written the findings and discussion carefully so that families cannot be identified by others (Holloway & Wheeler, 2010). Where quotes have been used, informed consent was given by the person making the quote before I included it in the research. Handling and storage of data followed the guidelines set by the Data Protection Act (1998) and the Freedom of Information Act (2000).

As a nurse I am governed by the Nursing and Midwifery Council Code of Conduct (2015) whether in clinical practice or engaging in research. Throughout this research I followed the code to ensure I justified participant's trust by: treating people as individuals and respecting their dignity; acting with integrity; being open and honest about the research process and potential outcomes.

There are ethical implications for methodological choices when conducting research and these have been incorporated in this study in the following ways:

- The voices of people living with bvFTD and their family were listened to and used to inform the development of the research questions and design, data collection and analysis tools.

- Participants were informed that research findings and recommendations, could lead to greater awareness, understanding of bvFTD, into how and why this affects social connectedness, and identify implications for future practice.

- All participants were actively involved in working through the collected data, giving comments and clarification and helping to inform the findings and recommendations.

I was aware of the potential for power imbalance and used reflexivity at each stage of the research to limit this and to promote self-awareness and reflection. Throughout the research I reflected on the research process and kept notes in a written form in a diary. I also had reflective sessions with a colleague who has clinical and academic expertise. Following the reflective session the thoughts and insights gained were written in the reflective diary. These written notes were then used as a basis for the reflexive analysis (see appendix 9).

Each participant in the research (including the pilot) was given a copy of the: information letter (see appendix 6); participation information sheet (see

appendix 7) consent form (see appendix 8) to ensure informed consent. They were also sent a copy of the semi structured interview prompt sheet (see appendix 3) diary template (see appendix 4) and observation template (see appendix 5) to provide information about expectations to aid transparency and informed consent.

4.8   Summary and transition:

In summary, this chapter has discussed the research approach used (qualitative inquiry, transformative paradigm and case study research) linking this to the research questions (see chapter 2, section 2.5), relevant literature and conceptual framework. It then proceeded to identify the methods and data collection tools used (semi-structured interviews, documentary review, diaries and participant observation), in order to collect data to address the identified research questions. The population sample and setting was justified, with key strategies suggested for actively involving PLWD in research. Ethical considerations were also described, with details of how PLWD were listened to, and actively involved in the research. Thematic analysis was selected to examine the data collected and this would firstly examine the data from each family individually (within case analysis) and then these individual case studies would be compared and contrasted with the others studied (cross case analysis) to identify differences and similarities.

In chapter 5, the research families premorbid functioning is discussed and in chapters 6 & 7, I will present the findings and interpretation of the individual case studies and the multi case study analysis.

Chapter 5: Understanding the Families: Premorbid Social Connectedness

5.1 The research families

In order to ascertain whether the social connectedness of the families in the research study had changed since the onset of bvFTD, it was necessary to establish a baseline of the premorbid personality, behavior and social functioning, of the family member diagnosed as having bvFTD. As each person living with the diagnosis was unable to recall, or recount how they functioned in the time before symptoms (premorbid) due to the stage of bvFTD they had reached (middle to late stages), this information was gathered from their spouse (or in the case of Diana, PLWD, F4, from her parents).

A brief outline of each of the 5 families including timelines of: pre diagnostic changes, seeking and getting a diagnosis and after the diagnosis are included in Tables 5.1, 5.2 & 5.3. All names and identifying details have been changed to protect the privacy of the family members involved in the research. [3] A brief outline of the premorbid personality, behaviour and social functioning of the person living with the diagnosis of bvFTD (PLWD) in each of the families is given in Table 5.1 (overleaf).

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[3] N.B: F1= Family 1; F2= Family 2; F3= Family 3; F4= Family 4; F5= Family 5

Table 5.1: Brief description of the premorbid personality, behaviour and social functioning of the PLWD of each family in the study (reported by their spouse (F1-5) and parents (F4).	
Andrew Allen (Family 1): Andrew was in his 50's when diagnosed in 2010.	Controlling personality, liked to get his own way and if he did not he displayed verbally aggressive behaviour towards his wife. Held a senior position in the Armed Forces, which involved managing and leading large groups of staff and high-level formal social occasions. Had an active social life whilst working, revolving around Forces events and occasions.
Brian Brown (Family 2): Brian was in his 50's when diagnosed in 2012.	Conscientious and capable. Liked to help people if he could. Held a senior position in the civil service when he was employed but was made redundant in 2008. His job involved high levels of communication, leadership, management and socialising. Worked for 2 years in consultancy after he was made redundant. Had a close group of long-term friends who he socialized with regularly.
Colin Cooper (Family 3): Colin was in his 50's when diagnosed in 2013.	Quiet, easy going and competent. Held a senior position in the commercial sector when he was employed which involved high levels of cognitive ability, communication, leadership and socialising. He and his wife had a close group of friends who they went out with regularly to social events.
Diana Davies (Family 4): Diana was in her 30's when diagnosed in 2012.	Organised, compassionate and sensitive. Held a senior position in public service in an environment that needed a high degree of communication and interpersonal skills. Her life revolved around her husband, children and birth family but she was also very friendly with her work colleagues.
Eric Evans (Family 5): Eric was in his 50's when diagnosed in 2012.	Organised and very capable, but could be stubborn. Held a senior position in charitable sector, which involved a high degree of communication, coordination and interpersonal skills. Enjoyed socialising and communicating with people. Had a close group of friends who both Emma and Eric socialised with regularly. In addition he had a few close friends who he socialized with.

All of the PLWD were employed in senior positions before they retired (Andrew, F1 and Colin, F3), retired on ill health grounds (Diana, F4), or were made redundant (Brian, F2 and Eric, F5). Their employment involved high degrees of social interaction and advanced interpersonal skills (i.e. verbal and non-verbal

communication; listening; negotiation; problem solving; decision making and assertiveness).

"Eric's working role was very suited to him as he has always been a good communicator and his friendly manner meant he was well liked". (Emma, spouse, F5, interview).

"She was very, very highly thought of, very respected. She was methodical and organised and very good at her job". (Diana's mother, F4, interview).

Andrew (PLWD, F1) and Colin (PLWD, F3) were very successful, but their chosen professions had a fixed retirement age of 55yrs. Once they retired neither sought an alternative occupation outside of the family home, arguing that they did not need to do so financially. Roach & Drummond (2014) and Hasselkaus & Murray (2007) state that employment is not only important for financial benefits; it also builds and maintains self-esteem and identity. In both Andrew (PLWD, F1) and Colin's (PLWD, F3) cases their retirement had a significant negative psychological effect on them, despite the fact they received a generous pension. They withdrew socially, lacked motivation, became depressed and missed the structure of a work life. In addition, as their careers involved a substantial amount of overseas travel, their social network was geographically scattered, so when they retired their social life reduced as a consequence.

"When I was 55 they told me I had to stop working. I did 34yrs with them, I was in charge for 14 of these." (Colin, PLWD, F3, interview).

When I was interviewing and observing Colin (PLWD, F3) it was very apparent he missed his previous employment, and whenever he spoke about it he became very animated, and continued to get documents from his career which he showed me, and told me about events that had happened when he was at work. This was in marked contrast to his interaction levels during the rest of the 2 hour visit, where he was quiet and disengaged from what was going on around him.

Andrew and Colin's family's said that they became "lost" when they retired i.e. loss of motivation, withdrawal, reduced social connections and an initial reduction in social and leisure activities was observed:

"After he retired at 55yrs of age he just retreated into his shell, he didn't want to find another job. He didn't want to know anyone or do anything. 2-3 years down the line his behaviour was worsening." (Ann, spouse, F1, interview).

The importance of work to a person's self-identity and sense of purpose is supported by Bartlett & O'Connor (2010) in their work on social citizenship (see chapters 1 & 3). All of the PLWD's careers gave them meaning; structure and a sense of identity, and the families in the study felt that after they left their jobs

they changed very rapidly. They became less interested in socializing or being involved with the family or usual family activities or events.

The Allen (F1), Brown (F2), Cooper (F3) and Evans (F5) families did not observe any changes in their family member pre-retirement, or redundancy, and there were no reports from work colleagues regarding concerns about performance or social interactions. Brian Brown (PLWD, F2) and Eric Evans (PLWD, F5) were made redundant (2008 and 2009 respectively) as part of a company re-organisation within their workplaces. When examining the information from the families more closely, it is probable that (in their situations) there were changes in workplace behaviour, personality and interpersonal skills that could have influenced the employers' decision to make them redundant. This would appear to be supported in the case of Brian (PLWD, F2). When he was made redundant, he worked in consultancy for 2 years, and unfortunately he had to give this up as his family, and others, noticed significant changes in his behaviour and performance:

> *"He got consultancy work for a couple of years and that picked up and he certainly did everything very conscientiously, but he was starting to get confused because towards the end, I was getting phone calls asking where he was. He was going to the wrong school! His communication was very poor. I think there was a time when he called a child something at one point, which was out of character." (Betty, spouse, F2, interview).*

The spouses in the Allen (F1), Brown (F2) and Evans (F5) families were in paid part time employment, which involved a high degree of social interaction

outside of the family home throughout their husband's premorbid stage. They all stated they enjoyed working and were successful in their jobs. They particularly enjoyed the friendships, social connections and the independence having a career or occupation outside of the family home gave them. Indeed, satisfaction is a recognised and important part of being employed and Whitbourne & Whitbourne (2014) identify intrinsic factors (e.g. sense of identity; competence; personal growth and autonomy), and extrinsic factors (e.g. salary, hours, co-workers and status) of employment. The spouses in this study appear to have a strong sense of vocational satisfaction with a focus on intrinsic factors, whilst the PLWD in the study appear to have had as their main focus, extrinsic factors. It could be argued that as the spouses in full time employment were earning the greater amount of money in high status occupations, the spouse who had the part time role could do something they enjoyed intrinsically rather than seeking something that conferred a high salary and status. This differed in the case of F4, as Darren Davies (spouse) had a full time job until his wife (Diana, PLWD) was diagnosed with bvFTD, which he then gave up to become a full time carer for their young children.

Employees spend much of their waking hours working and frequently these work colleagues also become friends or part of the social group of the employee and may continue to be so even if the employee then moves to another job or career (Whitbourne & Whitbourne, 2014). Unfortunately, chronic and progressive health conditions can force major changes to a person's ability to continue in employment, which can negatively affect their self-identity, self-

esteem, and in addition reduce their social network (Johannessen & Moller, 2011).

All of the families in the study described their lives with respect to social connectedness before the onset of bvFTD (see Table 5.2).

Table 5.2: Premorbid social connectedness levels of families	
Family	**Social connectedness**
Family 1- Allen's Andrew (PLWD)	Andrew's family contacts were always strained. Was married previously but did not keep contact with his ex wife or children once they divorced. Did have a social life that revolved around the army in his working life, and the church once he retired. Very active in the local community. *Spousal relationship:* was very controlling towards Ann.
Ann (Spouse)	Ann has close family connections. Had an active social life revolving around work, village and church activities. *Spousal relationship:* Andrew was very controlling and did not show affection to Ann. Had separate lives and social connections.
Family 2- Brown's Brian (PLWD) & Betty (Spouse)	Both Brian and Betty had very close family connections. Had an active social life revolving around work and close friendship group. *Spousal relationship:* Strong relationship, very close, activities together. *Relationship with children:* Very close relationship based on nurturing, caring and joint activities.
3 sons (late teens-early 20's)	Enjoyed a loving and supportive relationship with both parents. Had an active social life including school, sporting activities, friends and family activities.
Family 3- Cooper's Colin (PLWD) & Carol (Spouse)	Both Carol and Colin had close family connections. Had an active social life revolving around work and a close friendship group. Enjoyed do it yourself, golf, socializing with friends and attending the gym. *Spousal relationship:* Strong relationship, very close, activities together. *Relationship with children:* Very close relationship.
Son and daughter (over 20yrs)	Both married (son has a child). Both have active marital, family and social lives.
Family 4- Davies	

Diana (PLWD)	Diana had close family connections. Had an active social life revolving around work and a close friendship group. *Spousal relationship:* Strong relationship, very close, activities together. *Relationship with children:* Very close relationship.
Darren (Spouse)	Relationship with his own family has been more distant although are in contact. Relationship with his wife's family good on the whole but could be strained at times due to wife's closeness to her family. Social activities and contacts with close friends but mostly revolved around his young family. *Spousal relationship:* Strong relationship, very close, activities together. *Relationship with children:* Very close relationship.
2 sons (under 16yrs)	Very close to both parents, grandparents, maternal aunt and her family. Spent a lot of time with grandparents as both parents worked.
Diana's parents	Very close relationship to both daughters and their families. Had an active social life revolving around work and a close friendship group. *Spousal relationship:* Strong relationship, very close, activities together.
Diana's sister	Close family connections and close relationship with her sister. Had an active social life revolving around work and a close friendship group. *Spousal relationship:* Strong relationship, very close, activities together. *Relationship with children:* Very close relationship.
Family 5- Evans Eric (PLWD) & Emma (Spouse)	Eric and Emma had close family connections. Had an active social life revolving around work and a close friendship group. *Spousal relationship:* Strong relationship, very close, activities together. *Relationship with children:* Very close relationship.
2 daughters (in their 20's)	Both daughters had a very close relationship with both parents. Enjoyed usual family activities together. Both had active social lives.

What linked the families was the PLWD's success in their relevant careers at a senior position, which involved formal and informal socialising and a high level of interpersonal skills. Families stated that the premorbid high level of competence regarding performance, behaviour and interpersonal skills, contrasted sharply with the changes the PLWD displayed both pre and post diagnosis.

People living with bvFTD in the study described how they enjoyed their careers and the financial and social benefits that they gave them. In all cases, it was clear on interviewing the PLWD, that they missed their work life and the social aspects of this. With the exception of Diana (PLWD, F4), the social contact with work colleagues had significantly reduced pre diagnosis and all but ceased after the diagnosis was given. The family said this was partly due to the changes in behaviour, personality and social functioning of the PLWD, but they also thought that ex-colleagues no longer had things in common, and did not know what to say or do, so found it easier to avoid the family. Pre diagnosis, Diana told the work colleagues not to visit and hid away from them. When asked why she did this, she could give no reason, other than to say she did not want to see them.

The families involved in the research study also detailed the timeline of the pre diagnostic, diagnostic and post diagnostic changes of their family member with bvFTD (see Table 5.3).

Table 5.3: Details the timeline of pre diagnostic, diagnostic and post diagnostic changes of the participants in this study.			
Participants	Pre diagnosis	Getting a diagnosis	Post diagnosis
Family 1: Andrew Allen (PLWD) Ann Allen (wife)	2000- Andrew retired and Ann noticed his socially inappropriate behaviour. 2005- Ann noticed Andrew's inappropriate behaviour was worsening and he was also getting repetitive and forgetful. She considered leaving Andrew.	2010- Ann went to the GP and expressed her concerns over Andrew's behaviour. The GP told her nothing was wrong. 2010- Ann went back to the GP repeatedly with lists of Andrew's behaviour issues causing concern. GP eventually did a memory test and said all was okay. 2010- Ann contacted the GP again, in crisis,	2010- Visited by a psychiatrist following SPECT scan told Andrew had bvFTD, and told Ann to look it up on the internet and discharged them back to GP. 2010- Ann saw a programme about Singing for the Brain and searched on the internet for local service. 2010/11- Ann made repeated visits to the GP to get post diagnostic support. 2011- Community mental heath nurse (CMHN) visited for 1 year until she left her job.

	2007- Ann noticed that Andrew was becoming increasingly obsessive about collecting things.	and he referred Andres to a research clinic after Ann's insistence. 2010-Andrew sent for SPECT scan after Ann insists the GP do something and Andrew is given the diagnosis of bvFTD.	2012- given a replacement for the CMHN but this proved unsatisfactory. 2012- Andrew attended a day centre but due to his socially inappropriate behaviour asked not to attend. Now attends a nursing home for day care.
Family 2: Brian Brown (PLWD) Betty Brown (wife)	2008- Brian made redundant from his job. Took this badly. 2008-2010- Brian worked in consultancy. All was well initially but he then began to get confused about where he should be and his communication reduced towards family and others. 2010/11- Brian's decision making became problematic, was making errors of judgement with finances, became repetitive and obsessive about money.	2010/11- Brian's behaviour continued to deteriorate and this was noticed by family and friends. Betty took him for a health check as she thought he could be severely depressed. 2012- The family were on holiday in the UK and Brian went missing for 48hrs. Slept for 2 nights on a park bench near to a police station. Eventually he went into the police station and they contacted the family. 2012- Betty took Brian to the GP. Brian had a scan and this showed some cerebral atrophy. A second scan performed a few months later showed a marked atrophy in the frontal lobes and a diagnosis of bvFTD was given.	2012- No post diagnostic support given for 7 months after the diagnosis. As self funding had to find a "sitter/carer" independently. 2013/14- In contact with Alzheimer's Society for carer support and services. Brian tried a day centre but they said they were not suitable for younger PLWD. Betty referred to Social Services for a carers assessment. 2013/14- Betty attends a carers group when possible to fit this in with employment. 2014- Betty will contact the Alzheimer's Society again to see if there are any Singing for the Brain or Carers Groups on days when she isn't working.
Family 3: Colin Cooper (PLWD) Carol Cooper (wife)	2011- They went on holiday with friends, noticed Colin wasn't communicating as before and would behave in a disengaged way. Carol and the friends thought he was probably going deaf. Colin wouldn't go and see the GP.	2012- Colin and Carol went to the GP and she noticed Colin's behaviour and she thought he could have a brain tumour. So Colin was sent for a scan, which revealed shrunken frontal lobes. The GP diagnosed Alzheimer's Disease. 2012- Colin had several driving incidents and crashes and was told he could not drive. 2013- Daughter was	2013- No post diagnostic support given for 7 months after the diagnosis. As self funding had to find a care and support independently. 2014/present- Monday- Golf Wednesday & Thursday- Carer- Gym Friday- Day Centre Monthly- brother Every 2 weeks Colin visits son and daughter.

		convinced her father had bvFTD from his behaviours and scan results and insisted on a review. Colin was then diagnosed with bvFTD.	
Family 4: Diana Davies (PLWD) Darren Davis (husband) Diana's parents.	2011- Christmas-family noticed a change in Diana's behaviour especially towards the children and the rest of the family. When asked about this she said she had workplace stress. March 2012- Phone call from Diana's manager to her parents, as she was worried about Diana's behaviour at work. The consultant in the team thought Diana could have a frontal tumour. March 2012- Diana went to the GP and they said "take it easy". April 2012- Diana's behaviour was deteriorating further and saw GP again who prescribed antidepressants.	April 2012- Another phone call from Diana's manager asking her mother to collect her from work as her behaviour was causing significant concern. Assessed by Mental Health Team and they diagnosed stress. May 2012- Darren stated he couldn't cope with Diana's behaviour and she was admitted to a Mental Health Ward. She had an MRI scan revealing "global shrinkage". May 2012- Diana transferred to a neurological hospital. Behaviour continued to deteriorate. Lumbar puncture and EEG-nothing abnormal detected. Discharged to home address initially, but then Darren phoned Diana's parents in distress as he couldn't cope with her behaviour. June 2012- Took Diana back to hospital where she was admitted to admission/obs. ward. Mother of Diana said to neurologist she thought her daughter had Pick's and they said no she is too young. Transferred to a neurological hospital. Behaviour continued to deteriorate. Given the wrong diagnosis. Darren was convinced his wife had a mental disorder not a neurological one and was annoyed that her	November 2012- Darren states he doesn't want Diana home due to her behaviour and the effects on their sons. He wanted her to be admitted to a care home out of county, as he didn't want her to be near the boys. 2013- present-Relationship between Darren and Diana's mother is a little better now communicate by email or text but still somewhat strained and only have limited contact with one grandson.

		family were still seeking a diagnosis and treatment. Diana discharged home. July 2012- Readmitted into a mental health ward. Relationship between Diana's family and son in law broken down. August 2012- Diana transferred to a neurological unit as her behaviour continued to worsen. October 2012- Diana's family sought a second opinion from a Professor in London. MRI scan revealed "massive destruction of the frontal lobes of the brain."	
Family 5: **Eric Evans** **(PLWD)** **Emma** **Evans (wife)**	2007 to 2009- Eric's father moved into a home in 2007 and died in 2009 and the family noticed that despite having a very close relationship with him, Eric showed no real emotion or empathy. 2009- Eric was made redundant from his job. 2010- Eric's behaviour became more noticeably different to family and friends. He was derogatory, lacking in empathy, stubborn and was lacking insight into this. Eric's mother died but he displayed no real response to this. Family thought he could be depressed. 2011- Relationship with family and	May to August 2012- Eric seen by the GP and he referred Eric to the specialist neurologist. Seen by a specialist neurologist who confirmed the diagnosis of bvFTD following an assessment and an MRI scan which revealed "marked atrophy of the frontal lobes of the brain". Recommended a social services referral, anti depressants and Outpatient Appointments.	2012-13- initially 6 monthly outpatient appointments, reduced to annually and then discharged. 2012-13- further deterioration in Eric's behaviour including increased disinhibition; double incontinence; self neglect; aggressive behaviour; sexually inappropriate behaviour; reduced concentration and teeth grinding. October 2012- Carers attending to Eric's needs 5 days per week. Emma covering all other times and ensuring he is safe, clean and is able to live in the community. 2013- Applied for Continuing Healthcare funding as an increased package of care required to meet Eric's increasing needs. 2014- Eric admitted into a care home as the package of care in the community broke down due to his behaviours. 2014- present- Relationship between Emma and Eric good now she doesn't have the full time carer role. Their daughters visit their Dad now and the relationship is improving

behaviours worsened and he moved out into a rented flat saying he wanted a divorce. Then went to Australia to visit a friend. Whilst there it became apparent he was not coping and his behaviours were starting to get him into trouble outside of the family home. February 2012- Family decided Eric could no longer cope with living independently but he would not move back to the family home. Agreed to get a flat near to his family.		

The families described their struggle to get an accurate diagnosis and once this was received, the difficulty in accessing appropriate post diagnostic advice and support. This will be explored further in chapters 6, 7 and 8.

All of the families in the study had a strong sense that their family member was an active and valued part of the community and social network before the onset of bvFTD. The study families justified this by referring to their family member's pre-morbid functioning which included: successful careers with social status; hopes and aspirations for the future; a sense of purpose; positive self-identity; social connections and active involvement in their local community. These

correlate with the 6 components of social citizenship (see chapter 3, Table 3.2) described by Bartlett & O'Connor (2010).

5.2 Summary and transition

In summary, this chapter presented and interpreted data from the interviews with the families, regarding premorbid social connectedness. It highlighted the similarities and differences between the five families in this research study. All of the people diagnosed with bvFTD had very successful lives before they displayed the symptoms of the condition. They were achieving all 6 components of social citizenship as they had: social positions with status; a sense of purpose and scope for development; active involvement with their family and community and social connectedness. In chapter 6, (Findings and interpretation: social connectedness pre diagnosis and at the time of diagnosis), I will analyse and interpret the data collected from the research study to answer the research questions whilst considering possible alternative interpretations. A comparison will be made with what I have found in my research study with other available literature on the topic.

Chapter 6: Findings and Interpretation

Social Connectedness Pre Diagnosis and at the time of Diagnosis

6.1 Introduction to the findings and interpretation chapters

To fully understand how and why social connectedness changes for people living with bvFTD and their families, it was important to examine the pathway from the premorbid to the middle/late stages of bvFTD. For the purposes of investigation and analysis in this research, the social connectedness pathway has been identified as consisting of:

1) Premorbid social connectedness (see chapter 5).

2) Social connectedness pre diagnosis and at the time of diagnosis

3) Social connectedness after the diagnosis

The pathway became the central organising concept for the analysis and interpretation of the research findings, and was derived from families in the study. Six themes emerged from the thematic analysis:

i)   "Why are they behaving like this?": *Finding a reason*

ii)   "What is happening to our family?": *Changing family relationships.*

iii)   "Other people are noticing"

iv)   *"Getting a diagnosis".*

v)   "Now we know what it is".

vi)   *"Grief and loss".*

The stages of the pathway, and identified themes, were shared and reviewed with the five participating families, to ensure they accurately reflected their experience, and all the families agreed that they did. This commonality was surprising given that the families in the study had a varied background and experience. Table 6.1 highlights which themes were identified in each stage of the social connectedness pathway, and the key sub-themes that are discussed in each chapter.

Table 6.1: The Social Connectedness Pathway of the families participating in this study		
Central Organising Concept	**Themes**	**Key sub-themes**

Chapter 6:
 	
Social connectedness pre diagnosis and at the time of diagnosis
	Theme 1:
	"Why are they behaving like this?": *Finding a reason*
	
	Theme 2:
	"What is happening to our family?": *Changing family relationships.*
	
	Theme 3:
	"Other people are noticing."
		-Changes in personality and behaviour of PLWD
		-Employment
		-Perspectives of family and others on why they are like this.
		- Changes in social connections
		- Changed relationships:
		  -Spouse
		  -Parent-child

	Theme 4: "Getting a diagnosis."	-Birth family - Experience of health service.

Chapter 7:
 	
Social connectedness after the diagnosis
	Theme 5:
	"Now we know what it is"
	
	Theme 6:
	"Grief and loss"
		-Post diagnostic support
		-Changes in social connections and relationships:
		    -Spousal
		    -Family
		    -Social contacts and networks
		-Changes in social citizenship

The families identified that their lives and experience centred around three stages: before bvFTD started (premorbid stage); fighting to get a diagnosis (pre-diagnosis and at the time of diagnosis) and what happened after the diagnosis. These will be discussed in more detail, with this chapter covering the pre-diagnosis and time of diagnosis stages, and chapter 7, the time after diagnosis. Each of these chapters will discuss the findings and interpretations from the data collected from the 5 families, and include selected quotes to examine how and why social connectedness changed throughout the progression of bvFTD.

6.2   Introduction to chapter 6

As detailed in chapter 1, research into bvFTD has indicated people in the early stages start to exhibit changes in their personality, behavior, social functioning and empathy levels (Rascovsky *et al*, 2011). These changes can affect social connectedness due to misunderstandings about why the person is acting as they are, with the interpretation by others, that these observed changes are intentional. Without a diagnosis, such changes cannot be linked to dementia.

The behaviour and quality of life of a person diagnosed with dementia is not only influenced by biological changes in their brain but also by past experiences, situational context, sociocultural aspects, social interactions and how they are perceived by others (O'Connor *et al*, 2007). The participants in my study had careers that provided them with a sense of social status, financial stability, self-esteem and identity. After their family member ceased their career and the symptoms of bvFTD commenced, the families felt they had to alter much of their everyday life and family plans, hopes and aspirations. In addition, the families reported changes in their social position, due to the loss of employment and the associated salary. After the loss of their career, families reported that the person with the symptoms of bvFTD missed their usual routine and sense of purpose and they became withdrawn and lacking in motivation. They also started to reduce participation in social events and reduce contact with their usual social network, especially work colleagues. This had the effect of reducing the PLWD's social connectedness and altered their 6 components of social citizenship (see chapter 3, Table 3.2).

With the benefit of hindsight, families think that these behaviour and social changes may have been due to a combination of a sense of loss after they left their career, and also the early stage changes of bvFTD. Table 6.2 provides a brief description of the changes in personality and behaviour exhibited by each PLWD pre diagnosis. The changes occurred gradually over time, and families initially reported compensating for this by adjusting their behavior (e.g. avoiding arguments) and adjusting family routine to one favoured by the person with bvFTD.

Table 6.2: Brief description of the changes in personality and behaviour of the PLWD leading up to the diagnosis of bvFTD (as reported by their spouse; and in letters and diaries).

Family 1 (F1): **Andrew Allen** (Changes took place gradually since 2000)	Increasingly socially disinhibited; lack of regard for people around him; obsessive about collecting things; decision making compromised; only communicating about things he was interested in and not listening to others; irritability; personality traits intensified; singing aloud in public inappropriately; withdrawal from others and dependence on wife for activities of living.
Family 2 (F2): **Brian Brown** (Changes took place gradually since 2008)	Lack of regard for people around him; did not show any emotional reaction to his mother's death; obsessive about collecting receipts and money; decision making compromised; made unsound financial decisions; significantly reduced verbal communication; irritability; personality change; withdrawal; socially inappropriate behaviour; disappeared on a family holiday for 48hrs. Increasing dependency on his wife.
Family 3 (F3): **Colin Cooper** (Changes took place gradually since 2011)	Lack of regard for people around him; obsessive about the weather and switching electrical appliances off; decision making compromised; numerous driving offences; significantly reduced verbal communication and then only on topics he is interested in talking about; irritability; personality change; withdrawal; socially inappropriate behaviour; increasing dependency on his wife for activities of living.
Family 4 (F4): **Diana Davies** (Changes took place rapidly since 2011)	Lack of regard for people around her; did not show any emotional reaction to her families distress; obsessive about electrical switches; decision making compromised; significantly reduced verbal communication; irritability; screaming and wailing; over eating and craving for sweet things; personality change; withdrawal; socially inappropriate behaviour; biting; scratching; rolling around the floor and head banging; increasing dependency on others for activities of living.
Family 5 (F5): **Evans** (Changes took place gradually since 2007)	Lack of regard for people around him including his daughters; did not show any emotional reaction to his parent's death; poor concentration; obsessive about collecting music of his youth and driving; decision making compromised, especially re: finances; overeating sweet and junk food; irritability; personality change; withdrawal from family and previous interests; socially inappropriate behaviour; verbally aggressive; urinary and faecal incontinence; went missing for days in Australia; increasing dependency on his wife.

The families reduced their expectations of the social behaviour exhibited by the

person with the symptoms. Families also report that they reduced social

contacts with the extended family and friends due to their embarrassment of the

increasingly inappropriate social behaviour, and frequently made excuses for their family member.

6.3 Theme 1: "Why are they behaving like this?: *Finding a reason*

Families living with someone diagnosed with bvFTD stated that they observed changes in personality, behaviour and social functioning years before the diagnosis was made. They had sought a reason for these changes by attributing them to things that had happened in their lives e.g. redundancy, depression, stress, and had attended medical appointments to secure a diagnosis and treatment.

Information collected from documents (notes, diaries, letters), and semi structured interviews suggests that, for some families, delay in diagnosis may have lead to relationship problems and reduced social connections not only for the person living with the diagnosis but also for the family. Therefore it is important to explore what happens to social connectedness in relation to families, friends, work colleagues, spouse and children pre diagnosis.

Daly *et al* (2013) stated that in the early stages of dementia a carer can feel they are in an "unsettled place", as they sense there is a difference in the person's personality or behavior. According to Daly *et al* (2013) these changes are gradual and initially do not significantly interfere with everyday living and

are more likely to be "explained away" or "normalized". As dementia progresses the carer begins to notice significant changes which cannot be easily "explained away", so another reason is sought. Daly *et al*'s (2013) findings are reflected in this current research as families and people associated with the person living with the diagnosis of bvFTD, attribute a variety of reasons to explain why the person is behaving as they are pre diagnosis, and these include mental health issues, physical health issues and relationship breakdown. Each of these are presented below.

6.3.1 Mental health issues

The case study families reported that they noticed changed personality, behaviour and social functioning before people outside of the immediate family. Initially they attributed exhibited changes to external factors that had happened in their family member's life, such as redundancy (see Box 6.1 below).

Box 6.1: Quotes from participants in the current research study regarding the effects of redundancy.

"He was sort of moved aside when they (management) re-organised as he wasn't going to be the one to take them forward. That was hard for him to deal with, as he had been there 35yrs. There was a slump, which I thought was a slight depression. He was going to the wrong workplace about 3-4 years ago. His communication was very poor. So he was stopped by one of the consultancy firms due to his behaving inappropriately. So 3-4yrs ago I took him for a full health check as I thought he seemed terribly depressed. He just disengaged. He was diagnosed after about 2 years." (Betty, spouse, F2, interview).

"I imagined he would be curled up or so depressed as he had been behaving in a strange way. I imagined he would be thinking the relationships in his life were difficult and he decided to give up. He had actually walked along the beach and ended up lying on a park bench for two nights." (Betty, spouse, F2, speaking about the time her husband went missing for 48hrs).

"I kept thinking he was clinically depressed where everything was unbalanced, therefore they would give him medication and everything would become balanced again. Maybe the fact his mum died and he lost his job, maybe these things have caused all this to happen, and maybe these things have been going on for a while for different reasons. I just thought someone was going to make him better." (Emma, spouse, F5, interview).

A study by Lockeridge & Simpson (2012) indicated that initially, a spouse has a tendency to try to "normalize" their partner's behavior, make allowances and give extra support. This appears to describe the ways the families in this study reacted. Brian (PLWD, F2) and Eric (PLWD, F5) had both been made redundant after a long and very successful career, and their families described how initially they felt a great deal of sympathy for them. They attributed the changes in behaviour, mood, personality and social functioning to a depressive reaction. It was only when the changes became progressively worse over time that they searched for other causes.

Andrew and Colin (PLWD, F1 & 3) had both retired from fulfilling careers, which had provided structure and social standing and both found this hard to come to terms with, so initially the families were not surprised by these changes (see Box 6.2). In the case of Diana (PLWD, F4), she explained the changes in her behaviour towards her sons, husband and parents in terms of stress in the workplace, due to covering a colleagues workload as well as her own. She also displayed a reluctance to socialize with her birth family, which was unusual for her, and explained this was due to stress in the workplace and having too much to do at home (see Box 6.2).

Box 6.2: Quotes from participants in the current research study regarding attributing the changes to a mental health issue.

"15 years ago we moved to this part of the country and he shut down and did not want to know anyone or do anything. I started saying to friends "I think he has a mental problem as opposed to a personality disorder". I did begin to jot things down that he was doing. He was getting repetitive and forgetful…. He said inappropriate things but they were becoming more inappropriate and I noticed this changing over the last 15 years". (Ann, spouse, F1, interview).

"We usually picked the boys up from school and my daughter would then collect them from our house when she finished work. We would usually have a chat about what we had been doing during the day but all of a sudden she was coming in and behaving in an agitated way saying to the boys "Come on, come on we've got to go". When I spoke to her about it she said its work and her husband was off work with a bad back and didn't know if he would be able to go back into his job. So there were a lot of things causing her stress". (Diana's mother, F4, interview).

Initially, the families accepted these explanations, but as the behaviour, personality and social functioning changes became progressively worse, and were noticed outside of the family context, they realised that medical advice was required (see Box 6.3 below).

Box 6.3: Quotes from participants in the current research study regarding prompts for seeking medical advice.

"Following a social event I got a letter in the post from a friend saying "Are you all right because your husband was very odd and everyone said so". I thought, "Thank god I'm not the only one who has noticed he is acting oddly". I felt relieved it was "out there" and now I had permission to do something about it". (Betty, spouse, F2, interview).

"Darren (husband of Diana) said he had the neighbours coming round asking if she was alright as they could hear the screaming and banging. She was shunning people when they came to the door, tell them to go away and she was fine". (Diana's mother, F4, interview).

Families felt relief that others had noticed and felt that it gave them "permission to act", as stated by Betty (F2). However not everyone welcomed other people noticing what was happening, as described by Diana's mother (F4). Due to the changes in employment experienced by all of the participants with bvFTD in this study, there was a tendency to explain the changes as occurring due to depression.

6.3.2   Physical health issues

Some family friends attributed the pre diagnosis changes in the person to physical/health causes and in particular deafness (see Box 6.4).

Box 6.4: Quotes from participants in the current research study regarding attributing the changes to a physical health issue.
"At this time he was also very quiet and I thought maybe he was deaf as he didn't respond when you spoke to him". (Family friend, F3, interview). *"The couple we were with were very sociable, we got on really well and we went on his last retirement trip. In the evening my husband would sort of sit there and not communicate particularly well at all and initially they thought he was deaf. They thought he couldn't stand the noise and he would just sit there and then go off in a huff. When we got back from the trip our friends said get him to go for a hearing test but he wouldn't. This went on for another year. So I asked his friend to have a word with him. He came and spoke to him and Colin agreed to go to the GP". (Carol, spouse, F3, interview).*

When family friends spoke to the family about their concerns over potential health problems, the PLWD was more likely to visit the General Practitioner (GP) for a medical examination, especially if they thought it was a treatable condition.

6.3.3   Relationship breakdown

The family member's change in behaviour, mood, personality and social functioning became progressively worse and they also started to exhibit an increasing lack of interest in their family and friends. In addition, their emotional response and empathy levels had reduced, and families realized that they needed to seek an explanation for the way their family member was behaving. The families began to explain this in a number of ways, such as relationship break down. Rather than assuming external factors were at play, assumptions implied that the person was consciously and intentionally behaving differently

and hurtfully towards the family, which created misunderstandings, arguments and resentment:

"We did go to Relate at one stage to talk about the relationship, which might also because of the condition, that would've been about nine years ago. Actually thinking back to that, he was willing to go and he would talk during the meeting but when were out he wouldn't talk to me or even try and build on what we talked about". (Emma, spouse, F5, interview).

The families described how the nature of family as well as spousal relationships gradually changed as the condition progressed, reflecting Massimo *et al*'s (2013) study findings in which the spousal roles and relationships changed gradually with the progression of dementia. Massimo *et al* (2013) reported that relational changes are accompanied by a growing state of dependence of the PLWD on their spouse, and a change in roles for the unaffected spouse, from partner to a carer. This process was identified as difficult for the non-affected spouse, accompanied by feelings of isolation, anger and grief.

In the current study, the previously close relationship between the PLWD and birth family was also affected by the changes, and this was most shocking for those birth family members that had infrequent contact due to geographical distance and infrequent social events:

"His mother had just died a couple of months previously so his sister came over from Australia. He completely blanked her and most of his family and they were absolutely horrified. It was as if none of us existed". (Betty, spouse, F2, interview).

Parent-child relationships were also adversely affected by the changes in their parent's personality, behaviour, mood and social functioning. In the Brown (F2), Davies (F4) and Evans (F5) families, the relationships between the children and their parent, changed from being a supportive, nurturing relationship to one characterized by indifference and irritability (Brown, Davies and Evans families) and arguments (Evans family) (see Box 6.5 below).

Box 6.5: Quotes from participants in the current research study regarding the changes to the parent-child relationship.
"She was also getting very abrupt with the boys and they were seeing all of this weird behaviour". *(Diana's mother, F4, interview).*   *"We had one of those family rows, you know, one of those things you often do in families, "What are you doing?", "Why have you done this?" And it escalated into the girls saying that they didn't feel he was being very attentive towards them, he didn't seem very interested, the elder daughter got quite upset and said he wasn't the father she wanted him to be or the father he used to be. They were quite close those two, and she felt that he just wasn't being what she wanted. He seemed to be totally unbothered by what they had to say." (Emma, spouse, F5, interview).*   *"Diana (PLWD) had gone from being compassionate, sensitive, caring to not giving a damn. If I cried she asked why. I would say "Because I am worried about you" but she didn't care". (Diana's mother, F4, interview).*

All of the study families reported that the PLWD denied they had changed in any way, and were very resistant to seeking a medical appointment when prompted by their spouse or family.[4] They eventually agreed when family friends persuaded them to seek help with perceived physical causes or possible depression as a result of life events. Each spouse in the study stated that they were relieved when other people noticed the changes in their partner as they had confirmation that something was "wrong", and it was not just the immediate family that had noticed and experienced this.

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[4] As mentioned in chapter 1, lack of insight can be a symptom of the frontal lobe changes in the brain.

Brian, Colin, Diana and Eric (PLWD F2, 3, 4 & 5) were unable to either remember or vocalize what happened pre diagnosis with family, friends and employment. Andrew (PLWD, F1) however said the following, indicating a level of self awareness:

"I think as a result of explaining to them (church group) what I could or couldn't do they have approached the people (other members of the village) to do what I was doing. I played the role of minute taker of church meetings, originally. Over a period of time other people came along and took over (church duties)." (Andrew, PLWD, F1, interview).

Although Eric could not recount how he experienced changes pre diagnosis, he had described his experiences to his wife Emma soon after diagnosis and she recounted this:

"I have talked to him over this period, and I said "Did you realise you were doing this?" and he said, "Yes", "Did you wonder why you were doing it?" and he said, "I don't know why I was doing it". "Did it distress you?" and he said, "Yes". Looking back we used to have times when our daughter would go to him and say "Dad why are you doing this to me? Why do you want to be like this to me?" He would just sit there like this, just sort of stare at her, his eyes started to shift around and sometimes looked puzzled, but sometimes to us he acted like he didn't have a care in the world but I think actually he couldn't work it out, it was strange". (Emma, spouse, F5, interview).

The adult children in the case study families declined to be interviewed (F2, F3, F5), and most were in counselling at the time of the research (F2 and F5). The children in the Davies family (F4) were under the age of 16yrs, and as both were undergoing counselling, due to the trauma caused by their mother's

condition, I decided not to involve them in the research. I asked Betty Brown (spouse, F2), Emma Evans (spouse, F5) and the parents of Diana Davies (F4) how the children attributed the behaviour of their parent eventually diagnosed with bvFTD. According to their parents (and grandparents, F4), children thought that the changes exhibited by their parent with bvFTD was in some way their fault. This was particularly noticeable in the youngest children and teenagers who lived in the family home, but was not in evidence in the children who lived more remotely from the family home. Each family said that the children thought that their parent (Brian, F2, Diana, F4 and Eric, F5) had stopped loving them, and had no interest and concern for them. They found this very difficult to cope with, as before their parent had the symptoms of bvFTD, they were loving, caring and attentive towards them. This altered behaviour was very difficult to come to terms with as their previous family relationship had been centred around love and care.

6.4   Theme 2: "What is happening to our family?": *Changing family relationships.*

Various psychological theories have attempted to explain human relationships (e.g. social exchange theory (Thibaut & Kelley, 1959), equity theory (Walster *et al*, 1978), investment model (Rusbult, 2003). Each of these theories provide an understanding of why some relationships succeed and others fail (Whitbourne & Whitbourne, 2014). What these theories suggest, is when rewards outweigh the costs the relationship is maintained, but once the costs outweigh the rewards, the relationship is more likely to breakdown. There was evidence this cost- benefit model was used by family members and friends in this study to

describe how relationships and social connectedness changed over time. This section will examine the data collected from the families to explore how social connectedness, relationships and social citizenship changed before the diagnosis of bvFTD.

6.4.1 Spousal relationship

The spousal relationships in the Brown (F2), Cooper (F3), Davies (F4) and Evans (F5) families were described as close, loving relationships where they socialised (e.g. parties, Barbeques) and took part in household (e.g. shopping, chores) and leisure (e.g. sports, visits) activities together. In the case of Andrew (PLWD, F1) and Ann (spouse, F1) a controlling relationship was described, with some elements of bullying behaviour (e.g. shouting, disparaging comments), together with a lack of warmth and empathy displayed by Andrew towards Ann throughout the marriage:

> *"From being on honeymoon the control started, he literally changed over night. His outward personality was totally different to the way he behaved at home. People used to say, "You are so lucky to be married to your husband he is such a gentleman", and I remember thinking they should see just for a second his behaviour at home and how he treats his wife. He was just evil and I was terrified of him for years." (Ann, spouse, F1, interview).*

The spousal relationships had started to break down in all of the families in the study due to the changes in behaviour and personality (see Box 6.6).

Box 6.6:	Quotes from participants in the current research study regarding the changes to the spousal relationship.

"When I said, "I am feeling really sad" to him he would say "Aw, what a shame, poor old you", sarcastically. When it is said like that you just want to kill them!". (Emma, spouse, F5, interview).

"I started to think about getting a divorce. The reason for that action was he refused to move out initially. I couldn't because I didn't have any money to move out unless I sold the house. It felt like I was being abused, it was like mental torture. Eric eventually moved out and up to London, with a bit of help from me. I had to pack things up for him. By that stage he was someone who was really horrible, so horrible, I didn't want him anymore. I wanted to get some normality back and the relationship was very damaging. I also thought he was having a mid-life crisis." (Emma, spouse, F5, interview).

"I thought he was very depressed and that something awful would happen. I would come home to find him hanging or something. He just disengaged. The communication was very poor." (Betty, spouse, F2, interview).

"Initially I just thought he was getting more like his dad, he was very Victorian and I thought that this was the route we were going down. I thought I would soon sort this out, but no. He's not the person I married". (Carol, spouse, F3, interview).

There was an increase in arguments, feelings of abandonment, isolation and thoughts that the marriage was over. Ann (spouse, F1) had already started to consider separation and had bought a flat away from the marital home due to Andrew's behaviour towards her, which she described as: indifference; shouting at her whenever she expressed an opinion; saying she was "stupid" and controlling what she did within the home. When she became distressed he showed no signs of concern or empathy:

> *"I bought a flat against everyone's advice, it was the best thing I ever did. I then had a bolt hole for weekends." (Ann spouse, F1, interview).*

Ann said that as she worked in the week she could live at home with Andrew because she was seldom there. However, it was at weekends when she felt trapped, isolated and found it impossible to live with his change in behavior, so needed to have her own private space, her flat, to go to.

In 2010, Eric and Emma's (F5) relationship had started to experience difficulties partly because of his behaviour towards his daughters, which had become very antagonistic, leading to family arguments. He would make derogatory comments to his eldest daughter and argue with her for no identifiable reason. Emma (spouse, F5) tried to intervene in these arguments, without success, and felt trapped between her husband and daughters. This affected Emma and Eric's (PLWD, F5) spousal relationship as they began to argue more, and she felt he did not try to understand how his behaviour was affecting the household. She also thought he did not love her anymore and he would not discuss this with her. They tried marital counseling at Relate but there was no real change in the relationship following this. Emma said whilst in the sessions, he would give the impression that he would work on the relationship but as soon as they left he would refuse to speak or implement any of the agreed changes. He showed no concern or empathy when she became upset. Eric (PLWD, F5) told Emma that he wanted a divorce, but did not do anything about it; he eventually left the marital home and embarked on an extramarital relationship. Betty and Carol (spouses, F2 & 3) indicated that pre diagnosis they felt their marriage and spousal relationships were over. They took their husband's lack of concern or empathy as a sign they were no longer loved by their partner.

In the current research, the spouses of the Brown (F2), Cooper (F3), Davies (F4) and Evans (F5) families reported that in their current situation, the costs outweigh the benefits of the relationship. They all said they had decided to stay

together as in their wedding vows they had promised "in sickness and in health". Each spouse explained that even when they got the diagnosis, it did not repair the damage in their relationship that had already taken place (this will be discussed further in chapters 7 & 8).

6.4.2   Parent-child relationship

The transition from a partnership or marriage into a family tends to occur when a child is born into the relationship (Whitbourne & Whitbourne, 2014). The birth of a child introduces the new role of parent and this changes their status with other family members, and with society in general. In addition, there are gender expectations of what a man or woman does within the family home. Women tend to perform more of the household duties, whether or not she has external employment (Coltrane, 2000), and a man is expected to increase their involvement in paid employment (Christiansen & Palkovitz, 2001).

The Brown (F2), Cooper (F3), Davies (F4) and Evans (F5) families complied with the gender norms of mothers having occupations outside of the family home on a part time basis, as well as having parental and home care responsibilities. The male spouses had full time employment and were the main wage earners prior to their retirement or redundancy. Ann (spouse) and Andrew Allen (PLWD, F1) did not have children together, although Andrew did have 3 children from a previous relationship, however he did not keep contact with them after his divorce from their mother.

Having a parent with young onset dementia can lead to psychological and emotional problems in children and young adults, which can affect their schooling and relationship with the parent living with the diagnosis of dementia (Luscombe *et al*, 1998). Their study also identified that the children of a PLWD could experience shame, stigma and bewilderment due to these changes. Children and young adults can experience grief, distress, and isolation due to the effects of young onset dementia within their family (Allen *et al*, 2009). In particular, this may be exacerbated by the inevitable changes in the family relationships and cause significant psychological strain. Allen *et al* (2009) state this is most noticeable when there is a: change in the parent's behaviour; delay in diagnosis; lack of understanding about the diagnosis and fear and worry about their parent after they have been diagnosed. In addition, they also worry about their other parent who is now a carer, the loss of "real" parent to the changes of bvFTD and possibly having a new role providing care for their parent.

In the current study, Andrew's (PLWD, F1) relationship with his children from the first marriage had already terminated many years before the onset of bvFTD when he was divorced from his 1st wife:

> *"When the mother and father split up and one went one way with the kiddies and that was it. If you try to keep contact on a regular basis it is a lot more difficult for the child who maybe receiving different instructions from each on how to behave, and what to do and so on and so forth."* (Andrew, PLWD, F1, interviews).

Ann (spouse, F1) described how she wanted children and they started having fertility treatment but he refused to continue and gave no good reason for this. She felt very bitter about this refusal, as she always wanted children. Ann also stated this characterised their relationship, in that if she wanted something he would say the opposite to "spite her".

In the Evans family (F5), the relationship of the PLWD and the children in the family unit changed significantly with clear signs of distress, family conflict and withdrawal (see Box 6.7).

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Box 6.7: Quotes from the Evans family in the current research study regarding the changes to the parent-child relationship.

"After the death of his mother Eric started to act in an irresponsible way. He made some very ridiculous purchases and began to disappear, driving around randomly and becoming detached and very non- compliant. This behaviour caused distress and resulted in a family row with him, our daughters and myself. Our eldest daughter in particular expressed her upset at his detachment, especially as they had been close before and said he was not behaving as she would like her father to be." (Emma, spouse, F5, interview).

"From this point Eric targeted our eldest daughter because in his words she criticised him and he "doesn't do criticism". He decided to ignore her and kept this up for 7 months, right up to the day she left to go travelling in New Zealand and Australia. He would see her distress but looked on unmoved. If he saw her in the street he would turn his back." (Emma, spouse, F5, interview).

"It's a shame because we all got brought into it in a way, because it all started with him and our daughter. I remember his sister saying, "Well, why are they there?", and it's because that's where it all started. It wasn't a thing between my husband and my relationship it was him and the daughters, with me trying to repair it and trying to explain". (Emma, spouse, F5, interview).

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The eldest daughter in the Evans family was understandably distressed by her father's behaviour towards her and would try to connect with him but he would rebuff her. She spoke with her mother about the situation and asked for her intervention, but Emma (spouse, F5) was unable to encourage Eric to change his behaviour towards their daughter.

In the Cooper family (F3) the children of the relationship although distressed by the change in their father, were more concerned for their mother and how she was coping with their father's changed behavior, personality and social functioning (see Table 6.1). It should be noted that these children lived away from home in their own households with supportive partners, and unlike the other families in the study were not faced everyday with their father's behaviour.

Darren (spouse, F4) isolated himself from Diana's family and ceased contact with his friends and neighbours. He encouraged their son's to continue with their school friends and social activities, and focused all his attention onto his sons' wellbeing. Contacts at the school stated they were doing well with their studies and mixing with other children in much the same way as before and did not appear to be withdrawn or adversely affected by what was happening at home or with their mother.

In summary, the PLWD were described as good parents who spent time with their children when they were growing up and who had a very loving, caring and supportive relationship with them. In the Brown (F2), Davies (F4) and Evans (F5) families this began to change in the pre diagnostic stage and was characterised by a lack of care, interest and empathy. Once the diagnosis was given, the previous close parental relationships were not resumed.

6.4.3 Birth family relationships

Alm *et al* (2014) indicated that relationships changed in families living with dementia and identified a sense of longing, lost closeness, loneliness and changed sibling relationships, that not only affected the spouse and children but also the PLWD's siblings.

Sibling relationships are often the longest lasting of our lives (apart from parents) as they share so many early experiences, background situations and memories. In the current study the sibling relationships were described as becoming more distant, with infrequent to no contact when their family member began to exhibit changes in the way they behaved, communicated and engaged in family activities and social events. In two families (Brown and Evans, F2 & F5) this change in relationship became evident when a parent died. Brian and Eric (PLWD F2 & F5), did not behave in a way expected of them by their siblings and when given "tasks" to perform following their parent's death, they did not follow them through, which led to arguments and strained relationships:

> *"His sister expected him to clear out their mother's house when she died so it could be sold but he wasn't getting on with it and she was getting very frustrated and annoyed because they had a buyer for the house and this caused annoyance and rows." (Emma, F5, interview).*

The behavior of Brian and Eric was totally out of character from their usual organized, helpful and caring approach with their siblings. When challenged regarding their lack of support and apparent lack of concern, they did not

respond in the way they usually did. The consequent arguments and breakdown in relationships was beyond the level of usual sibling disagreements.

In a study by Hsieh *et al* (2013), lack of care and concern was highlighted as a particular issue affecting people diagnosed with frontotemporal dementia. They attribute these changes to atrophy in the right ventromedial frontal and anterior temporal pole regions of the brain, areas that are thought to be critical for empathy and social behaviours. As this was the time before diagnosis, the current study families would not have known that the changes in their family member, where in fact due to brain changes, over which the person had no control. An example of this lack of empathy is regularly demonstrated by Eric (PLWD, F5) towards his sister. Eric's brother was murdered in 1987 and now he has bvFTD he talks about this a lot when his sister visits him, which she finds very distressing. He still continues to talk about what happened in detail to his sister, despite seeing her distress and her requests to stop talking about it. She still visits him on a quarterly basis but says she finds it very distressing to do so.

The sister of Brian (PLWD, F2) lives in Australia and they used to keep contact by phone until the onset of bvFTD, but now Betty (spouse, F2) hears from her infrequently, and she feels upset about what she interprets as an apparent lack of care and concern. Brian's brother has not contacted Brian or Betty since the onset of bvFTD. Andrew (PLWD, F1) had strained relationships with his birth

family for the majority of his adult life, and there was no change in this pre or post diagnosis. Colin (PLWD, F3) has two brothers but only see's one of these regularly since the onset of bvFTD.

Brian (PLWD, F2) and Eric (PLWD, F5) showed no emotion on the death of their parents when they had previously had a caring and close relationship with them, and this was a signifier to Betty (spouse, F2) and Emma (spouse, F5) that there was something "wrong" with their husband's emotional expression and behaviour, and this prompted them to seek a medical opinion.

Diana's sister (F4) visits regularly but finds it very difficult to do so, and often spends the time after the visit crying due to her grief in seeing her sister as she is now. Diana's parents (F4) have been devastated by the behavioural changes that they have noticed in their daughter:

> *"She didn't really have emotion, she neither laughed nor cried, she didn't ask after her family or her boys. She was rude to the staff and other patients." (Diana's mother, F4, interviews).*

This distress was further compounded by the breakdown in relationship between themselves and their son-in-law. Diana's parents and sister state that the relationship with Darren (spouse, F4) had always been strained prior to the onset of symptoms:

> *"The relationship issues between Darren and I goes back over years. He would make us feel uncomfortable when we went around to their house for*

a social situation. He didn't like the closeness of the family relationship and our role in the childcare of their children. We fitted our lives around their careers and helped with childcare. He was resentful about all of this". (Diana's father, F4, interviews).

Family 4 was unusual in the study because Diana (PLWD) was in her 30's, as compared with other PLWD who were in their 50-60's. The sudden onset and rapid progression of behavioural, personality and social functioning gave Diana's parents high levels of distress and they pushed for a diagnosis and treatment, at times against the will of Darren (spouse, F4). This caused significant conflict between the families:

"I think that's when the relationship between my son in law and I started to breakdown because I wanted to push for more tests and opinions but the ward did not see the need for this. My son in law was reluctant for more tests as he listened to the ward staff and doctors when they said it wasn't that bad. I tried to say to him this wasn't normal and we should get more tests. I must admit I was a bit like a dog with a bone and insisted she should be taken to the neurological hospital and he eventually agreed." (Diana's mother, F4, interview).

Darren initially believed Diana when she said she was stressed, and when these changes continued to worsen over time, he then believed the medical practitioners when they said the changes were due to a psychiatric disorder. Her parents knew that what they were seeing did not fit either of these explanations so continued to seek a diagnosis and potential treatment, despite the fact that it was causing a rift in their relationship with their son-in-law.

In the Brown (F2), Cooper (F3), Davies (F4) and Evans (F5) families the birth family relationships for the PLWD started to break down in the pre diagnosis stage from the relationships they once were and were not reinstated to the previous level after diagnosis.

6.5   Theme 3: "Other people are noticing"

Friendships and social groups provide many forms of support and help to maintain a person's emotional well-being (Whitbourne & Whitbourne, 2014). According to Hartup & Stevens (1997) what underpins close friendships is reciprocity (give and take at a deep, emotional level), or a sense of mutuality. Close friends are also expected to help each other through life transitions, such as relationship changes, employment issues, family challenges and health changes (Whitbourne & Whitbourne, 2014). According to Adams & Blieszner (1994), friendships follow a trajectory from formation to dissolution where the quality and benefits of the friendships are informally evaluated, and a decision is then made to increase or decrease involvement. They also state there is a tendency for individuals to choose friends who are similar in gender, socioeconomic status and ethnicity.

The majority of close friendships remain throughout life, but there can be a reduction in individual friendships, and an increase in joint friendships when a person enters a long-term intimate relationship with a partner (Kalmijn, 2003). In addition to close relationships, there may be peripheral ties, which are social

friendships at a surface level and these include neighbours, co-workers, friends or children's friends parents (Fingerman & Griffiths, 1999). Chapter 5 described how the PLWD life and social connections changed after they left their career, as they reduced or ceased contact with their previous work colleagues and social network.

6.5.1 Work colleagues

Diana (PLWD, F4) was in paid part-time employment when her behaviour and personality changed rapidly during late 2011 and throughout 2012. Diana's inappropriate behaviour included: lack of regard for people around her; rudeness to the public; irritability; running and skipping around the workplace without shoes. It was these changes that led to her leaving her job (see Box 6.8).

Box 6.8: Quotes from the Davies family in the current research study regarding the behaviour exhibited by Diana with work colleagues.
"Her manager started to describe some of the things she was doing at work: whistling; taking her shoes off and skipping down the corridor; obsessive about switching lights, sockets and taps off; being rude to people and behaving inappropriately." (Diana's mother, F4, interview).   *"All of her colleagues that she worked with would come to see her but the minute she saw them coming she would run into the bathroom and lock herself in, she wouldn't see them at all. All of this hurts me, I wonder whether she knew that there was something wrong and had some insight and was hiding because she did not want them to see her." (Diana's mother, F4, interview).*   *"She was shunning colleagues when they came to the door. She would tell them to go away and say she was fine." (Diana's mother, F4, interview).*

Once Diana left her job and went into hospital for investigations, she ceased contact with her work colleagues even though she previously had got on well with them socially as well as at work. According to Diana's parents (F4) the friendship group and work colleagues wanted to keep contact, but they were

told not to visit by Diana (PLWD, F4). Diana's mother was worried by this, as to her it meant her daughter probably had insight into what was happening to her and did not want her colleagues to see her.

In all of the families, the spouse was in paid employment whilst the family member started to exhibit the changes in behaviour and personality. All of the spouses cited the difficulties they had "juggling" the role of employee, spouse, parent and family members whilst their relationship faced difficulties, due to the change in personality and behaviour of their spouse or family member. They described how they left home in the morning not knowing what they would be going home to at the end of the working day. Darren Davies (spouse, F4) gave up his job to focus on the needs of his family full time as he worried about how his young son's would cope with their mother's "ill health" and her behaviours towards them. The spouses in the Allen, Brown, Cooper and Evans families (F1, F2, F3 & F5), stated that despite these worries, they wanted to continue going out to work, as it gave them some respite from what was happening at home, and they had the social and friendship support from their work colleagues. In addition, they all stated that it helped them to retain a sense of self-identity, self-esteem and connectedness.

6.5.2 Social relationships

Social relationships are the social interactions between people over time and are made up from: social supports (from family, friends, relatives, co-workers and social group); social networks (size, composition and frequency of

contacts) and these can lead to social connectedness. The connection with other people, including family, friends, work colleagues and people with similar interests, leads to a feeling of belongingness, attachment, identification and self-worth, if it is a positive experience. If, in the experience the person feels detached, excluded or identified in some way as not being part of the group, it can lead to feelings of social isolation (Lee *et al*, 2001).

In all of the families in the study, the person living with dementia (PLWD) was reported by family members and spouses to have had an active social life before the onset of bvFTD, mainly revolving around work and close friendship groups:

> *"He was the sort of person who was part of a very sociable group and it was this that pushed him to maintain friendships."* *(Betty, spouse, F2, interview).*

> *"Colin was an easy going, competent man. He was always a quiet type, but was sociable".* *(Family friend, F3, interview).*

As the PLWD's behavior, personality and social functioning changed towards friends, work colleagues and the local community, social connectedness gradually decreased over time, with reduced invitations to socialise or join activities. When I asked families did they think their family member (PLWD) withdrew from social contact, or was it others that withdrew from them, they stated that it was a bit of each. Andrew Allen (PLWD, F1) cut himself off from his previous friendship and work colleague group when he retired and relocated, but eventually started to forge new relationships within the local

community, actively involving himself in the church and community events. However, the church community withdrew from the family when Andrew's behaviour (singing loudly at inappropriate times) and social functioning (socially unacceptable comments) changed, and both Ann (spouse, F1) and Andrew felt very upset by this. This was a few years before the diagnosis and it upset them greatly, as they had been very active in the local community and had made a significant contribution through their work with church groups. Even after the diagnosis, the church group still remained intolerant of Andrew's behaviour and excluded both Andrew and Ann from church events:

> *"It's more people who I think should know better that are bad. People at church and things like that. I think how can you not come to talk to me because of his behaviour!" (Ann, spouse, F1, interview).*

Brian Brown (PLWD, F2) did not actively maintain friendships or develop new ones, but would attend events if invited. He appeared to enjoy attending, but his socially inappropriate behavior started to cause concerns (e.g. asking people for computer passwords, drinking excessively and not following usual social conventions of behavior). Colin Cooper (PLWD, F3) attended social events, but tended to be more withdrawn in his friends company, and did not take part in conversations as he did previously. Diana Davies (PLWD, F4) cut herself off entirely from friends and family, and at times actively removed herself from situations involving friends, work colleagues and neighbours by running away and hiding.

Eric Evans (PLWD, F5) remained sociable with friends and contacts outside of the family home, but also began to occupy himself with his own interests to the exclusion of family and friends (see Box 6.9).

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Box 6.9: Quotes from the Evans family in the current research study regarding the behaviour exhibited by Eric (PLWD) in social situations.

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"He began to immerse himself with his music collection downloading and purchasing re-mastered CD's to recreate his old record collection. He became rather obsessive and disruptive when friends came over, playing music loudly and switching tracks but not engaging with the conversations." (Emma, spouse, F5, interview).

"Eric was someone who prided himself with his ability to empathise with others but stopped engaging with his immediate family. He did still continue to offer support to others outside of the home which was upsetting for us and a source of resentment." (Emma (spouse), F5, interview).

"We didn't go anywhere together, we didn't go to friend's houses. I stopped that probably at the beginning of 2011 really because I didn't want to have someone who was hostile to me in the home environment to then go into someone else's house and put on a show. He was quite capable of putting on a performance for others." (Emma, spouse, F5, interview).

" Eric kept fiddling with everything in the house, he kept turning everything off at the mains (hot tub; toilet flush; switches). He kept wandering into the bedroom unannounced in the early morning when told it wasn't right to do this he still kept doing it!" (Friend of Eric, F5, document).

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Emma (spouse, F5) decided not to go to social events with Eric due to how he was behaving in the family home. She found it distressing that he behaved in a hostile and argumentative way in the home environment, and in a superficially friendly way when in the company of others. She surmised from this that his behaviour towards her was intentional and he had some control over it. This impacted on her social networks as these were joint friends and as she declined the invitations they gradually stopped inviting her.

Eric's (PLWD, F5) erratic behaviour, deficits in his problem solving and decision making ability became more apparent when he went to see his friend in Australia. This behaviour consisted of: losing the car at the airport; crashing a

car but not reporting it; sleeping rough and missing flights. In addition, his friend had concerns that he was acting in a socially inappropriate way.

Each family's spouse spoke about having 2-3 close friends who were consistently supportive in times of difficulty, but the total number of close friends had reduced over time. The friends who were described as "true" or "consistent" friends, were people who would contact or visit the spouse and offer: help; a "shoulder to cry on"; someone to talk to about their feelings and thoughts and respite (e.g. going out to a café, cinema or walk). The spouses stated that they were "true friends" as they were there through "thick and thin", were not judgmental and were trusted:

> *"When this started you realised who your true friends are. I have two or three really, really close friends who are in and out of this house." (Carol, spouse, F3, interview).*

The value of "true" friendships and support of work colleagues was described as very important for the spouses who were living with uncertainty and family distress at this time. They cited this as helping them to cope and make some sense of what was happening to them. This also enabled the spouse to retain their sense of social connectedness with a supportive network at a time when other social networks were reducing, and their family relationship and roles were changing.

6.6   Theme 4: Getting a diagnosis

Diagnostic difficulties occur because of the diverse initial symptoms of young onset dementia (Luscombe *et al*, 1998), the rarity of the diagnosis (Rossor *et al*, 2010) and the gradual changes over time (Liebson *et al*, 2005). It is important to secure a quick diagnosis, since delay in diagnosis can lead to increased distress for the family (Vogel *et al*, 2010). In the current study, the range of years between families noticing the first changes to diagnosis ranged from 1 and 15 years.

The diagnostic process described by the families in the current study was characterised by repeated visits to the doctors to get a diagnosis, with increasing levels of distress and thoughts that they were not being listened to (see Box 6.10).

Box 6.10: Quotes from participants in the current research study regarding getting a diagnosis.
"*Just before mum's 90th birthday I went to see the doctor with my concerns and he just sent me away. It took a good two years of noticing problems and thinking I must see someone and then just to be told to go away.*" *(Ann, spouse, F1, interview).*   "*When I went back I went with a list and I had this list in front of me. When he started questioning me I said I will take questions at the end and want to have my say first and I want you to listen to what I have to say.*" *(Ann, spouse, F1, interview).*   "*Brian (PLWD) was very depressed (after he was made redundant) and I thought something awful is going to happen here and so I took him for a full health check. When it was time to go back for the results I thought I must be more proactive here and said, "I want to come in," which Brian agreed to. I went and at in for the results and the doctor was talking about slightly high blood pressure. I then said, "Excuse me, but I was the one who urged my husband to come and I am seriously worried about him". I then saw the doctor suddenly switch and he obviously hadn't thought of the effects of losing his job had on my husband and his behaviour change. Things moved quickly after that. We were sent to neurologists and we were already thinking it could be Alzheimer's.*" *(Betty, spouse, F2, interview).*

> *"When Colin (PLWD) visited the doctors (for his hearing test), the doctor realised that she would tell him things and then he had to write them down and she was concerned and thought he had a brain tumour. So she sent him for a scan the next day. He had the scan and it came back showing shrinkage to the front of the brain. They diagnosed him as Alzheimer's disease. Our daughter thought the symptoms were more suggestive of frontotemporal lobe dementia, so she pushed for her father to have more tests, which he did and frontotemporal dementia was diagnosed after 18 months with the wrong diagnosis."* (Carol, spouse, F3, interview).
>
> *"I had also helped with a patient with Pick's disease…. I knew she (Diana, PLWD) had the same thing. The neurologist said, "No she is far too young. We don't know what it is, but it's not Pick's. I will get her readmitted to a neurological ward", but we had to wait for a bed. About a week later the original consultant came up with a diagnosis. He phoned us and seemed quite excited, my son-in-law, my husband and I went into the hospital and they said we have some results that come back suggesting she has PANDAS (childhood autoimmune disorder). He said there is research going on into adult PANDAS and because of this borderline result of the blood test it was thought that my daughter had adult PANDAS. I asked "Is it treatable?", "Yes", he said, "Is it curable?", "Yes" he said. "So she should be able to come home as a normal wife and mother?" He said, "It may take some time but yes". "What about work?", "Yes" he said. We walked out of the hospital like we were on a cloud, we celebrated."* (Diana's mother, F4, interview).
>
> *"We were looking for improvements all the time and thought Diana might have been a little bit better each time. After a week we start to realise that there was no real change and after two weeks she was getting worse. We went back to the hospital but the previous consultant had now left for another job. The Department said they could do no more and she would have to go back to mental health. So she duly went back and had more memory tests. Her doctor there laughed about what the other neurologist had diagnosed. My son-in-law was now beginning to get quite angry and said he wouldn't accept it was neurological and we have to accept it's a mental disorder… He started to get cross with me and said that I had insisted on her being on the antidepressants, which was not the case. So things were starting to break down. He was very resistant to getting a second opinion and when I reminded him of the scan results he said, "You heard what the neurologist said it was only slight amount of shrinkage and nothing to worry about. All the tests were clear". I was still in touch with my daughter's colleagues and they kept saying there was something very wrong and said I should get a second opinion. I was getting stuck now because I wasn't the next of kin and my son-in-law was beginning to resent my pushing."* (Diana's mother, F4, interview).
>
> *"Diana (PLWD) had another scan and I sat in the room with her whilst she had it done. I could see the staff looking at the screen and pointing and then calling other people in and I knew there was something very wrong. They asked her husband and I to come for the results and they told us there was massive destruction of brain cells in her frontal lobe."* (Diana's mother, F4, interview).
>
> *"After all of the family arguments and crises in 2010, Eric (PLWD) went to Australia in December to see one of his friends. His behaviour whilst there caused his friend serious concerns. I made an appointment with the GP on his return and said, "I want Eric to see a psychiatrist. I don't want therapy (she had already seen the GP months before and he advised attending Relate), I want you to look at his brain to see if anything is happening to it and I want him to have a brain scan". I took pages of notes with me for the GP and the referral to the psychiatrist. Eric had the scan but I still believed it was a psychological problem following his parent's deaths, redundancy and his brother's murder. I thought he would be given treatment and he would be well again. I went for the appointment with the GP again and he said they had found shrinkage of the brain on the scan and Eric had frontotemporal dementia."* (Emma, spouse, F5, interview).

In addition to the distress these families felt, they were also given the wrong diagnosis initially, which led to feelings of hope as the disorder was often misdiagnosed as a "treatable" and potentially "curable condition". This lead to additional distress for the

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families when they realised the diagnosis was incorrect, and their hopes of a return to normal were dashed, as their family member continued to deteriorate. The families then had to return to the medical practitioners for another assessment and diagnosis. As can be seen from the quotes in Box 6.10 the families in the current study then went back to the medical practitioner with additional evidence, and a determination they would be listened to and not dismissed.

The case study families' described, and gave examples of their fight to get the correct diagnosis, they indicated how they felt frustrated and powerless in the diagnostic process. As an attempt to address this, families gave examples of how they tried to regain control, and challenge the power imbalance they felt e.g. making lists of symptoms; seeking second opinion; pushing for further tests; telling health professionals to listen to them. This helped the families to eventually get a diagnosis for their family member.

For Diana's family (F4), not having the correct diagnosis and the search for the right one adversely affected her parent's relationship with their son in-law (Darren, spouse, F4) but also led to a breakdown in Darren's relationship with Diana's sister. From letters to and from Darren, it is clear that he felt Diana's family were critical of his role in the assessment, diagnostic process and the decisions he made on behalf of Diana (PLWD, F4). In the letters from Darren, he asserts his legal right to make decisions regarding Diana in his role as her husband and next of kin above that of her parents. This contributed to the breakdown in the relationship between Darren and his in-laws.

During the interviews and observation sessions, only Andrew (PLWD, F1) could speak about when he got the diagnosis, all of the other PLWD were unable to communicate what had happened:

> *"I didn't get any information about what it was they just said we will see you a bit later. They said they would let me know the next time when they saw me what it meant and they would give some help and so on and so forth but this hasn't happened yet."* (Andrew, PLWD, family 1, interview).

It should be noted that the PLWD in the Brown (F2), Cooper (F3) and Davies (F4) families were exhibiting extreme difficulties with verbal expression when interviewed or observed, and when asked several times about the diagnosis they did not reply. The families said that when the diagnosis was given, their family member did not show any reaction or acceptance that there was a problem with their behaviour, or that they had changed in anyway.

In the current research, there was a general feeling of relief when a diagnosis was given because it gave a reason for the observed changes in behaviour, personality and social functioning. The families were able to understand that this was due to a physical cause rather than something they had done, or was something intentional on the part of their family member.

6.7   Summary and transition

This chapter analysed and interpreted data regarding social connectedness pre diagnosis and at the time of diagnosis for the 5 case study families. The results from each family were compared and contrasted with the other case study families to explore similarities and differences in their experiences.

Each of the case study families described the gradual progression of bvFTD, from the first signs of changed personality, behavior and social functioning, to their struggle to get an accurate diagnosis. Theme 1: " Why are they behaving like this?":*Finding a reason*, described when the changes were first noticed and how they presented. The family attributed a variety of reasons to explain why the person had changed, including mental health issues, physical health issues and relationship breakdown.

The case study families stated that they believed their family was breaking apart due to the changes exhibited by their family member with bvFTD (Theme 2: "What is happening to our family?": *Changing family relationships*). They described how this affected the spousal, parent-child and the birth family relationships. The experience voiced by each participant, indicated the lack of understanding of what was happening to the family, the depth of distress and breakdown in family relationships.

The first signs of bvFTD were more noticeable by the family initially, but eventually other people began to notice in the workplace and in social settings (Theme 3: "Other people are noticing"). The social contacts and networks started to reduce, as the PLWD behaviour did not conform to social norms and caused embarrassment. As a result of the socially inappropriate behaviour by the PLWD, the families withdrew from social events due to embarrassment and feelings of shame. This negatively impacted on the families' social connectedness and their sense of social citizenship.

The families in this study recounted their various experiences of getting the correct diagnosis for their family member (Theme 4: Getting a diagnosis). They expressed similarities regarding medical practitioners inability to listen to the families when they were expressing their concerns about their family member's changes. This resulted in a delay in the diagnosis and further distress for the families in the current study. Families described how they challenged and managed the process of getting the correct diagnosis for their family member.

In chapter 7, (Findings and interpretation: social connectedness after the diagnosis) I will analyse and interpret the data collected from the research study to address the research questions whilst considering possible alternative interpretations. A comparison will be made with what I have found in my research study with other available literature on the topic.

Chapter 7: Findings and Interpretation

Social Connectedness after the Diagnosis

7.1   Introduction

As stated in chapter 6, the six components of citizenship (Bartlett & O'Connor, 2010): growth; social position; purpose; participation; solidarity and freedom from discrimination had all been altered pre-diagnosis not only for the person living with bvFTD, but also for the rest of their family. There was an assumption by the families, that once the diagnosis was given, this would change how friends, relatives and the local community would treat their family member, and the rest of the family. However this was not the case, families felt let down by the perceived community and social network lack of understanding and support, and the further reduction of contact with family and friends.

Once the diagnosis was given, each family in the study described how they coped, what support they received and what happened to their social connectedness. This information was collected from: participant observation; semi structured interviews and documents (notes; diaries and letters).

7.2   Theme 5: "Now we know what it is".

The assessment and diagnostic process was traumatic for families and was characterised by, a difficulty in receiving the correct diagnosis from medical practitioners (see chapter 6). Other researchers have also recognized this in their studies and state it is due to the lack of awareness of young onset

dementia by medical practitioners, and the range of signs and symptoms that do not always include a memory disturbance (Luscombe *et al*, 1998; Rossor *et al*, 2010; Johannessen & Moller, 2011). This can lead to an incorrect diagnosis and a delay in referring the symptomatic person for further investigations, diagnosis and support.

The medical practitioner's lack of awareness, understanding and their inattention to what the families were saying about their family member, gave rise to frustration, as the study families attempted to make them understand the situation they were experiencing. This was also recognized in a study by Johannessen & Moller (2011), which found that families faced difficulties getting health professionals to understand there was something wrong with a family member. Initially the health professionals frequently gave an incorrect diagnosis, such as depression or "burnout", instead of recognising the symptoms of young onset dementia. Families thought that this delay in diagnosis further compounded relationship difficulties with family, friends and social groups. Had this diagnosis been made earlier, their relationships might not have deteriorated to such an extent, as knowledge of the condition might have enabled them to make allowances and seek support.

When the correct diagnosis is given families should receive specialist post diagnostic advice and support but unfortunately this is not always the case and families are left to try and seek out information and support themselves, so adding to the distress they experience (Johannessen & Moller, 2011). The

families in the current study stated that they fought to receive the correct diagnosis with the expectation they would then receive appropriate treatment and support, however this did not prove to be the case and the following sections of this chapter will explore this further.

7.2.1 Post diagnostic support

Following the diagnosis there was a lack of specialist advice and post diagnostic support for all of the families in the current study and consequently the families felt disappointed, angry, abandoned and trapped by the situation they found themselves in. This affected how they managed to live with the diagnosis as a family (see Box 7.1). These feelings have also been highlighted in other studies of young onset dementia (Armari *et al*, 2013; Werner *et al*, 2009; Lockeridge & Simpson, 2012).

Ann (spouse, F1), Emma (spouse, F5) and Betty (spouse, F2) described how they felt lost, isolated and abandoned by a service they expected to get help from. This need for specialist advice and support to enable families to cope was also identified in other studies (Johannessen & Moller, 2011; Lockeridge & Simpson, 2012; Diehl-Schmid *et al*, 2013; Kaiser & Panegyres, 2007). In a similar vein, both Eric's (PLWD, F5) and Brian's (PLWD, F2) families, felt they were victims of age discrimination, in that the dementia services and support were provided for people over 65yrs of age. Eric's and Brian's needs and presentation of dementia were very different to the rest of the people who used the dementia services, and so they were excluded from attending by the staff.

The staff delivering services and support designed for people under the age of 65yrs with various mental health and neurological disorders, excluded Eric and Brian, as their services and centres would not accept people with a diagnosis of dementia.

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Box 7.1:   Quotes from participants in the current research study regarding post diagnostic support.

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"That summer after the diagnosis was the worst time as we had no support whatsoever, no-one to ask, no-one to go to, I didn't know what to do with Andrew (PLWD). I spent a lot of that summer crying. This time, as well as pre diagnosis, is when you need someone and there just wasn't anyone! Seven months after diagnosis the GP referred us to the Mental Health Team." (Ann, spouse, F1, interview).

" We have a person who comes on the Monday and Tuesday but Brian (PLWD) doesn't speak to them. In the last week I spoke to social services to get a carer's assessment. I visited two centres around here on the recommendations of social services. It turns out one of those doesn't accept people with dementia. So I had to accept the other one. The bus came and he got on and went to the centre from 10m-2pm. Brian went a few times but then the social worker contacted me to say the centre believed they were unsuitable for my husband. There is another centre but it is a quite a way away and is more expensive at £50 per day for 10-2pm. I need him to attend for my peace of mind but I don't think he will enjoy it." (Betty, spouse, F2, interview).

"Our daughter suggested her father goes to a day centre to give me a break. We have a lovely carer who comes and takes Colin into a pub on a Friday night and on Wednesday afternoon he comes around to check he has had something to eat."(Carol, spouse, F3, interview).

"They sat us down and told us it was frontotemporal dementia. They explained how it was often called Pick's and what it meant; there was our son-in-law and I both in tears. Our son-in-law ended up by saying she couldn't come home. So we started looking for a care home. He didn't want her at my home either because geographically we were very close. That was our first little blip. The awfulness, the devastation, the horror, the emotion, everything came at us, and then looking for a nursing home." (Diana's mother, F4, interview).

" I asked a GP what could I expect and he said, "Very little". I had been to the GP sometimes and I was constantly trying to find out as much as I could. We did finally see somebody (after 18 months) and I was, 'Oh at last'. They came around and spent about two hours with me, it was this psychologist from the team, and I never saw them again. When I saw them I was like, 'Thank goodness, I've got somebody', I said "I don't know, I'm not skilled at dealing with this….I don't know, the right thing or the wrong thing, I don't know, I just need a bit of help. Then they disappeared and somebody else came, another six months later, another one. I got to the point I wasn't even going to bother with them they were so useless. Then another one came with a whole load of stuff, then at the end the woman said, "I don't know if I can help you". The reason being is that, because he's got dementia, he fits into older adults, but because he's young, and they don't know how to do it they kept pushing me around. (Emma, spouse, F5, interview).

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In families 3 & 4, no post diagnostic support was offered or given by the medical practitioners; it was left to the family to make the decisions regarding future care and support. It was Carol and Colin's daughter (F3) who suggested the post diagnostic support as she thought her mother should receive respite from caring. The only post diagnostic advice given by the practitioner was to arrange Lasting Power of Attorney immediately. In the case of Diana (PLWD, F4), Darren (husband of Diana, PLWD) decided that for the sake of their children he could no longer have his wife in the family home and the only solution was for her to go into long-term care. Diana's parents found this difficult to accept, but could understand why he came to that decision as by this point, Diana was screaming, shouting and neglecting her personal hygiene and this was frightening the children.

Role overload, role conflict and associated stress and frustration were evident in the current study. Although having employment outside of the home added to the role overload, Ann Allen (spouse, F1), Betty Brown (spouse, F2), Carol Cooper (spouse, F3) and Emma Evans (spouse, F5) did not want to give up their employment as they thought this gave them: financial stability; personal growth; a social network; time away from caring and a sense of personal identity. For Darren Davies (spouse, F4), the role overload and conflict of having a job, young children, household responsibilities and visiting his wife, created too great a personal burden and he decided to stop working to become

a full time father for his young sons to reduce the disruption the family had experienced when Diana (PLWD, F4) became unwell.

All of the spouses spoke about how supportive their employers and colleagues were when they found out about the diagnosis of their family member. However, the family caregivers in this study often found it difficult to continue in employment due to the lack of age appropriate, reliable and good quality services to support their family member (PLWD).

7.2.2 Spousal connectedness

In the UK, the peak age for a person beginning to have a family (adult) caregiver role, is 45-64 years of age (Hirst, 2002). In addition to the caregiver role, at this age a person is more likely to have: dependent children; a career or job demanding time and effort; a mortgage and financial pressures. What is not expected is caregiving for a spouse who is diagnosed with young onset dementia, which can coincide with being the caregiver for elderly parents and children. This was certainly true for the spouse caregivers in the study and in the case of Darren Davies (spouse, F4); he was considerably younger as he was in his 30's.

Rolland (1994) wrote about the difficulty of maintaining a balanced relationship whilst at the same time adopting the role of carer, and indicated that some

psychological strain is unavoidable in these circumstances. This was echoed in the experiences of the families in the current study. For example, the spouses in the Brown, Cooper and Evans families stated that they no longer felt like they occupied the status of "wife" but were instead a carer, not a role they had volunteered for or wanted:

"I don't want to think of myself as a carer, I have to tell myself, yes you are get over it!" (Betty, spouse, F2, interview).

"I think one of the saddest aspects was that before you get the diagnosis you already feel alienated from the person. I started to feel not interested in this person or his behaviour's" (Betty, spouse, F2, interview).

In Betty's quotes (spouse, F2), she spoke about her feelings of alienation from her husband, and how her relationship had changed in her eyes from wife to carer. She stated she looked after her husband because other members of the family expected it, and she promised to do so in her wedding vows but love, affection and the relationship they previously shared had now gone.

As previous research has indicated, additional roles and expectations taken on by spouses can cause stress and frustration (Svanberg *et al*, 2010). This is partly due to competing demands and lack of control over the situation they find themselves in (Baikie, 2002). In the current study, the spouses in the Brown (F2), Cooper (F3) and Evans (F5) families described how they felt that their relationship, and role as a partner had already broken down in the pre diagnostic stage (see Box 7.2). They all felt the relationship could not be rebuilt

to how it was before the changes in personality and behaviour occurred, as this was no longer the person they loved and shared a happy life with. This could suggest that to retain relationships within families living with bvFTD and maintain their social connectedness, early recognition and diagnosis is crucial.

Box 7.2: Quotes from participants in the current research study regarding spousal connectedness.

"*Now I've got the diagnosis it gives a reason to some of the behaviours. I don't get irritated by Brian's (PLWD) behaviour; I just ignore it and avoid getting into a situation where it could become annoying. Sometimes I think it's such a long period without having anybody to talk to about this, no one to rely on and always working around the other person. Always working around their needs and it gets very wearing. (Betty, spouse, F2, interview).*

"*Carol (spouse) has become more like a mother, and in some ways it's worse than looking after a child. She looks more tired and is less patient." (Family friend, F3, interview).*

"*There are times when the frustration is so great that I feel like hitting Colin (PLWD). I don't of course. This is so sad because he was such an easy going, competent person." (Carol, spouse, F3, interview).*

"*Thank goodness it was diagnosed because if it hadn't been I wouldn't be here now. I just knew this wasn't the person I married, he is a really changed character from being the easiest going person and very clever to what he is like now. I have to be honest I'm not really trying to keep track of everything that is happening now to my husband because I don't have the wherewithal to do it anymore." (Carol, spouse, F3, interview).*

"*I kept feeling a bit guilty that I wasn't looking after Eric (PLWD), actually I was really looking after him in his flat. Everyone around me says "You do so much for him", I meet some ladies at this group who have dedicated their whole lives, I mean I have dedicated my life but I still have some freedom to do things and go places." (Emma, spouse, F5, interview).*

Betty (spouse, F2) indicated that she had changed the way she interacted with Brian (PLWD) to avoid situations that could cause distress or an escalation of behaviours. She indicated that having a reason for the behaviours gave her more control over how she responded to his changed behaviour. Although Carol (spouse, F3) knows that the behaviours Colin (PLWD, F3) displays are

due to bvFTD, she still feels frustrated at times when coping with this. She spoke about her feelings of sadness on seeing her husband as he is now, and compared that with how he was in the past. She also voiced how her relationship had transitioned from wife to carer, a fact supported by a family friend.

When interviewing Emma (spouse, F5) she voiced how she felt other people (e.g. family, friends, local community and other carers) would be judging her for placing her husband in long-term care. She compared herself with other carers who were in their 80's and 90's who managed to care for their partners at home. Emma accepted she had to place her husband in a nursing home due to his high safety risks when living in the community, e.g. walking in the road, walking around the streets late at night and neglecting his self care needs and his home environment. However, she still felt guilty, especially when she went out or enjoyed an activity.

In the current study, transitional relationships were not all negative experiences. As Harris (2009) found, the losses experienced when a partner has been diagnosed with dementia are evident, but for some couples, the experience could be a positive experience characterized by personal growth, appreciation of each other and small things, commitment and understanding. This appeared to have been in the case in the Allen family (F1). The relationship between Ann (spouse, F1) and Andrew (PLWD, F1) improved after the diagnosis as the bvFTD progressed. Andrew used to be controlling with some elements of

bullying behaviour (e.g. shouting, belittling comments, lack of warmth and empathy), but is now loving and affectionate towards Ann. He constantly expresses how much he loves Ann and appreciates her, he sings and is jovial much of the time.

In the observation visits with Andrew and Ann, I noted that the love and attention they paid each other was in marked contrast to the other case study families observed. When examining further why this spousal relationship appeared so different to the others, it was apparent that Ann's expectations of her relationship with Andrew differed because of her previous experience before the onset of bvFTD. Ann engaged Andrew in activities he enjoyed and made the most of the strengths he still had e.g. singing, eating out and visiting garden centres. I observed that she was able to predict activities or situations that could make Andrew feel uncomfortable and irritable, and avoided or diverted him from these before it became a problem for him, much as Betty expressed earlier. There was warmth and affection expressed and observed between Ann and Andrew throughout my visits. Ann had made necessary adaptations to their lifestyle to accommodate Andrew's needs and limit his distress, by the use of: routine; distraction; engaging in activities he enjoyed; visiting people and places that were aware of Andrew's behaviours (e.g. singing loudly, saying inappropriate things and not following usual social conventions). Ann felt really happy about the change in Andrew's personality, and his behavior towards her and stated that this was what she thought married life

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would be like. However, she also reported feeling sad, as she knows that his condition can only get worse over time and is life limiting.

In contrast to Ann and Andrew, in the other spousal relationships I observed, I noted that when a spouse was talking about the PLWD and the changes since the onset of bvFTD, they tended to: reduce eye contact; lower the volume of their voice; sounded tired; spoke in more of a monotone and their body language became more closed and stooped. When they were speaking to the PLWD they tended to raise the volume of their voice, and give short instructions rather than hold conversations. This was not surprising however as all participant PLWD in this study had significant communication difficulties and a very limited response to direct verbal communication (see chapter 6, Table 6.2). These communication difficulties were also observed when the person with bvFTD was in the presence of their children.

7.2.3 Family connectedness

In the current study, some families (F2, F4 & F5) described the relationship between the parent with bvFTD and their children as being characterized by arguments, withdrawal and sadness. This was especially noticeable in families with children still living at home, where the symptoms of bvFTD influence everyday life in a very direct and immediate way (see Box 7.3). This negative effect on the parent-child relationship was also noted in other studies (Allen *et al*, 2009; Luscombe *et al*, 1998; Roach & Keady, 2008).

Box 7.3: Quotes from participants in the current research study regarding parent-child connectedness.

"When our son came back from university after six months Brian (PLWD) showed no signs of recognition at all. He (son) is very reluctant to come home now, as I don't think he feels he fits in. In the event of course, he feels guilty that he isn't here and helping us. They (sons) know as well as I do there is only horribleness to come". (Betty, spouse, F2, interview).

"He (Diana's son) went up to her to give her a cuddle and she bit his nose so badly and they had a problem getting her off him. Her husband was distraught, her son was destroyed, her other son scared and she didn't know what she had done. Our son in law said he wouldn't take the boys to see her again." (Diana's mother, F4, interview).

"We had a holiday booked with our other daughter and her 2 little girls. I didn't want to go. I was torn between this family and that family and I knew the only way I could go would be if I could maintain daily contact with Diana (PLWD) and I didn't want to burden Darren (spouse) with daily calls. I wrote a letter to the home saying, "I'm away but my aunt and sister will be phoning so they could report to me" but reiterated that Darren was next of kin. The staff told him about the letter he was very angry saying we circumnavigated his authority and we "were not fit parents!" If we could do such a thing he felt very strongly that we could not see our grandsons either. He told our eldest grandson what we did and he said he didn't want to see us anymore". (Diana's mother, F4, interview and also backed up with letters between Diana's mother and Darren).

"He (Eric, PLWD) didn't speak to our daughter for 7 months, can you imagine that? I remember in the October saying to him, "Look why are you doing this? Why are you letting this happen? Its silly, what would happen if she got married?" and he would say, "Urgh, I don't want to be there anyway!" and he would be rude about both daughters. He would say things like, "It doesn't matter anyway. I find her very narrow minded". He was very rude about them and said I don't mind if I don't see them again." (Emma, spouse, F5, interview).

"Anyway we got the diagnosis and people said "I am sure the girls will be better, now they know, that'll be great". Everybody assumed they would both go, "Oh, poor old dad, give him a hug, poor man, he's ill" and they didn't. (Emma, spouse, F5, interview).

The Brown family (F2) reported that when Brian (PLWD) was diagnosed, his condition progressed rapidly and he did not recognise his sons consistently. As a consequence their son, who is at university remains reluctant to go to the family home when he has study breaks, which causes his mother sadness.

Betty (spouse, F2) tries to keep contact with their son to maintain their connection but misses day-to-day communication and involvement, which was an important part of their relationship before Brian's diagnosis.

Brian's (PLWD, F2) lack of communication and apparent lack of interest in his family and others was observed during my visit. He spent his time walking in and out of the room, sweeping the same bit of floor, then standing and staring. Any attempt to engage Brian in conversation resulted in either a one-word answer or he would stare and then walk away. According to his wife this is how he behaves when anyone is in the house, including his sons' and herself.

A particularly traumatic experience occurred to one of Diana's (PLWD, F4) sons (who was under 10 years of age at the time). During a visit to his mother at the care home, she started to hug him and then bit him very seriously on his nose and would not let go. As a result, the sons no longer wanted to visit their mother. There did not appear to be any intent or malice in this behaviour towards her son, but understandably Darren (spouse, F4) and Diana's sister have now made the decision that their children cannot visit Diana in case they are harmed. The decision to cease the children's visits further reduced the social connectedness of Diana with her children and her sister's children.

The parents of Diana (PLWD, F4) displayed love, affection and concern for Diana even though both stated she does not appear to reciprocate this. In my

observation visit, I noted that Diana's mother talked to her daughter in a conversational style with a great deal of warmth, friendliness and empathy. Diana's mother kissed, stroked, cuddled, smiled and maintained eye contact with Diana throughout the visit. Diana has no verbal expression, but does make vocalisations, such as shouting and various noises, however they do not appear to be in response to anything or express any emotion in particular. During the visit Diana's mother tried to engage Diana in everyday activities of living, such as eating and drinking, walking in the garden and spending time in her room and sitting room. This relationship was different to the ones observed in the Brown (F2), Cooper (F3) and Evans (F5) families as there was obvious love, affection and care expressed by Diana's mother, but this could be due to being a mother-daughter rather than a spousal relationship.

Emma (spouse, F5) reported that her daughters felt that the damage that had occurred to the parent-child relationship due to the change in behaviours and verbal communication was so great they could not forgive or forget. In addition to this, they had both read on the internet that bvFTD could be genetic, and they had worries over whether they would have bvFTD later in life. As a result of their worries and concerns, both needed to undergo counselling after their father was diagnosed to help them to come to terms with their thoughts and feelings towards him, and to help them face the future. As they did not consent to be interviewed, due to the issues they were facing, I was unable to explore this with them.

The distress displayed by children in this study has also been identified in other studies (Allen *et al*, 2009; Luscombe *et al*, 1998; van Vliet *et al*, 2010; Denny *et al*, 2012). Various reasons for this distress have been given including: lack of information and support (Denny *et al*, 2012); grief and loss (Denny *et al*, 2012; Svanberg *et al*, 2010); social isolation due to embarrassment (Hutchinson *et al*, 2014); behavior and personality change of parent (Denny *et al*, 2012).

However, some children adapt well to living with a parent with young onset dementia by progressing through grief to emotional detachment, and then begin to take on more adult responsibilities ("becoming grown up") (Svanberg *et al*, 2010). There is a risk that children in households with young onset dementia could appear to be coping when in reality they may need time and space to talk about their feelings. The children from the Brown (F2), Cooper (F3) and Evans (F5) families declined to be interviewed, so I could not ascertain from them how they were maintaining their social connectedness within, and external to the home, and how they were coping with their parent with bvFTD. In addition, I decided not to interview the young children in the Davies family (F4) due to the trauma they were experiencing, for which they were receiving counseling.

The parent- child relationship difficulties described in the Brown (F2), Davies (F4) and Evans (F5) families were not in evidence in the Cooper family (F3). Their children were married and living in their own household's and helped their mother by giving her space and time to express some of the difficulties she faced. Both daughter and son suggested their dad attend a day centre for one

day per week in order to give their mother a break. They also gave their mother respite time by regularly looking after their father for the day. While this provided important support for her, Carol noted that when Colin is at the children's houses he follows his usual routine and does not really communicate with them, preferring to complete his crossword or Sudoku. His wife interpreted these visits as part of his routine and, as such, they no longer held any personal or social meaning for him:

> *"I've noticed that Colin will frequently ask our son, daughter or his brother when he can come to see them but when he's there he simply sits and completes crosswords as he would do at home." (Carol, spouse, F3, interview).*

During my visit, I observed that Colin did not communicate with anyone around him (present were family friend, wife and myself) and continued to complete the Sudoku. Even though we tried to engage him in conversation, he would not answer at all other than to talk about the numbers on the Sudoku grid. When we were in conversation, he would regularly interrupt and talk over us to say what he had completed on the Sudoku. This inability to join in the usual social norms of behaviour could lead to a further reduction in Colin's social connectedness.

As discussed in chapter 1, family relationships can be an important source of support and connectedness (Allen & Killick, 2008). However, when a person is diagnosed with bvFTD some families experience a difficulty adapting to the

diagnosis and instead of providing the connectedness and support expected, respond in a different way (Riedijk *et al*, 2006; Diehl-Schmid *et al*, 2013). The families in the current study reported difficulties and disappointment with family members outside of the marital home (see Box 7.4).

Box 7.4: Quotes from participants in the current research study regarding birth family connectedness.
"Even my own mother who sees it from the inside because she stays with us, finds it difficult to accept what is happening. I find this very upsetting. They seem to play it down or not comprehend how this must be for me trying to work, run a house, raise a family and becoming a carer." (Betty, spouse, F2, interview). *"When his family visit they haven't at any time tried to pull me aside to say, "How are you coping?" That's really difficult." (Betty, spouse, F2, interview).* *"At least one Thursday a month Colin (PLWD) goes to see his youngest brother but most of his family and friends are dropping away now." (Carol, spouse, F3, interview).* *"I dislike going to see Diana (PLWD), I find it incredibly difficult to communicate with her. I watch Mum effortlessly interact with my sister, I can't and just don't know what to do when I get there. It causes pain when I visit her and I feel bad. I used to take my girls but can't now as I don't want them to see her like this and she bites now. I try to go every 10 days to 2 weeks even though I find it difficult." (Diana's sister, F4, interview).* *"Our oldest granddaughter, who was under 5 years of age, started behaving badly and started to get really distressed if anyone around her was ill, if her mummy was ill or I was. She started to wonder why they could not see their Auntie because we had to stop them seeing her due to the biting. She started talking about dying, then the next thing we knew she had to visit a behavioural therapist". (Diana's mother, F4, interview).* *"Eric's sister visits him about 4 times per year. Her view is she wants to preserve him from risk and as he is in a home, he is safe. She doesn't understand that my daughters have been traumatised by their Dad's behaviour and don't want to see him" (Emma, spouse, F5, interview).*

As previously discussed in chapter 6, the birth family relationships of the person with bvFTD had deteriorated in the Allen (F1), Brown (F2), Cooper (F3), and Evans (F5) families before the diagnosis, and having the diagnosis did not improve these relationships. These families felt disappointed that extended family members (external to the family home) failed to recognise the extent of the problems the person living with bvFTD and their family faced. This finding has been reported in other studies (Lockeridge & Simpson, 2012; Riedijk *et al*,

2006). As a result of this lack of understanding by others, families in the current study felt that their caring role was underestimated and undervalued, and they resented this. The Brown, Davies and Evans families (F2, F4 & F5) described how they did not feel the same sense of connectedness to these extended family members as they did before their family member was diagnosed with bvFTD.

The relationship between Diana's parents and spouse (Darren, F4) deteriorated further in that there were frequent arguments over Diana's care needs. Darren thought that Diana's birth family were interfering or usurping his role as next of kin, which culminated in him restricting the children's visits to their maternal grandparents, Diana's sister and her children. This has reduced the social connectedness between members of Diana's birth and marital family. All the members of Diana's birth family and the marital family are now receiving individual counselling to help them to work through their feelings of distress following the diagnosis and the rapid progression of bvFTD.

7.2.4 Social contacts and networks

Friendships and social connectedness are an important part of being a human being (Harris, 2011), but unfortunately people living with dementia often experience a reduction in these due to others people's reactions to the changes. This appears to be particularly so in people diagnosed with bvFTD

due to the changes in personality, social functioning and the behaviour displayed.

The families shared the diagnosis with their friends and social networks as they expected them to understand and support them to cope. Unfortunately, they found that this was often not the case, and were upset when close family and friends did not understand or support them more (see Box 7.5). They also thought their social connectedness could have been maintained longer, as friends and colleagues now had a reason for the changes in behaviour and personality. The families in the study reported that now they had a diagnosis, friends and colleagues would not attribute these changes to the PLWD being "difficult", rude, or choosing to act in that way.

Box 7.5: Quotes from participants in the current research study regarding social contacts and networks.
"Some of the church members can't deal with Andrew's (PLWD) behaviour. I have always offered to help others at church and expected others to help us, but no one did". (Ann, spouse, F1, interview).
"Brian (PLWD) even turns people away, a very rare visitor came the other day and he said, "No" and shut the door on them. He didn't recognise them and just shut the door". (Betty, spouse, F2, interview).
"The friend (who is a nurse) who phoned me about my husband's behaviour at the social event hasn't been to visit once or called me since that time. These are friends he has had since 32yrs of age!" (Betty, spouse, F2, interview).
"I think other people avoid the family and don't understand what is happening. I think they are sympathetic but don't know how to respond. They come stay for a few hours then just go again. I don't think many people understand what the disease is". (Family friend, F3, interview).
"Because Colin (PLWD) looks normal and he is quiet I don't think people realise he has a problem. I don't think people understand his wife's situation. He needs someone with him 24hrs a day." (Family friend, F3, interview).
"I try to keep Colin's (PLWD) friends going as much as possible. I did have a very down moment before Christmas and had a serious go at three of his friends who used to drink with him but who he

> *has not seen for ages. One said he had been a coward, as he didn't know what to do. I said, "You and me both!" Most of his family and friends are now dropping away. We do still have some joint friends but social events are becoming less possible due to Colin's behaviour." (Carol, spouse, F3, interview).*
>
> *"Diana (PLWD) did not in any, way shape or form want any of her friends to visit but because she was who she was, her friends have been in touch with me, every single one of them, her colleagues and friends. Darren asked them all not to contact him." (Diana's mother, F4, interview).*
>
> *"They are probably saying, "Well I want to remember her as she was", because that's another thing people do. They are lucky they get that choice!" (Diana's mother, F4, interview).*

The families felt a sense of disappointment, annoyance and bitterness about how other family members; friends, work colleagues and local community avoided not just the PLWD, but also the whole family both before and after diagnosis. Pre diagnosis, Brian (PLWD, F2), Colin (PLWD, F3) and Eric (PLWD, F5) had long-term friends with whom they had close supportive relationships, but once the bvFTD progressed, these friendships ceased for Brian and Eric. Carol (spouse, F3) confronted three of Colin's friends about their apparent lack of understanding and support. One of them said he had been avoiding the contact, as he did not know what to do or say, so it was easier to avoid them, but he resumed contact after the confrontation. Carol's friend highlighted an important point about Colin looking "normal" but being different to how he was before, which often lead to misunderstandings about why he behaved as he does in social situations. Evans & Lee (2014) highlighted how some spouses of a person with bvFTD describe their experience with their spouse as still looking the same but there is a "loss of the person within".

Diana's mother voiced her appreciation that Diana's friends and work colleagues had not given up on her daughter, even though Diana (PLWD, F4) told them not to visit or actively avoided them when they did so. Darren (spouse, F4), said he preferred not to be contacted for updates from friends and family, as he wanted to concentrate on his sons and trying to maintain as normal a life for them as he could, so Diana's mother offered to do this. The behaviour of Darren could be construed as ceasing all social connections with anyone previously involved with Diana. An alternative explanation could be he is progressing through the process of emotional detachment, as described by Walters *et al* (2010) or "moving on" as described by Hellstrom *et al* (2007).

Not all families in the current study experienced the same reduction or cessation of social contacts and networks. Since diagnosis and the progression of bvFTD, Andrew Allen (PLWD, F1) has been much more sociable and he enjoys going out to cafes, singing for the brain group, respite, and local community events. So although previous social connections have ceased, Ann (spouse, F1) and Andrew have developed new ones, focused around living with dementia. Both Ann and Andrew enjoy this time together, meeting with others in similar situations and sharing experiences. I observed Andrew and Ann in the local community in the various activities, and it was apparent that they both gained a great deal from attendance, both in the actual activity and also in the social network opportunities it gave:

"This is the happiest I have ever known him. Andrew is content with life." *(Ann, spouse, F1, interview).*

Ann states that she only found out about these services and supports by searching for them herself on the internet and through the media, and wondered why these were not suggested by health and social care professionals who had been involved with them after diagnosis. This is an important point, and implies that if families are given timely and appropriate post diagnostic support, social connectedness could be maintained and new contacts and networks developed.

7.2.5 Contacts with the local community

Although Betty (spouse, F2) shared the diagnosis with family and close friends, she does not share this with her neighbours or the local shop that he goes to regularly. When asked why, she said it was because she felt embarrassed about his behaviours, and what the neighbours and local shop would think, and do, if they knew about the diagnosis. Although she has not shared the diagnosis with the local shop and neighbours she is certain they must realize Brian (PLWD) has changed but not necessarily why:

"I try to avoid telling people what the issues are. I haven't been to the local shop to speak with them about my husband. I think I have avoided the embarrassment of going there." *(Betty, spouse, F2, interview).*

Carol (spouse, F3) shared the diagnosis widely with family, friends and the local community in an attempt to raise awareness of why Colin behaves as he does. She stated that she thought if they knew why he behaves like he does, he may be able to continue attending activities and events:

> "However I have asked the club to tell other golfers about my husband and frontotemporal dementia to aid with understanding as I don't feel at all embarrassed about telling people what is happening. I don't mind who knows, the more people who know the better". (Carol, spouse, F3, interview).

The PLWD in the Allen (F1), Brown (F2) and Evans (F5) families were actively excluded from services and support in the local community as a direct result of their behaviours (e.g. invading personal space; pushing in front of people in queues; voicing inappropriate comments and taking things from others) (see Box 7.6).

Box 7.6: Quotes from participants in the current research study regarding the PLWD conditions of attendance or exclusion from activities.
"Brian (PLWD) went a few times but then the social worker got in contact with me to say the centre believed they were not suitable for my husband but gave no alternatives." (Betty, spouse, F2, interview).
"The gym asked us to send someone with him because of his behaviour and then the golf club phoned and said they couldn't cope with his behaviour towards other players." (Carol, spouse, F3, interview).
"Eric was always interested in classic cars and I organised a voluntary job at a motor museum. He could go as long as his carer went with him but they had to say he couldn't go anymore as they had a few embarrassing incidents, like his trousers fell down in front of a load of school kids and he was swearing and stealing food from the canteen". (Emma, spouse, F5, interview).

Some groups or activities made it a condition that in order for Colin and Eric (PLWD, F3 & F5) to attend, they would have to be escorted due to concerns about their behaviour. This finding is supported by Oyebode *et al*'s (2013) study, who found that loss of inhibition leading to socially embarrassing behaviours provoked strong reactions in others, which could lead to withdrawal of contact and some degree of stigmatisation.

Eric (PLWD, F5) is now in a care home and socialises with the staff and residents. He socializes more now than when he lived in the community, and Emma (spouse, F5) stated he has a much better quality of life, as the staff and residents understand and accept his behaviour:

> *"Eric (PLWD) goes past everybody in the home and says, "How are you doing mate, you alright?" I mean he doesn't talk about anything really, it's all the same conversation, his conversation, it's a repeat of the day before and the day before that, but he's quite lively. The other day he said, "You are looking bonkers" and other personal things like, "You've got a fat bum" but they are used to it. He is in a very loving, kind and relaxed place now". (Emma, spouse, F5, interview).*

Local communities differ in how they behave towards people living with dementia despite the focus in the United Kingdom on Dementia Friends and Dementia Friendly Communities. The families in the study described how they felt that the local community were less understanding of their family member

(PLWD) as they were younger, looked "normal" and had behaviour disturbance rather than problems with their memory, the more common sign of dementia.

Families in this study reported that the changes in social connectedness were experienced both pre and post diagnosis for the whole family not just the family member diagnosed with bvFTD. They voiced their feelings of disappointment, anger and frustration about many of their previous social connections. They also identified thoughts of being ignored and abandoned, especially by the people who they assumed could help and support them e.g. health professionals, family and friends. This abandonment and feelings of isolation added to the stress and burden experienced by the families.

7.3   Theme 6: "Grief and loss".

The spouses in the study spoke about the losses (which included: loss of relationships; change of role; loss of personal freedom and control; loss of social connectedness) and grief they and their family experienced (these have been covered in this chapter and chapters 5 & 6). In addition, it was noted in observations, documents and interviews with family members, the person living with the diagnosis of bvFTD experienced a loss in role, personhood and career. Whilst all PLWD voiced their feelings about the loss of their career, none expressed any thoughts or feelings of loss of anything else, including their family relationship or social contacts and networks with friends or local community. This could be due to the stage (middle to late stage) of bvFTD that each was experiencing, and the effects on the parts of the brain that regulates

social behaviour, insight and empathy or maybe indicative of something else, but exploring this was beyond the scope of this study.

This section explores the losses and grief a family living with bvFTD can experience and suggest some possible reasons for why this can occur.

7.3.1 Changes in social citizenship

The families in this study started to experience changes in their sense of social citizenship at the early stages of the onset of bvFTD, often years before a family member was diagnosed. As the change to social citizenship in the study families commenced before diagnosis, it could be argued that this was not entirely due to the local community discriminating against someone with dementia. It would appear the reduction in social connectedness and citizenship could be due to the PLWD's gradual changes in behaviour, personality and social functioning from the onset of bvFTD.

Before the diagnosis, the person in the early stages of bvFTD can also become more introverted, withdraw from social engagement, fail to reciprocate in relationships and cease to maintain or initiate social networks, which can also lead to a reduction in social connectedness. As their family member's behavior, personality and social functioning progressively deteriorated, the study families tended to avoid social situations due to feelings of embarrassment, and this started to have an impact on the whole family's social connectedness.

7.3.2 Loss of relationships and grief

Families in this study stated they missed someone to talk to, to discuss and plan things, and to share their hopes and dreams. Adams *et al*'s (2008) study on losses and relationship quality in caregiving, also found that this loss of intimate exchange contributed to role overload and depressive symptoms. This was observed in the spouses within the study, and was most noticeable where the PLWD had reduced communication abilities as well as the other changes associated with bvFTD.

Children in the study (F2, F4 & F5) found it difficult to adapt to the changes in their parent with bvFTD. According to their non-affected parent, they displayed varying degrees of psychological distress that necessitated formal counseling in order to manage their emotions (see Box 7.7).

Box 7.7:   Quotes from participants in the current research study regarding their children's feelings of loss, grief and distress.
"The oldest son had a bit of a breakdown at school suddenly in his exam year. He was put on Prozac for 2 years. He said this was because of his dad. (Betty, spouse, F2, interview).   *"Diana's (PLWD) son has been back to visit his mother once since the when she bit his nose. He said he didn't like her and didn't want to go again nor does his brother. (Diana's mother, F4, interview)*   *"Unfortunately Eric's (PLWD) behaviour resulted in a breakdown of his relationship with us. The girls have been deeply disturbed and have both had counselling. They prefer not to see him anymore for their own well- being." (Emma, spouse, F5, interview).*   *"I remember the younger daughter saying to me in tears when I had a row with her about seeing her Dad, "I am not sure if I want to let him into my life anymore". He has been horrible to them and said he didn't want them anymore. They had to learn to build a*

> *protective shell around themselves, and they don't want to let the shield go, she said "Why would I want to open myself up and let Dad back in to wreck my life because he is only going to die, and why do I want to do that to myself?" She didn't want to let in her emotions of love for him because she said, "I've said goodbye to him already, I'm going to have to say goodbye to him again. The eldest daughter says things like "He is not my Dad anymore, I don't even like this person, and I don't know what I am connecting to.""" (Emma, spouse, and daughters, F5, interview).*

The non-affected parents spoke about the conflicting emotions they experienced when they tried to balance the needs of all parties within the family. This was particularly problematic in the Brown (F2), Davies (F4) and Evans (F5) families and resulted in additional distress for the families.

In family 5, although the daughters did not immediately reengage with their father (Brian, PLWD), after receiving their counseling, they are now beginning to visit him in the care home, and their relationship is improving. The care home is very welcoming and supportive to families who have a loved one in residence, and has a very homely atmosphere that makes it easier for both daughters to visit and reengage with their father.

In the Davies family (F4), Diana's sister at times thought her mother spent so much time with Diana (PLWD) and not enough with herself and her children. Diana's sister felt guilty having these thoughts, as she knew that Diana's bvFTD was rapidly progressing and she would be unlikely to live much longer.

The relationship between Diana's parents, sister and son in law (F4) gradually deteriorated due to differences of opinion over the assessment, diagnosis and treatment of Diana. This eventually led to Darren (spouse, F4) stating that he did not want contact with any of Diana's family:

> *"I would go upstairs (at the home) to ask what my daughter (Diana, PLWD) had had for her medication because she was banging her head, shouting, screaming or tearing things up. I would get distressed as well and end up in tears. I would phone my son in law and say "They are not doing this or not doing that" and it was just insidious and ever so quick that he started to resent it. He stopped talking to me, he said to the staff "If my mother in law asks for anything then you should come straight to me". (Diana's mother, F4, interview).*

> *"When I visited the home Diana was in pain and I phoned our son in law and probably I didn't handle it properly and said "Please get some sedation for her". That just dug the hole deeper, and after that we were told not even to acknowledge our eldest grandson in the street. He also said that he didn't want anything to do with any of us including Diana's sister and her little girls. He said I had destroyed them as a family." (Diana's mother, F4, interview).*

In the above quotes, Diana's mother spoke about her distress and feelings of powerlessness when she visited her daughter in the care home. These feelings and thoughts were also expressed in letters to Darren (son-in-law). By her own admission, she would question and instruct the staff at the home if they were not caring for her daughter as she would expect, and would insist they respond to her concerns. This irritated the staff and they would phone Darren (spouse, F4) and complain about her behaviour towards them. He in turn would get

angry with Diana's mother and an argument would ensue. The situation escalated until the two families stopped communicating and Diana's mother and father were told to cease contact with their grandsons.

Chan *et al* (2013) indicated that families living with the effects of dementia experience anticipatory grief due to the multiple losses that can occur after diagnosis, and this was observed in the study families. Denial and avoidance was noted and reported in the extended family and some friends, with them saying to the families:

> *"I can't visit as I don't want to see them like this, I want to remember them as they were". (Various family and friends to families 2, 3, 4 & 5)*

This led to the families living with the effects of bvFTD feeling angry, disappointed and distressed by the apparent lack of understanding of what the family was coping with, leading to increasing feelings of isolation. All families (with the exception of the Allen (F1) family) exhibited signs of depression and distress but this was not in evidence in the person living with bvFTD. In contrast, the Allen family did not exhibit any signs of depression and on the contrary appeared happy in their life at the present time, indicating that grief and loss reactions are not always experienced when living with bvFTD.

The families accepted the reality of the diagnosis but felt very distressed about the effects that it had on themselves as a family. The Davies (F4) and Evans (F) families stated that they wished their family member had cancer instead of

bvFTD and felt they would have coped better, especially as their loved one would then have had their personality and behaviours intact:

> *"It is an ugly, ugly illness. I wish I had a pound for every time I thought I wish she had cancer." (Diana's mother, F4, interview).*

> *'I often say to people, I could have dealt with it better if it was cancer". (Emma, spouse, F5, interview).*

> *"I put my hand on my heart and I just want Diana (PLWD) to die because to watch her and to know what she was and to not be able to talk to her is hell." (Diana's mother, F4, interview).*

Diana's family (F4) feel very distressed for thinking about their daughter like this but realise that she will never get better and worry she may be aware of what is happening to her. Diana's mother visits very regularly, but her father and sister state they have to "force themselves" to visit, as they feel so distressed after seeing her. Meeting and observing the family and reading some of the letters written by Diana's mother revealed the depth of feelings and grief experienced by the family. The letters indicate how, as Doka (2010) suggests, grief intensifies as the PLWD condition worsens and their personality, behaviour and social functioning become further removed from their pre morbid state (Doka, 2010).

The breakdown in communication and animosity between Darren (spouse, F4) and Diana's parents resulted in Darren making decisions about Diana's care, treatment and end of life decisions without discussing this with her birth family:

"My son in law said he had sorted out our daughter's (Diana, PLWD) funeral arrangements and that her ashes would be split, he would have half and we would have the other half. He was saying this matter of fact as though she was a bag of onions but he was talking about my daughter". (Diana's mother, F4, interview).

"I can't understand why all of this has happened to our family, all of this is nothing to do with Diana (PLWD) and her illness. I will never forgive Darren for what he has done. I feel that the family has broken up already and the family has lost focus on Diana". (Diana's sister, F4, interview).

Diana's mother and Darren (spouse, F4) have now started to try to communicate in the best interests of Diana (PLWD, F4), but he will not communicate with the rest of the family. The eldest grandson is still refusing to have any contact with his grandparents:

"I will not over step the mark and I said that to our son in law. We have been through a year of hell that hasn't stopped, but at least he and I know now why I did what I did. We have that unspoken understanding. I don't feel the anger towards him that I did. I've never dismissed his distress. He says visiting our daughter is a "necessary chore". I feel that with our daughter I visit her as her mum." (Diana's mother, F4, interview).

The breakdown in the relationship between both of Diana's families is likely to have long-term effects on the families involved. All members of both families are receiving counselling to help them to manage their emotions, feelings and thoughts. Diana's parents and sister hope they will be able to rebuild the relationship with Diana's sons.

Emma (spouse, F5) stated she felt guilty when Eric (PLWD) moved out into his own flat and ultimately into the care home. However, she still provided active daily care when he lived in his flat and visits frequently now he is in the care home. Emma also spoke of the losses she has experienced e.g. loss of a future, loss of a partner, loss of relationship:

> "I feel deeply sad now. It's like, say you have someone who has died, or a child who might have died, you go on with your life. I can still go out and have fun. I go out and do things, I can still enjoy things. I don't go around super depressed all the time, thinking there's no point in life. I sort of, its deep inside me, it's like a deep sadness. Sometimes it's more at the top than others, sometimes you get on with it and sometimes it's sort of on a parallel universe. Sometimes it hits me when I am out and about and I think this is all a bit superficial. Now I've got this sad space because I've lost my sort of mate and the future we thought we would have." (Emma, spouse, F5, interview).

Emma also spoke about the feelings of relief she felt when Eric went into a care home, as he would be safe and therefore she would not have to worry, however she felt guilty doing this. These feelings have been expressed by spouses in other studies (Cabote *et al*, 2015; Mioshi *et al*, 2009; 2013).

The Allen family (F1) is unusual in that the spousal relationship improved after the diagnosis:

> "How Andrew (PLWD) is now is what I thought I was marrying. This is why it seems so unfair". (Ann, spouse, F1, interview).

"It's really bizarre that this is the bit I will always remember which is positive, I can ditch all the bad memories…. I want to remember how his behaviour is towards me now". (Ann, spouse, F1, interview).

Ann (spouse, F1) and Andrew's (PLWD, F1) experience is not one of loss, but of gain, their relationship is much stronger now than it has ever been during their marriage, and both have a very positive and loving relationship. Ann says that she feels very sad about this, as she knows he will continue to deteriorate due to the bvFTD, and this time of such positive change will eventually end. This is contrary to what has been found in the other study families, who expressed the opinion that the family member's death would be a relief because they thought the person they were before was no longer there. It should be noted that in contrast to Ann and Andrew's improving spousal relationship after bvFTD was diagnosed each of the other families in the study had loving, caring and in their words, *"strong relationships"* before the onset of bvFTD so their experience is one of *"loss of the person and relationship that was"*. This is supported by Doka & Aber (2002) who wrote about the deep sense of loss when a person's identity changes so much that family and friends no longer recognise them as the same person. Evans & Lee (2014) wrote about the "loss of the person within" to describe how the person still looks the same, but due to the changes in personality, social functioning and behaviour they are no longer who they were.

7.4   Summary and transition

In summary, families in this study identified the difficulties they experienced obtaining appropriate post diagnostic advice and support (Theme 5: "Now we know what it is"). They thought that this support would have helped them to make sense of what was happening, and assisted them to cope with the changes to their roles, relationships and social connectedness.

The study families expressed their feelings of grief and loss as bvFTD progressed in their family member (Theme 6: "Grief and Loss"). They believed that the social connectedness and social citizenship of the whole family gradually altered due to the changes in personality, behaviour and social functioning of the person with bvFTD.

In Chapter 8, I will progress my discussion on the findings by discussing the broader social and health issues to emerge from the study, placing my findings within the context of previous knowledge in this area. Implications for practice and recommendations for future research studies will be identified.

Chapter 8: Discussion, Implications for Practice, Recommendations and Conclusion

8.1 Introduction:

In chapters 6 & 7, I presented the study findings and provided initial interpretations of specific issues, highlighted in the data collected from 5 case study families over the period 2014-15. The findings from the Brown (F2), Cooper (F3), Davies (F4) and Evans (F5) families were similar in that they experienced a marked reduction in the quality and quantity of their family and social relationships, contacts and networks, and consequently this had an effect on their social connectedness. The Allen family (F1) was different in that the premorbid and pre diagnosis connections and relationships were not

maintained, but new ones were established and the marital relationship strengthened.

In this chapter, I will progress my discussion on the findings in chapters 6 and 7, by considering the broader social and health issues to emerge from the study, placing my findings within the context of previous knowledge in this area. From this, implications for practice and recommendations for future research studies will be identified.

8.2   Early recognition and diagnosis

Early recognition and diagnosis of a condition is important to ensure that appropriate advice, support and interventions can be given in a timely way (Iliffe *et al*, 2003). However, van Vliet *et al* (2013) report that in cases of young onset dementia, it can take an average of 4 years to receive an accurate diagnosis. Having an early diagnosis can enable the person with the diagnosis to actively participate in making plans and decisions for their future, and the later stages of their life (Dening *et al*, 2011). In addition, early diagnosis and post diagnostic support can reduce the amount of distress the person with the diagnosis and their family experience, and reduce relationship breakdown (Nicolaou *et al*, 2010; Johannessen & Moller, 2011). Early diagnosis is predicated on early recognition of the signs and symptoms of bvFTD. In this study, the families recognised signs of behavioural and personality change and sought help, but health professionals were poor at diagnosis making it difficult for the families to

receive appropriate early advice and support. The reasons for this are outlined below.

8.2.1 Early recognition

The early signs of bvFTD are insidious in nature and include changes in behaviour, personality and social functioning of the symptomatic person (Fernandez-Matarrubia *et al*, 2014). In the absence of memory problems and older age, families do not consider the symptomatic person could have a young onset dementia (Lockeridge & Simpson, 2012). People who are symptomatic, and their family; seek alternative, perhaps more commonly understood reasons for the changes they have noted, i.e. stress, "mid life crisis", physical or mental ill health and relationship difficulties (Johannessen & Moller, 2011; Lockeridge & Simpson, 2012; Griffin *et al*, 2015; Pressman & Miller, 2014).

Families in the current study described their experience of recognizing that their family member's behaviour, personality and social functioning was changing (see chapter 6, Table 6.2). They identified how they sought potential reasons for this, initially attributing the changes they noticed to: mental health issues (see chapter 6, Box 6.2), physical health problems (see chapter 6, Box 6.4) or relationship breakdown (see chapter 6, section 6.4). The findings in the current study appear to accord with the previous studies by Johannessen & Moller (2011), Lockeridge & Simpson (2012) and Pressman & Miller (2014). As identified in chapter 6, the families in the current study eventually attributed the progressive changes in behaviour, personality and social functioning as

intentional on the part of the symptomatic family member, and this led to misunderstandings and deterioration in family and social relationships.

In the current study, once other people outside of the immediate family began to observe and comment on the changes, the families decided to seek a medical opinion (see chapter 6, section 6.5). At this stage, all of the families suspected that their family member had a treatable condition, and when diagnosed and treated, everything would return to "normal". This initial "explaining away", or "normalising", of the early symptoms of dementia have also been described by Daly *et al* (2013) and Lockeridge & Simpson (2012). This normalization is understandable given that dementia under 65 years of age is relatively rare (see chapter 1, Table 1.1 & Box 1.5). Also, the early symptoms of dementia can be similar to other common conditions, such as stress, depression, psychoses and relationship difficulties (Manoochehri & Huey, 2012; Pressman & Miller, 2014; LaForce, 2013). The long period of attributing personality, behavioural and social functioning changes to a cause other than bvFTD, lead to a marked delay in approaching a medical practitioner for an assessment or diagnosis. The findings show that this delay in diagnosis can lead to increased family distress and relationship breakdown.

The findings also indicated that pre diagnosis, the social connectedness of the whole family was negatively affected by the changes they, and others (e.g. extended family, friends, work colleagues, social network), observed in the symptomatic family member (see chapter 6). They began avoiding events and

social situations, being embarrassed by the behaviours displayed by their family member (see chapters 6 & 7). In addition, there was a reduction in the invitations to social events for the symptomatic family member and their family (see chapters 6 & 7). This indicates that family withdrawal from social life was accompanied by social exclusions in a two way process of loosing connectedness. It is clear that maintaining connectedness at the early stages is not under the control of families, but requires an accepting social context, something difficult to negotiate when diagnosis has not been made.

8.2.2 Diagnosis

Delays in diagnosis could lead to increasing family distress, lack of support, further reduction in relationships and, as mentioned above, negative changes to social connectedness (Allen *et al*, 2009). Studies have identified the multi-various reasons behind diagnostic problems in young onset dementia, including: medical practitioners lack of awareness, knowledge and skills (Cabote *et al*, 2015; Johannessen & Moller, 2011; DH, 2009; Luscombe *et al*, 1998); not listening to the family's concerns (Johannessen & Moller, 2011); symptomatic person's lack of insight (Clare, 2003; Stephan & Brayne, 2014; Rascovsky *et al*, 2011) and the symptomatic person's and family's presentation in the medical consultation (Lockeridge & Simpson, 2012).

Obtaining a diagnosis was difficult for all the current study families, an issue they described as frustrating and distressing. Initially they delayed booking a medical appointment for a prolonged period until there was a family crisis. When they did book an appointment, they felt extreme pressure, and wanted

immediate answers. This diagnostic delay compounded their feelings of anxiety and distress, a finding also identified by Chow *et al* (2011). The current study families felt that they had to "fight" for the correct diagnosis after an incorrect one was given, adding to their distress (see chapter 6, section 6.6). In a study by Werner *et al* (2009), it was found that 30-50% of younger people with dementia were wrongly diagnosed initially.

In the current study, spouses and family felt that medical practitioners did not listen to them regarding the extent of the changes to behaviour, personality and social functioning of their symptomatic family member (see chapter 6, Box 6.10). This apparent lack of attention to family experiences could reflect medical practitioners lack of knowledge and skills regarding recognizing bvFTD as a diagnostic possibility (Davies-Quarrell *et al*, 2010; Werner *et al*, 2009). In addition, medical practitioners are bound by an oath of confidentiality and their duty is to their patients, and as such they will ask the patient (in this case the symptomatic person) their viewpoint, and whether or not they can share information with their family and others (GMC, 2009). If the symptomatic person does not have insight into their condition and how it is affecting them, they are less likely to confirm the changes the families have observed or to consent to having information shared. Hughes & Louw (2002) state that in these situations the medical practitioner should consider the context of the situation and recognise the role of the family and carer's needs for information and involvement.

Another important factor could be the presentation of the families in the medical appointment. In the current study, families spoke about how, during the appointment with the medical practitioner, they were distressed and anxious when voicing their concerns over changes in marital and family relationships. Whilst doing this, they described how their symptomatic family member presented as calm, sitting and listening to what was said in a dispassionate way. A medical practitioner observing could misconstrue this as the non-symptomatic person having the problem e.g. anxiety, depression, stress or relationship disturbance rather than the person who has the symptoms of bvFTD.

A study by Stewart (1995) indicated that the unequal distribution of power and control in a medical appointment between the practitioner and patient could lead to the incorrect diagnosis, communication problems and dissatisfaction. Stewart (1995) suggests this could be addressed by power sharing[5] to address the imbalance, improve communication and health outcomes. The issue of medical power and control emerged in the current case studies during the family's attempts to obtain a diagnosis from medical practitioners. It appeared that the medical practitioners had a tendency to adopt the "expert role" in consultations, when in fact they did not have the knowledge and experience to do so. This may have prevented them from listening to the family's experience of living with a symptomatic family member. As a result they missed the symptoms of bvFTD that should have alerted them to the need for a referral to

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[5] Power sharing could include listening carefully to what the patient (or family) says, giving them a choice over treatment and care and actively involving them in decision-making.

a specialist practitioner for the correct assessment and diagnosis. Manoochehri & Huey's (2012) and Pressman & Miller's (2014) studies suggested that medical practitioners should listen to the concerns of families, as they are able to give a first hand experience of the changes indicative of bvFTD. If medical practitioners had followed this advice, a specialist referral and the correct diagnosis could have been given earlier, which may have reduced the distress the families experienced.

In the current case study families, I noted that families tended to use words like: "fight", "challenge", "I want you to listen", "proactive", "pushed" and "I do not want therapy", when recounting medical appointments. Such words and sentences suggest families experienced power imbalances with the medical practitioner when seeking a diagnosis and were trying to regain control of the situation. The families did this when they thought the medical practitioner was not listening or did not understand the severity of the situation (see chapter 6, Box 6.10). They insisted on attending the appointment with the symptomatic family member (F2, F4, F5), writing lists of symptoms (F1, F4, F5) and challenging the medical practitioner when an incorrect diagnosis was given (F1, F2, F3, F4, F5).

Once the diagnosis was obtained there was a feeling of relief as the families finally had a reason for what was happening to their family member. However there was also a sense of disappointment, anger, and isolation when they were

not offered appropriate information, advice and support (see chapter 7, Box 7.1).

8.3   Post diagnostic support

Post diagnostic support is important in developing an understanding of what the diagnosis means for the person diagnosed and their family. The goals of post diagnostic support are to: increase the families knowledge base regarding dementia; sustain and improve the psychological and physical health of family caregivers; maximize independence of the person diagnosed for as long as possible and delay inappropriate admission to hospital or long term care (Fortinsky, 2014). However, it has been identified that there is widespread variation in giving appropriate post-diagnostic support and advice (Fortinsky *et al*, 2010; Koch & Iliffe, 2011). Receiving timely post diagnostic support, focused on retaining and developing the family's links with their local community, family and friends, could ensure that their needs for social connectedness are achieved.

The study families stated that they were not offered appropriate post diagnostic advice and support following the diagnosis of their family member (see chapter 7, Box 7.1). This added to feelings of isolation, distress, anger and abandonment. They stated that they had to seek out advice and support for themselves through family members (F3), on-line (F1) and by repeatedly asking for advice and support from medical practitioners (F1, F2, F4 & F5) (see chapter 7, Box 7.1). This lack of support also compounded feelings of loss and

grief they experienced when they realized that this was a progressive life limiting condition and not something that could be cured or controlled (see chapter 7, section 7.3).

Younger people with dementia and their families have a greater need for emotional, psychological and relationship work than people diagnosed later in life (Nicolaou *et al*, 2010; Spurlinger & Furst, 1994; Johannessen & Moller, 2011; Diehl *et al*, 2003) as post diagnostic advice and support can help families adapt to the changing relationships and roles that occur with the onset of bvFTD (Daly *et al*, 2013; Nicolaou *et al*, 2010; Mioshi *et al*, 2009, 2013; Kipps *et al*, 2009). Post diagnostic advice and support could also enable families to develop coping strategies in order to build resilience, prevent distress and crises.

Post diagnostic advice and support could take the form of developing techniques and coping strategies with families to prevent and manage problematic behaviours (Mioshi *et al*, 2013; Lockeridge & Simpson, 2012; De Mattei *et al*, 2008), and regular respite and peer group support (Lockeridge & Simpson, 2012; Daly *et al*, 2013). In addition, the studies by Peel & Harding (2013) and Doherty *et al* (2009) indicated that families wanted an identified, knowledgeable professional to provide continuity, guidance and support throughout their journey with dementia to help them cope with the added responsibilities of caring for their family member with dementia.

The current study families stated that once their family member became symptomatic with bvFTD, no one outside of the immediate family understood their situation, and there was a tendency for outside contacts to withdraw from the family. This caused distress, anger, social isolation and feelings of being abandoned by wider family, close friends and colleagues. Feelings of isolation from the community, family and friends has also been described by Daly *et al* (2013) in their theory of "sustaining place" (see chapter 2, Table 2.3). This theory indicates how informal carers attempt to maintain citizenship and social connectedness for the PLWD and themselves. Daly *et al* (2013) suggests that this can be achieved by: respecting their family members personhood (and ensuring other people do so); accepting advice, support and services to assist in the caring role and peer support network. However as discussed previously, services and support are often not offered immediately after the diagnosis, and families are left to find this for themselves with varying degrees of success.

In the current study, the Brown, Cooper, Davies and Evans families (F2, F3, F4 & F5) described stable, happy, supportive and loving relationships before the onset of bvFTD. Accordingly, they found changes in their family member's behaviour, personality and lack of empathy particularly hard to come to terms with; a finding paralleled by Massimo *et al* (2013) and Kipps *et al* (2009). In the Allen family (F1), Ann (spouse) and Andrew (PLWD) successfully sustained their place in the local community post diagnosis, but this was not evidenced in the other families (F2, F3, F4 & F5). Ann and Andrew's experience differed because they actively sought out services, support and local community venues

and activities that were accepting of Andrew's behaviours, and supported the strengths and interests he now had.

8.4   Changes to social citizenship

As mentioned previously in chapter 3, Bartlett and O'Connor (2010) identified 6 components of citizenship: growth; social position; purpose; participation; solidarity and freedom from discrimination (see chapter 3, Table 3.2). In chapters 6 and 7, the changes to social citizenship and connectedness have been described and possible interpretations have been suggested. In this section, I will explore in more depth how and why social citizenship changed for the study families using the 6 components of citizenship to structure the discussion.

i). Growth: The hopes, aspirations and plans for the future of people diagnosed with dementia, and their families, start to change from the onset and through the progression of dementia (Lockeridge & Simpson, 2012). This can be a traumatic time for the PLWD and their families, as there is a realization that their previous hopes and aspirations may need to be revised (Evans & Lee, 2014). However, it should be noted that the these changes to a person's growth and development are not solely due to the effects of the condition, but can also be exacerbated by other people's attitudes and behaviour towards the symptomatic person and their family (Henwood & Downs, 2014; Kitwood, 1997). Negative imagery portrayed in the media contributes to the public's viewpoint that dementia is something to be feared, and is a condition with "no cure, no hope and no help" (Litherland & Capstick, 2014). This can lead to a

perception that PLWD are not in a position to have hopes, aspirations or plans for the future.

In the current study, families described how they redefined their hopes, aspirations and plans for the future when they felt relationships within, and external to the family were breaking down (see chapter 6). After the diagnosis of bvFTD, families faced the reality that their family member had a progressive and life limiting condition. This necessitated further revision of the family's hopes, aspirations and thoughts for the future (see chapter 7). In addition, from the family's perspective, their family member living with bvFTD was excluded from employment, activities and social events they previously enjoyed, due to the changes in their behaviour, personality and social functioning, which led to a negative change in their social citizenship.

ii). Social position: People occupy multiple identities and positions in their lives e.g. family member, employee, community member, with varying levels of status attached to each; by others in the family, local community and society in general (Bartlett & O'Connor, 2010). It has been argued that when a person is diagnosed with dementia there can be a downgrading of the person's status (Sabat, 2001). In their theory of "Sustaining place", Daly *et al* (2013) describe how families living with dementia can be isolated from experiences and interactions with other people external to the immediate family, due to the lack of understanding about dementia and their caring responsibilities.

In the current study, when the person living with bvFTD left their employment, not only did they have a reduction in family finances, but also their social network reduced significantly. The previous sense of purpose, and social standing began to reduce for the person with bvFTD. The family found they were not invited to social and community events in the same way as before the onset of the symptoms of bvFTD (see chapters 6 & 7). Family members found this difficult to accept, and felt a sense of betrayal as these friendships and social supports were reduced for all the family, and not just the person living with the diagnosis. As the social positions and networks changed for the person living with bvFTD and their family, social connectedness and social citizenship began to reduce.

iii). Purpose: People living with dementia generally want to have a purpose and meaning in their life, and want to be valued (Bamford & Bruce, 2000). However, when a person is diagnosed with dementia there is a tendency for others associated with them, to focus on what the person has a difficulty doing rather than their strengths and abilities (Litherland & Capstick, 2014). This can unintentionally reduce the PLWD's sense of purpose, confidence and self-esteem.

Families in the current study, spoke about trying to find meaning in their life after the diagnosis was given, but found it difficult to think of what would happen in the future (see chapter 7). With the exception of the Davies family (F4), each of the wives of the person living with bvFTD continued with outside

employment, as they said it gave them a purpose, retained a sense of normality and enabled them to retain a social network. These spouses were living a dual identity, retaining a clear sense of purpose in one part of their life, whilst living with uncertainty in their family life. This could be evidence of the spouse trying to "Move on" in their life and become an "I" rather than a "We". This "Moving on" behaviour was also recognized and described in a study by Hellstrom *et al* (2007). There was an apparent lack of purpose for the PLWD once they left employment, and their role within the family changed. Social connectedness of the person living with bvFTD reduced, within and external to the family. Social citizenship also reduced, as the PLWD and their family, began to be isolated from their local community and social networks.

iv). Participation: In social citizenship, as defined by Bartlett & O'Connor (2010), participation includes active involvement in decision making, with the PLWD's input being valued and respected. Cantley *et al* (2005) and Bartlett & O'Connor (2010) wrote about how PLWD could be actively involved in national and local groups to increase public awareness, influence policy development, plan and develop services and support. In their theory of "Sustaining place", Daly *et al* (2013) indicated that family carers enabled continued citizenship of the PLWD and themselves, by challenging exclusion, abandonment and disconnection from the local community and society.

In chapters 6 & 7 of this thesis, quotes from families indicated how they challenged health and social care professionals to receive an accurate

diagnosis (see chapter 6, Box 6.10) and post diagnostic support (see chapter 7, Box 7.1). These examples do not indicate that the study families were listened to, actively involved in the process of decision-making or future care and support needs. This could indicate that the health and social care professionals did not consider that they needed to actively involve the PLWD and their family in decisions. This may demonstrate that these professionals did not respect or value the opinions of the family, and therefore did not accord them the status of active social citizens.

v). Solidarity: In social citizenship, Bartlett and O'Connor (2010) describe solidarity as when a PLWD unites with others to make a difference and make social connections. In 2014, Bartlett conducted a study into PLWD who campaign for social change, and identified that they felt energized, part of society and saw this as their occupation. However, PLWD also found this work tiring, and worried that they would forget things. In addition, PLWD stated that during their campaigning and awareness raising there were challenges from others in the local and national community, who would question their diagnosis, because they were able to campaign and live positively with dementia, and not conform to the stereotype (Bartlett, 2014; O'Sullivan *et al* 2013).

The spouses of the families in the current study stated they felt very isolated, as they did not know any other families living with the effects of bvFTD. Common questions asked by families during interviews and observations visits were, "Is this what normally happens?" and "Is this what other families say or do?" This is

not surprising given the lack of specialist information, advice and support given to families at the time of diagnosis or in the post diagnostic stage. Family members felt that if they had received timely post diagnostic support, and were linked with other families living with bvFTD, they would have been able to join forces to campaign for improved services and support. This could have increased the family's sense of agency, belonging and citizenship.

vi). Freedom from discrimination: PLWD have been described as the most socially excluded and stigmatized members of society (Alzheimer's Disease International, 2012). PLWD can be discriminated against and have a reduced status in society, resulting in less power and influence (Bartlett & O'Connor, 2010). Woods (2001) and Bartlett & O'Connor (2010) have identified how PLWD can be denied access to services and benefits due to their diagnosis and are perceived as lacking the ability to speak on their own behalf.

In the current study, the person living with bvFTD was excluded or was allowed limited attendance at services, events and activities due to the symptoms of their dementia. Families in this study spoke about how they felt discriminated against, because they had a person in the family who was diagnosed with dementia under 65 years of age. Although each family fought against this stigmatization and discrimination they generally felt powerless to change the systems they encountered.

As discussed, social citizenship changed for the whole family, from the commencement of the symptoms of bvFTD. Citizenship further reduced over time as the symptoms of bvFTD progressed, with families stating they felt isolated from society, and their previous social supports and networks. This further exacerbated the family's sense of loss and abandonment.

8.5   Limitations of the study

Five families living with bvFTD participated in this study. Although this is a small sample size in comparison with some research approaches, the depth of study and the range of data collection tools used, enabled me to collect rich data to make a contribution to the body of knowledge on social connectedness changes in bvFTD. It should be noted that 4 out of the 5 people living with bvFTD were male and their spouses were female, and all were from a white British, middle class professional background. It could be surmised that white, middle class, professional families are more likely to engage with research studies, so are more likely to volunteer for this type of study. This could have had some influence on the findings. Further studies could explore whether social connectedness changes in people living with bvFTD from other sociocultural and socioeconomic groups.

The study was conducted over a short time scale (2013-15), as required by the award of the Doctorate of Professional Practice. The information on the pre morbid and pre diagnostic stages were based on the retrospective memories of family members, and their recollections of these stages. However, retrospective

memories may be inaccurately recalled or influenced by the present thoughts and feelings of the reporter. This study could have been improved by being conducted over a longer timescale to commence immediately after diagnosis and followed through until the end of life.

Although four of the study families (F2, F3, F4 and F5) had children, the children in the Brown (F2), Cooper (F3) and Evans (F5) declined to take part in the research. The children in the Davies family (F4) were under 16yrs of age and undergoing couselling so I chose not to interview them in case they found this traumatic. The lack of interviews of children in these families has meant that children's voices are missing from this analysis and their understandings of social connectedness might yet need to be revealed. If future studies commence at the time of diagnosis, in the early stages of bvFTD, there could be the opportunity to involve children, as it is important to seek their viewpoint of how and why social connectedness changes in their family.

I decided to study people with bvFTD in the middle to late stages of the condition, as so little research had taken place with people in these stages. However, a result of this was that the PLWD were less able to communicate due to verbal expression difficulties, and their general withdrawal from communication. The study could have been enhanced if the PLWD could have expressed their thoughts and feelings regarding social relationships and social connectedness. Another limitation of studying families who were in the middle to late stages of bvFTD was that their social contacts, networks and social

connectedness had already significantly reduced at the time of the study. As a result, there were very few "Identified others" to interview or contribute to the study, and a reduced opportunity for conducting participant observation. This could be improved by conducting a longitudinal study from early diagnosis through to the end of life.

8.6   Implications for practice

The current study families indicated that they thought the following areas needed to be improved for families living with bvFTD:

- Early recognition and diagnosis (see section 8.6.1), these areas were also highlighted in studies by Lockeridge & Simpson (2012) and Svanberg *et al* (2010).

- Specialist post-diagnostic support (see section 8.6.2). Other studies also indicated that post-diagnostic support should be improved for families living with bvFTD (Oyebode *et al*, 2013; Griffin *et al*, 2015; Allen *et al*, 2009; Arai *et al*, 2007; Diehl-Schmid *et al*, 2013 & Denny *et al*, 2012).

- Awareness raising and maintaining citizenship (see section 8.6.3 p211-214), this was also recognised in studies by Johannessen & Moller (2011), Nicolaou *et al* (2010) and Lockeridge & Simpson (2012).

These areas for improvement are addressed in the next sections of this chapter.

8.6.1   Early recognition and diagnosis

As indicated earlier in this chapter (see sections 8.2.1 & 8.2.2), the lack of early recognition and accurate diagnosis of bvFTD can prove to be problematic for families. The changes in behaviour and personality of the symptomatic person, can lead to a gradual reduction in social connectedness and social networks, as others in their family, and social group, misinterpret why the person is behaving in an uncharacteristic way (Lockeridge & Simpson, 2012; Johannessen & Moller, 2011; Daly *et al*, 2013). Without an accurate diagnosis, there is no identifiable reason for the changes in behaviour and therefore, there is a tendency, by others, to consider the behaviour change as intentional (see chapter 6, section 6.3.3). This misinterpretation could lead to loss of employment, a breakdown in relationships, stigma and a reduction in the social connectedness of the person with bvFTD (Lockeridge & Simpson, 2012).

All of the families in the current study spoke about the problems they had getting a diagnosis, and stated that this was due to a lack of awareness and recognition of the signs and symptoms of bvFTD by medical practitioners, and other health and social care staff (see chapter 6). They called for more training for medical practitioners; health and social care staff regarding the early recognition and diagnosis of young onset dementias (and bvFTD in particular). Box 8.1 details a recommendation for improving the early recognition and diagnosis of bvFTD.

Box 8.1: Recommendations for early recognition and diagnosis
1. A programme of training, information and resources should be devised by national groups (e.g. Young Dementia UK, DEEP, Alzheimer's Society and Dementia UK) for medical practitioners, health and social care staff regarding the signs and symptoms of bvFTD.
2. People living with bvFTD and their families should be actively involved in designing and delivering the training programme.

3. A referral pathway should be designed and implemented to aid clarity for medical practitioners on how and who to refer symptomatic people to.

The families in the current study described how the changes in the person diagnosed with bvFTD had a negative effect on the whole family's connectedness, relationships, self-identity and self-esteem (see chapters 6 & 7). By the time the diagnosis was given connectedness and relationships had already broken down and families stated that they could not be rebuilt to their previous level and this caused them sadness and distress (see chapter 7). It could be argued that if the symptoms of bvFTD were recognized and diagnosed earlier, and appropriate post diagnostic support given, the family's social connectedness and citizenship could have been maintained longer so reducing the isolation and distress experienced.

8.6.2 Specialist post-diagnostic support

Chapters 7 (see section 7.2) and 8 (see section 8.3) detailed the difficulties that could arise when a person is diagnosed with bvFTD, and they and their family do not receive post diagnostic support. Families participating in research studies into young onset dementia and bvFTD state that post diagnostic support should be delivered by health and social care staff who have specialist knowledge and skills, e.g. Young Onset Dementia Team or specialist dementia nurse (Admiral Nurse). Families in other studies have indicated that specialist post diagnostic support is needed to enable the family to understand what is happening to their family member, to develop coping strategies (Lockeridge & Simpson, 2012; Nicolaou *et al*, 2010), to retain relationships (Hellstrom *et al*,

2007; LaFontaine & Oyebode, 2014; Lockeridge & Simpson, 2012) and social connectedness (Lockeridge & Simpson, 2012; Harris & Keady, 2009; Johannessen & Moller, 2011).

As detailed in chapter 1 (section 1.3), although there has been a focus on developing community awareness, research and policies regarding dementia, the specialist needs of younger people living with dementia, and their families, have not been adequately addressed. In addition, of the 18 main aspirations of the Prime Minister's Challenge on Dementia 2020 (see chapter 1, Box 1.4), only 2 are focused on post diagnostic care and support. These aspirations focus on delivery of services and support for the majority of people with dementia, i.e. older people, and as detailed earlier their needs can be very different to younger families living with dementia (see chapter 1, Box 1.5). Future policy development regarding dementia should include the need for specialist post diagnostic support for younger families, including bvFTD.

As detailed in chapter 2, the changes in social behaviour of the people diagnosed with bvFTD can lead to deterioration in relationships with family and others (Mendez *et al*, 2014), and a reduction in the social networks, and connectedness of the person with the diagnosis of dementia and their family, and can result in feelings of increased stress and burden (Mioshi *et al*, 2009; 2013 & Nicolaou *et al*, 2010). Studies by Johannessen & Moller (2011) and Daly *et al* (2013) indicated that social support and connectedness changed for families living with young onset dementia throughout the progression of the condition. The lack of recognition of the early signs of dementia both by the

families and the medical practitioners added to the distress of families, and resulted in a significant delay in receiving specialist support and advice.

Families in the current study felt that earlier specialist post diagnostic support could have enabled them to cope with the changes experienced and to develop coping strategies. Spouses of the person diagnosed with bvFTD indicated that home based services and respite, could enable them to continue with employment, spend quality time with their children and take part in interests external to the home. Families in the study had experienced situations where there was no respite or services available to meet the identified needs of their family member diagnosed with bvFTD.

Peer group support was requested by the families in the study, they all said they would like to meet with and talk to other people who are experiencing the same issues as they are. They spoke about the isolation experienced and the feeling of being "different" from their peer group. Each family in the study stated that the carers groups they attended, were focused on the issues faced by older people who had different forms of dementia and as they did not see the relevance of them in their situation they had stopped attending. This intensified their feelings of being different and isolated from their local community. For recommendations to improve post diagnostic support see Box 8.2 below.

Box 8.2: Recommendations for specialist post diagnostic support
1. Families living with bvFTD should be offered specialist post diagnostic support immediately after diagnosis.   2. People living with bvFTD and their families should be actively involved in designing and delivering information and support sessions for families, alongside

specialist practitioners.

3. Activities and groups should be designed around the needs of the person living with bvFTD and their family. The activities should be focused on maintaining and developing social connectedness and citizenship.

4. Specialist respite services should be developed that can deliver support in the person's own home or in a specialist age appropriate setting.

However, as previously mentioned families often do not recognise the early symptoms of bvFTD, and tend to present at a medical practitioners when they are in a crisis situation. This leads to a delay in diagnosis and in receiving post diagnostic support. The nationwide dementia awareness sessions could include information on the recognition of symptoms that may indicate the possibility of bvFTD, which could enable families and others in their social network to recognise the condition much earlier.

8.6.3 Awareness raising and maintaining citizenship

The national growth of Dementia Friendly Communities and Dementia Friends initiatives could help to address the lack of understanding about the needs and requirements of PLWD, by building community support and upholding citizenship (Henwood & Downs, 2014). The National Dementia Declaration (2010) (see chapter 1, Box 1.1) was created to stimulate a change in how society responds to dementia. If these "I statements" are followed, PLWD and

their families could maintain their citizenship and have the appropriate support to enable them to live as well as possible with dementia.

All of the family members in the current study stated that the lack of awareness of bvFTD within health and social care professionals, family members, social contacts and local communities, led to a lack of understanding, absence of support and a growing sense of social isolation of the whole family. They thought it essential that awareness campaigns should also include information on frontotemporal dementias, and the different signs and symptoms that can be exhibited. This could increase the recognition and awareness of the condition and hopefully decrease the stigma and discrimination families' experience. For recommendations to improve awareness of young onset dementia including frontotemporal dementias see Box 8.3.

Box 8.3: Recommendations for increasing awareness of young onset dementia
1. People living with young onset dementia and their families should be actively involved in designing and delivering information for dementia awareness sessions. This could be incorporated in the Dementia Friends & Dementia Friendly Communities training. 2. The Dementia Friendly Business training could incorporate information on how to recognise the possible signs of young onset dementia and what to do if they suspect this. 3. The Dementia Friendly Business training could also include how to support someone living with young onset dementia in the workplace. In addition, it could also include how to support a family member of someone living with young onset dementia.

In the current study, the person with bvFTD had already left their employment. However, families stated that they thought it was important that employers should be aware of the possible early signs and symptoms of young onset dementia and in particular bvFTD, as the symptoms may initially be more apparent in the workplace than at home. Had the families in the current study realised that their family member was having difficulties at work they would have urged them to seek help.

The National Dementia Strategy (2006) is currently under review and a new guideline is being developed. It is important that the needs of younger people living with dementia are recognised within the new guideline and Alzheimer's Society, Dementia UK and Young Dementia UK, among others, have been inputting the findings from research and from families living with young onset dementia into this. In addition, commissioning guidance for dementia is being developed which should be completed and actioned, in the latter part of 2016. Younger people with dementia, their families and specialist practitioners are actively participating in the formulation of the commissioning guidance. The key issue is to ensure all guidance regarding dementia, and specifically young onset dementia becomes a reality throughout the UK, by negotiating with, campaigning, and if necessary challenging, commissioners and service providers.

In summary, the lack of awareness, recognition and early diagnosis of bvFTD leads to significant feelings of distress for affected families. It can lead to a delay in diagnosis, lack of advice and support after it is given. Current practice

could be improved by working with health and social care staff to build an awareness of the signs and symptoms of young onset dementia (including bvFTD). Awareness raising could lead to early recognition of young onset dementia, and an appropriate referral for an assessment and accurate diagnosis, at an earlier stage of the condition. Once the person is diagnosed with bvFTD, there is a need for timely post diagnostic support to help PLWD and their families, to explore and manage their feelings and thoughts, and help them to develop coping strategies. This could enable the family to maintain relationships and social connectedness. In addition, referral to peer support groups could help families to feel connected to others in a similar situation and reduce the feelings of isolation. In addition, the peer group could give advice and support regarding some of the behaviours exhibited.

8.7 Recommendations for future studies

There is a paucity of research into the experience of living with bvFTD, and its effects on social connectedness and relationships. In searching the literature and conducting the study, other potential areas of research were identified that need to be addressed in the future. This is essential to not only assist in the development of a body of knowledge, but also to challenge and change current practice. This will ensure that families living with bvFTD receive a timely diagnosis, followed by specialist post diagnostic advice, support and interventions to meet their current and developing needs. The areas identified for future research are detailed overleaf in Table 8.1.

Table 8.1: Areas identified for future research.	
Topic	**Rationale**

i). A longitudinal study of families living with bvFTD to explore how and why social connectedness changes over the progression of the condition.	My research into social connectedness and bvFTD involved participation of 5 families and identified others during 2014-15. In order to study this topic in more depth over a longer period of time, families could be recruited from the time of diagnosis and followed through until end of life. This would enable the researcher to collect longitudinal data regarding social connectedness through the stages of bvFTD, and explore how and why connectedness changes. This could lead to the development of interventions and support that could help to prevent the reduction in social connectedness.
ii). Research studies on various interventions that can be used when working with families living with bvFTD to reduce family distress and social isolation.	A range of interventions and supports could be evaluated to establish which ones are effective in meeting the needs of families living with bvFTD. Interventions and supports could include: peer group support; family therapy; relationship-centred care; counseling approaches; different forms of respite; social groups and activity based interventions. Families living with bvFTD could be actively engaged in designing and providing evaluations of the interventions implemented.
iii). Research into how people living with bvFTD could be supported in the workplace.	Ideally the person living with bvFTD would still be in employment, but as described earlier in the thesis, frequently the person has already ceased employment before diagnosis. The alternative would be to work with a selected employer as a pilot study with people diagnosed with bvFTD and evaluate how the employee could best be supported in the workplace.
iv). Research into the early recognition and referral of people suspected of having bvFTD.	Research examining what happens in the pre diagnostic pathway of people diagnosed with bvFTD. This could be a retrospective study to explore the missed opportunities and problems of recognizing the signs and symptoms in primary care (e.g. General Practitioners). The families living with bvFTD could be actively involved in the design of the study. The gathered information could then be utilized to design a training programme, with active involvement of the families living with bvFTD to improve the recognition of bvFTD in primary care and assist in a timely referral and diagnosis
v). Studies in identified areas of the country piloting an awareness campaign of bvFTD.	The pilots could follow a similar approach as the Dementia Friends and Dementia Friendly Community programme but focus on bvFTD, and the very different presentation as compared with Alzheimer's disease. Families living with bvFTD could be actively involved in the design and delivery of this. These pilots could then be evaluated, and if effective, repeated in other areas of the UK.
vi). Studies which explore the experience of people diagnosed with bvFTD and their families from different socioeconomic and cultural groups.	The current study families were from the same socioeconomic (middle class) and cultural group (white British) and so further studies could explore whether gender, socioeconomic and cultural group differences influence the experience of families as regards social connectedness.

In summary, bvFTD is a rarer diagnosis affecting younger people and as a result there is a paucity of research into its effects on the person with the diagnosis, family and identified others. This could be a contributory factor as to why there is a limited awareness of bvFTD, and a consequent delay in recognition and diagnosis.

8.8   Conclusion

In the current study I have addressed the following main research question:

> *"How and why does social connectedness change in families living with the effects of bvFTD?"*

The sub questions derived from this were:

> 1. *"How and why does social connectedness change for people diagnosed with bvFTD?"*
> 2. *"How and why does social connectedness change for family members of a person living with a diagnosis of bvFTD?"*
> 3. *"How and why does social connectedness change for identified others (e.g. work colleagues, siblings, faith group, neighbours, parents) who have a social relationship with a person living with bvFTD?"*

Each of these sub questions will be addressed in this section of the thesis. Following this, the main research question will be answered.

1.   *"How and why does social connectedness change for people diagnosed with bvFTD?"*

Chapters 5, 6 & 7 detailed how social connectedness changed for people diagnosed with bvFTD from the pre morbid stage (chapter 5), through the pre diagnosis stage (chapter 6) and after diagnosis (chapter 7). A key feature of bvFTD is how insight into the changes in behaviour, personality and social functioning, is often lost early in the condition (see chapter 1). This lack of insight can result in the symptomatic person denying there is a problem and therefore not willing to seek help or support. This denial can cause distress for the family as they try to make sense of what is happening to their loved one. It can also lead to a prolonged time before a symptomatic person will agree to attend an appointment with a medical practitioner to investigate the cause of the changes (see chapter 6).

Throughout the pre diagnostic, diagnostic and post diagnostic stages, PLWD in the current study experienced a reduction in their social network, a deterioration in the quantity and quality of relationships with other people, both within and external to the family, due to the changes in their behaviour and social functioning. In addition, it was noted by the family that the PLWD started to withdraw from activities, events and hobbies that previously interested them, which could have been due to the symptoms of bvFTD (see chapter 1, section 1.4) or the response of others to the changes.

2. "How and why does social connectedness change for family   members of a person living with a diagnosis of bvFTD?"

At the commencement of the first signs of bvFTD (pre diagnostic stage) the family attributed the changes in their family member to issues such as depression, stress or loss of employment (see chapter 6). At the beginning, when they thought that the changes were due to circumstances beyond the control of their family member, they were supportive, made adaptations and allowances. As the changes progressed the families began to think the changes were intentional, or evidence of relationship breakdown. This damaged their relationships and affected their connectedness as a family (see chapter 6). At the same time, the study families reported that their friends, colleagues, local community and extended family found the socially disinhibited behaviour of the PLWD embarrassing and gradually stopped inviting the whole family to events and social activities. In addition, the family started to avoid social situations due to the embarrassment caused by their family member with bvFTD. Although these changes to social networks, relationships and connectedness commenced in the pre diagnostic stage they continued to reduce during the time after the diagnosis (see chapters 6 & 7).

3. "How and why does social connectedness change for identified others (e.g. work colleagues, siblings, faith group, neighbours, parents) who have a social relationship with a person living with bvFTD?"

The social network and friendship group had already significantly reduced for the families involved in this research study, so there was a limited opportunity of discussing their perspectives of how and why social connectedness had reduced over the progression of bvFTD. The friends who were still in contact and who agreed to be interviewed, stated that in their opinion the relationships

had broken down due to the changes in the person with bvFTD before they were formally diagnosed. Once broken, these social connections could not be rebuilt once the diagnosis was given (see chapter 7). This could in part be due to the social network finding it difficult to reengage after the absence, or not knowing how to communicate with the PLWD.

Main research question: *"How and why does social connectedness change in families living with the effects of bvFTD?"*

Social connectedness is important for a person's sense of belongingness, attachment, self-identity and self-esteem. In the case study families, social connectedness had significantly reduced during the progression of bvFTD, which had an impact on their relationships and quality of life (see chapters 6 & 7). The current studies revealed that the families experienced stigmatisation and isolation, due to the lack of awareness, recognition and understanding of bvFTD, by the local community, health and social care professionals, family and friends.

There has been an increased focus on improving the general public, community and business awareness of dementia over the last few years, which has started to reduce the stigma and fear of dementia as a diagnosis. However, the focus has been on the older age group and the more common diagnosis of Alzheimer's disease, which is characterized by a reduction in cognitive and functional abilities. In the early stages of bvFTD, the person is less likely to exhibit memory, orientation and functional impairment, so a diagnosis of

dementia may be overlooked leading to a significant delay in presentation and referral for a diagnosis. The awareness campaigns should include information on more unusual presentations of dementia. Families in the current study did not have an awareness of the signs and symptoms of bvFTD, and hence did not go to a medical practitioner when their family member was in the early stages of bvFTD. If the countrywide awareness campaign was modified to include information on the early signs and symptoms of bvFTD, more families in the UK could benefit. It could also lead to earlier recognition of the signs of bvFTD whilst relationships, employment and social connectedness are still relatively intact.

If bvFTD was recognized, diagnosed and specialist advice and support given earlier by medical practitioners, it could enable families to understand what is happening to their family member and develop coping strategies, build family resilience and prevent crises. This could enable the families to live as well as possible with dementia, with appropriate support.

The findings from the current study, suggests medical practitioners, health and social care professionals, should receive training regarding the recognition of the signs and symptoms of young onset dementia and bvFTD. The health professionals should also receive advice on how to refer people with possible young onset dementia and bvFTD, for specialist assessment, diagnosis, advice and support. Families stated that they would have benefited from specialist support and advice post diagnosis, as they believed it would have helped to

prevent relationship breakdown and social connectedness difficulties. Peer group support should be available for all families who have a family member diagnosed with bvFTD, to reduce the feelings of isolation.

The current research was different to other studies, as it explored the experience and perceptions of family members living with bvFTD (and identified others) concerning how and why social connectedness changed from the pre-diagnostic to after diagnosis stages. Each of the people living with bvFTD in the study was in the middle to late stages of the condition, whereas the reviewed literature into the lived experience of bvFTD tends to be conducted in the early to middle stages. Although only 5 families living with bvFTD participated in this study, the results could add to the body of knowledge and stimulate further research. This study also suggests possible changes to current practice that could improve the experience of families living with bvFTD and areas for future research. When the case study families were asked what final message would they like to give to health and social care staff, they all said: **"Listen to the families!"**

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Appendix 1: Literature Search Template

Identification of Search Themes			
Term	**Alternative Terms (in abstract or subject)**	**Databases searched**	**Results**
"Social Connect*" **AND** **"frontotemporal dementia"**	"Social contact*" "Social network*" "social relation*" "behavioural variant frontotemporal dementia" "Frontal* dementia" "Young* onset dementia" "Young* person with dementia" "Young* people with dementia" Pick's "working age dementia" "Presenile dementia"	EBSCO: - CINAHL - ebook collection - Medline NELSON (gateway) APA PsycINFO Ovid Online Proquest SAGE Journals ScienceDirect Wiley Online Library	**EBSCO:** "Social connect*" AND "behavioural variant dementia"=**0** "Social connect*" AND dementia= 6,830 (further refined by searching frontotemporal dementia) AND "frontotemporal dementia"=160 (10 duplicates and 142 not relevant for my research)= **8 relevant for my literature review.** "Social network*" AND "frontotemporal dementia"=228 (5 duplicates and 215 not relevant for my research)= 8 relevant for my research but all counted in NELSON articles or social connect*= **0** "social network*" AND dementia= 120 (4 duplicates and 104 not relevant for my literature review)= **12 relevant for my literature review** **NELSON (gateway- University of Northampton):** "Social connect*" AND dementia=6 (none relevant for my research)= **0** "Social contact*" AND dementia= 9 (1 duplicate and 6 not relevant for my research)= **2 relevant for my literature review** "Social network*" AND dementia=81 (7 duplicates and 72 not relevant for my research)= **3 relevant for my literature review**

| | | | | **APA PsycINFO:**
"Social Connectedness"
AND dementia=**0**

Ovid Online:
"Social connect*" AND "behavioural variant dementia"=1 (not relevant for my research)= **0**
"Social connect*" AND "frontotemporal dementia"=12 (none relevant for my research)= **0**
"social connect*" AND "young* onset dementia"=4 (3 not relevant for my research)= 1 but already counted in NELSON search= **0**
"social connect*" AND "young* person with dementia"=2 (2 duplicates)= **0**
"social connect*" AND "Pick's"=12 (11 not relevant for my research)=**0**
"Social contact*" AND "frontotemporal dementia"=30 (2 duplicates and 28 not relevant for my research)= **0**
"social network*" AND "behavioural variant frontotemporal dementia"=2 (not relevant for my research)=**0**
"social network*" AND "young* people with dementia"=6 (4 not relevant for my research)= **2 relevant for my literature search**
"social network*" AND "young* person with dementia"=3 (3 duplicates)=**0**
"social network*" AND "working age dementia"=1 (1 duplicate)=**0**

Proquest:
"Social connect*" AND dementia=4 (4 not relevant for my |

			research)= **0**
			"Social contact*" AND dementia= 19 (19 articles not relevant for my research)= **0**
			"Social network*" AND dementia=38 (38 not relevant for my research)= **0**
			Sage Journals:
			"Social connect*" AND dementia=10 (8 not relevant for my research)= **2 relevant for my literature review**
			"Social contact*" AND dementia= 12 (12 articles not relevant for my research)= **0**
			"Social network*" AND dementia=34 (32 not relevant for my research)= **2 relevant for my literature review**
			Science Direct:
			"Social connect*" AND dementia=4 (4 not relevant for my research)=**0**
			"Social contact*" AND dementia= 13 (13 articles not relevant for my research)=**0**
			"Social network*" AND dementia=25 (24 not relevant for my research)=**0**
			Wiley Online Library:
			"Social connect*" AND dementia=11 (10 not relevant for my research)= **1 relevant for my literature review**
			"Social contact*" AND dementia= 28 (28 articles not relevant for my research)=**0**
			"Social network*" AND dementia=27 (26 not relevant for my research)=**1 relevant for my literature review**

			33 identified on 1st search (23 articles excluded on second examination as did not meet the set criteria) **Total number of articles/studies that met the inclusion criteria= 10**

Appendix 2: Case Study Protocol

	Outline
1. Background	
Topic	Exploration of the impact of bvFTD on social connectedness
Research problem statement	There is very little research into the experience of families living with the effects of bvFTD and how and why this effects their social connectedness. What is in evidence within the literature, from clinical experience and within carers groups is that the social relationships, contacts, networks and connectedness of not just the person living with a diagnosis of bvFTD but also the family, significantly reduces.
Theoretical framework	• Transformative paradigm • Social Citizenship:- conceptual framework • Case Study research
2. Design	
Design	Qualitative research Case study: Multiple case and Holistic design. Exploratory- as it helps to gain insight into the structure of the phenomenon in order to develop hypotheses, models, theories Replication logic of multiple case study design. Literal replication- predicting similar results.
Describe who will be studied and how they are selected	Purposive sampling. Family of a person diagnosed as having bvFTD.

	Person who has been diagnosed as having bvFTD Identified others who have social relationships with the person living with bvFTD. These categories have been chosen in order to explore the impact of bvFTD on social contacts, networks and connectedness from the perspective of the person living with bvFTD, their family and any other people who have social relationships to provide the breadth and depth of understanding of the how and why social connectedness changes.
Purpose statement	The purpose of this multiple case study research is to understand how and why social connectedness changes in families living with the effects of bvFTD.
Central question	"How and why does social connectedness change in families living with the effects of bvFTD?"
Sub questions derived from research question and the measures	The sub questions derived from this are: 1. "How and why does social connectedness change for people diagnosed with bvFTD?" 2. "How and why does social connectedness change for family members of a person with a diagnosis of bvFTD?" 3. "How and why does social connectedness change for identified others (e.g. work colleagues; siblings; faith group; neighbours; parents) who have a social relationship with a person living with bvFTD?" This will be explored by the use of: semi structured interviews; participant observation; documents and diaries.

3. Case selection	
Ethics	Participants will be fully informed of the purpose of the research before they consent to take part. Will be given an information letter, participation information sheet, and consent sheets written in terms that can be easily understood. Participants can withdraw at any time without any implications and without being required to provide an explanation. Information gathered during the research will be kept confidential at all times and will be suitably anonymised. Handling and storage of data will follow the guidelines set by the Data Protection Act (1998) and the Freedom of Information Act (2000).
Criteria for case selection.	The principle inclusion criteria will be families living with the effects of bvFTD. The principle exclusion criteria will be families living with the effects of any other form of dementia.

4. Case study procedures and roles	
Field procedures	All participants will be given a choice of setting for the interview and observation study. Participants may chose to be interviewed in their home setting. In the observation these can be conducted in various settings including: care home; groups; social situations; café; activities. All participants will be given information about the research to enable them to give informed consent.
5. Data Collection	
Data to be collection	Semi structured interviews Participant observation Diaries Documents
Define a data collection plan	Northamptonshire for pilot study- September 2013 Approach FTDSG to send out call for participants for the case study- November 2013 FTDSG London- I will present my research plans and recruit participants for my research- March 2014 Visits, observation and interviews to be conducted in the families' choice of environment from January 2014 to April 2015. Transcription of interviews, observation visits completed as soon as possible after the event.

Define how the data will be stored	Handling and storage of data will follow the guidelines set by the Data Protection Act (1998) and the Freedom of Information Act (2000). The data will be stored in a locked filing cabinet for which only I have the key.
6. Analysis	

The criteria for interpreting case study findings	Thematic analysis will be used to analyse the data collected, including the diaries and documents, as this approach will assist me to identify patterns and themes in the collected data (Braun & Clarke 2006).
	
	The six phases of analysis will be followed for interpreting data (Braun and Clarke (2006):
	    • phase 1: familiarising with data
	    • phase 2: generating initial codes
	    • phase 3: searching themes
	    • phase 4: reviewing themes
	    • phase 5: defining and naming themes
	    • phase 6: producing the report
	
	After the themes have been identified in the individual case studies (within case analysis) they will be compared and contrasted with what is known from other research, publications or policy. Also the results of each case studied will be compared and contrasted to identify similarities and differences (cross case analysis). After this is completed consideration will be made as to how transferable the results will be from one context to another.

Range of possible outcomes and edification of alternative explanations and identification of any information that is needed to distinguish between them	Possible outcomes of the study: • interventions can be suggested to improve social connectedness for family and person living with bvFTD • no interventions can be suggested to improve social connectedness for the family and person living with bvFTD • there maybe decreased social connectedness for family and person living with bvFTD due to: i). family disengaging from their social network ii). Person living with bvFTD disengaging from their social network iii). Identified other(s) disengaging from the family and person living with bvFTD iv). Community in general disengages from the family and person living with bvFTD Support or rebuttal of these possible outcomes would come from the data collected in the interviews, documents, observations and diaries.
7. Plan validity	
Multicultural validity	I will use: • pattern matching, explanation building and theory to back explanations in the data analysis stage. • multiple sources of evidence e.g. interviews, observation, diaries, documents and once transcribed they will be given to the participants to review and make comments.
Reliability	Reliability, credibility and dependability in the research will be demonstrated by devising and following this case study protocol to ensure consistency across each case study family and using the same data collection procedures in each case with the same procedures.
Triangulation	For this research data triangulation will be achieved by using a variety of data sources (families living with the effects of bvFTD and identified others). I will also use a variety of data collection methods (methodological triangulation) including semi-

	structured interviews, observation, documents and diaries. Theory triangulation will be achieved by checking transcripts, data, findings and themes with participants throughout the research and comparing the findings with other researchers in bvFTD.
8. Study limitations	
	This is a research study prepared for submission of a Doctorate of Professional Practice with limitations on time, word limit and budget. I will conduct all of the research processes, procedures and analysis throughout the study which will be very labour intensive but have the advantage of consistency. Potential for observer bias. In order to prevent this, I will engage in reflexivity and also share the case study transcriptions, findings, themes and overall assertions with the individual participants for their views. Lack of generalization to other situations and therefore it does not add to scientific development. Five families living with the effects of bvFTD will participate in this study and although this is a small sample size in comparison with some research approaches the depth of study and the range of data collection tools will enable me to collect rich data to make a contribution to the body of knowledge on the how and why social connectedness changes in bvFTD.
9. Reporting	
	Conferences. Publication in professional journals. Presentations to professional groups and general public. National Forums for dissemination: Dementia UK; Alzheimer's Society; FTDSG.

10. Schedule	
	Northants for pilot study- September 2013
	Approach FTDSG to send out call for participants in the case study- November 2013
	FTDSG London to present my research plans and to recruit participants for my research- March 2014
	Visits, observation and interviews to be conducted in the families' choice of environment from January 2014 to April 2015.
	Transcription of interviews, observation visits completed as soon as possible after the event.
	Write up of Chapters of the research: August 2014- 31st July 2015.

Semi-Structured Interview

Prompt sheet

Introduction to the Interview Process:

- Introduce self and research.
- Check consent.
- Explain why the interview is important.
- Ensure confidentiality and explain how I will protect privacy.
- Ask permission to record interview by recording and taking notes.
- Ensure the environment is conducive to interview e.g private, limited distractions.
- Explain visit after the interview to check accuracy and seek clarification if needed.
- Put interviewees at ease with culturally relevant "polite conversation".

The plan would be to encourage the participant to tell their story with the minimum of interruptions and as little prompting as possible.

Part 1: Recognition of change:

Possible prompt questions:

"When did you first notice changes?"

"What were these changes?"

"What did you think?"

"What did you do?"

"How did your partner get the diagnosis?

Part 2: Experience of behavioural variant frontotemporal dementia (Pick's Disease)

Possible prompt questions:

"Did you discuss the diagnosis with anyone? If so, what was their response?"

"Can you tell me about your experience of Pick's Disease?"

"What type of care do you provide?"

"What is a typical day like?"

"How have things changed since getting the diagnosis?"

"What are the main differences between a good day and a bad day?"

"How do you feel on a bad day?"

"What things cause you the most distress?"

"Can you tell me about things that help?"

"Can you tell me about things that don't help".

Part 3: Social networks and support

Possible prompt questions:

"Has your social life changed since the diagnosis?"

"What sort of social activities are you currently involved in?"

"Have these changed since the diagnosis?"

"Have your social networks or relationships changed since the diagnosis?"

"How have family and friends reacted since the diagnosis?"

"Why do you think these changes have occurred?"

"How do you feel about these changes?"

"What is your experience of support from organizations such as health & social care?"

"Can you tell me what support would help?"

Part 4: Finally

Thank you for your time.

"Do you have any questions that you would like to ask of me?"

At the End of the Interview:

- Explain what happens next.
- Ask the person are they still giving consent to the information collected to be used in the research.
- Arrange the follow up visit to check the transcript from the interview and set up an observation visit.

Appendix 4: Guidance Sheet for Diaries

Keeping a Diary

As part of this research participants are being asked if as well as being interviewed they would also keep a diary, for at least one month, of their experience of living with someone with behavioural variant frontotemporal dementia (Pick's Disease).

Diaries can be very useful to record your experiences, feelings, thoughts and behaviours.

What kind of diary should I use?

The diary can be:

- written by hand
- on a computer
- a video or audio diary
- or any other method that you prefer.

You can decide the frequency of diary writing, some people prefer to write daily and others only if something particularly noteworthy happens. Most people find writing about an event soon after it has occurred helps with their accurate recall of the event.

What should I put in a diary?

For this research the diary could be used for recording your:

- experiences
- feelings
- thoughts
- behaviours.

The diary can also be used to write about things that you find particularly pleasurable or difficult.

This research would also like to identify approaches and interventions that may help you to cope with the impact of Pick's Disease. In view of this could you write about any approaches or interventions that you find do or don't work.

A template (in paper or electronic form) has been devised to give suggestions about how to structure your diary entries which you can choose to use if you find it helpful (see page 3).

Confidentiality

You should be as honest as possible and include anything you feel is relevant to you. Anything that is written will be kept in strictest confidence and will be stored in secure locked cabinets. All information will be anonymised so no connection could be made to the source.

What to do if you need help with the diary

Please do not hesitate to contact me if you have any questions or concerns about keeping your diary.

You can contact Hilda Hayo (Researcher) as follows:

ADDRESS

EMAIL

PHONE NUMBER

An example of a diary template

Date & time	Who was present?	What happened?	What did you feel, think and do?	What helped? What didn't help?
11.11.12 5pm	My husband	He started to insist he wanted to go for a walk even though it was pouring with rain and was getting dark. I tried to talk him out of it but he kept getting angry, started shouting and eventually pushed me out of the way to try to get out of the door.	He has never done this before and so I was shocked. He didn't hurt me physically but I felt so shocked and hurt by his behaviour because he was always such a placid and kind husband before he was diagnosed with Pick's.	It didn't help when I stood in front of him at the door as it seemed to make him more angry and determined to leave.
12.11.12 2.30pm	My best friend and my husband.	When my friend arrived my husband told her she looked awful and her clothes were too tight!! She was shocked initially but then laughed it off.	I was mortified and didn't know what to say, luckily I have known my best friend all my life and she has been so very supportive. When she joked and laughed it off it really helped to lighten the mood.	My best friend's approach really worked as after she made it into a joke my husband joined in and what could have been a tense situation was quickly turned into a laugh and joke.
13.11.12 8am	My husband	He looked at me today and said "I really love you."	It was a really lovely thing for him to say, as he hasn't said it for many months. However it gave me mixed emotions: sorrow for what we have lost; happiness that he said this to me and hopes that things might be getting better.	Having a cuddle helped us both.

Appendix 5: Participant Observations – Template

Preparing for Participant Observation:

- Arrange the meeting and venue as agreed with participant including date, time and duration.

- Agree how the researcher will explain their purpose.

- Agree how the observation will be recorded.

- Assurance re: confidentiality and involvement.

Participants (using codes for names):
Venue:
Date:
Start:
End:
Who and how many people are present in setting? **What are their characteristics and roles?**   **What is happening in setting?**

Where are people located in the physical space?

Where do interactions take place?

When do conversations and interactions take place?

What are the actions and interpersonal communication?

What are the variations in the interpersonal communication observed?

Why are there variations in interpersonal communication?

Why do people in the setting act the way they do?

After the Observation:

- Remind participants about consent and confidentiality.
- Writing up notes and share contents for verification and comments.

Appendix 6: Research request letter

ADDRESS

EMAIL

PHONE NUMBER

Date

Dear…………,

Research Request Letter

I am currently undertaking a small scale research study as part of a Doctorate of Professional Practice. The research consists of an interview and observation visit(s) which should last no longer than 1 hour for each visit. In addition you will be asked whether you would keep a diary for the duration of the research.

The full title of the research is:

"How and why does social connectedness change in families living with the effects of bvFTD?"

Before you agree to the interview I would like to confirm that:

- University of Northampton has given approval for this research to be conducted.

- The research has been given Ethical Approval by the University Research Ethics Committee and the NRES Committee East Midlands- Nottingham 1 Research Ethics Committee.

- Participant Information Sheets (one for family members and one for the person living with the diagnosis) are included to explain the research in more depth so

you can make an informed decision about whether you would like to take part in the research.

- A sheet with the focus of the interview is attached so you know what to expect.

- With your permission the interview will be recorded.

- A transcript of the interview will be sent to you after the interview and I will arrange to meet you to go through what is written.

- Your anonymity will be maintained and no comments will be ascribed to you by name in any written document or verbal presentation. Nor will any data be used from the interview that might identify you to a third party.

- You are free to withdraw from the research at any time and/or request that your transcript not be used.

- I will arrange to meet with you on completion of the research and a copy of my final research report will be made available to you upon request.

If you would like to be involved in this research I can be contacted by:

- phone: -----
- letter: -----
- email: --------

If you have any queries concerning the nature of the research or are unclear about the extent of your involvement in it please contact me as above.

Finally, can I thank you for taking the time to consider my request and I look forward to your reply.

Yours sincerely,

Hilda Hayo

Student on the Doctorate of Professional Practice at University of Northampton

Appendix 7: Participant Information Leaflet

Research Title:
The full title of the research is:

"How and why does social connectedness change in families living with the effects of bvFTD?".

Researcher:
Hilda Hayo – Research Student on the Doctorate of Professional Practice
Qualifications: RMN, RGN, PG (Dip), M.Ed (Man.), BA (Hons), BSc (Hons), RNT, Dip.Counselling

Email address: ---------. Telephone number: -----

Introduction:
I have been working with people under the age of 65yrs with dementia for the last 12 years. I set up the Younger People with Dementia Team for Northamptonshire and during this time I have been responsible for: the assessment; diagnosis; treatment; advice giving and provision of support to people who are suspected to have or who have dementia.

As part of my Doctorate of Professional Practice I decided that I would like to explore how living with behavioural variant frontotemporal dementia affects social relationships.

Invitation to take part:
You are being invited to take part in my research study. Before you decide to take part, it is important that you understand why this research is being done and what it will involve. Please take time to read the following information. If you require further information or are unclear about any aspect related to this study please feel free to speak direct to Hilda Hayo

Why is the study being carried out?
This research is being carried out as part of a Doctorate of Professional Practice that the researcher is undertaking at the University of Northampton.

Research questions: The objectives of the research are to explore:

- "How and why does social connectedness change for people diagnosed with bvFTD?"

- "How and why does social connectedness change for family members of a person with a diagnosis of bvFTD?"

- "How and why does social connectedness change for identified others (e.g. work colleagues; siblings; faith group; neighbours; parents) who have a social relationship with a person living with bvFTD?"

Why have I been invited to participate?

You have been asked to participate as you have either been diagnosed as having frontotemporal dementia or are a family member. To date there have been no research studies on the social impacts of living with this diagnosis and this study plans to address this and also to identify interventions and approaches that could help people to maintain relationships and social connections.

Do I have to take part?

No, you are *invited* to take part in the research. After reading this information, you will be offered the opportunity to ask questions, I will explain anything which you do not understand. Only when you feel happy to proceed will you be asked to sign a form giving your consent to take part in this study. If at any time you wish to terminate your participation you can do so.

What will I be asked to do if I agree to take part?

This research will involve being interviewed for up to 1 hour by myself whilst you are being interviewed I would like to record this on a digital recorder for accuracy of reporting, but in order to use such equipment, I must be certain that you are happy to have your experiences recorded in this way. Your recording will be anonymised (no name will be recorded). Notes will be made of your interview and I will share these with you after the interview to ensure you are happy to proceed with the use of these in the research study. (These, as well as the tape recordings, will also be destroyed on completion of the study).

In the research interview I will ask you if you would agree to draw a diagram of who you are socially connected to. The aim of this is to understand your social world from your and other's perspective. The people identified by you may include your family, friends, work colleagues, neighbour, faith group, social and sporting activities. Once this has been completed you can make a decision about who you would like to approach to ask if they would like to be involved in the research. I will give you Research Request Letters, Participant Information Sheets and Consent Sheets for those people who you would like to involve so they can consider whether they would like to take part in the research.

If you agree I would also like to observe your interactions with other people who have agreed to be involved in the research. This would involve me attending social situations with you both to understand your social world from your and other's perspective. If you both agree that I can observe the social situation

then agreements will be made as to: what social situation will be observed; when this will take place; where; for how long and how frequently.

If you would feel more comfortable having another family member or an advocate present during your involvement in the research this can be arranged.

At any time in the observation either party can request that the observation session is stopped and the notes destroyed. I will take notes of the social situation and will then go away and write these up for the research. These notes will be shared with yourself and the other people involved in the social situation as soon after the event as is possible.

Are there any risks?

There are no physical risks to you as a person, however you may find as a result of the interview (and the recording) or the observation part of the research you feel uncomfortable to continue. You will not be expected to discuss anything with me, which you do not feel comfortable with. You are free to withdraw from the study at any time and you wouldn't be expected to justify your withdrawal from the study.

Will the information collected about me be kept confidential?

All information collected from you during the course of this research will be entirely anonymous and will be kept strictly confidential. It will be stored in a secure place, and will be protected by a password if saved on a computer.

In research on rare occasions the researcher may identify situations or information which raises concern for the safety of yourself or others. In these cases the researcher is ethically and legally obliged to share this information with relevant parties. If this does become necessary in the course of this research I will ensure that I discuss this with you and explain what is to happen and why before this information is shared.

What should I do if I want to take part?

If you want to take part in this study you can contact the researcher by any of these methods:
- email
- by phone on: ------
- by writing (or sending the consent form)

What will happen with the results?

After I have written the transcript from the recording and observations I will contact you to arrange another appointment to discuss the transcript. At this appointment you can read through the transcript and seek clarification or make

any modifications as necessary. Once the research is complete I will arrange another meeting to discuss the overall results of the research and to answer any questions you may have. This research will be submitted as part of the Doctorate of Professional Practice in 2015/16. Anonymised data may be shared with the researcher's supervisor but care will be taken to protect the participant's identity.

It is planned to publish the research in an academic publication and to present the work at conferences. If you would like a copy of the research or of any subsequent papers that are submitted for publication please let me know and this can be arranged.

Who has reviewed the study?

University of Northampton and Northamptonshire Healthcare NHS Foundation Trust have given approval for this research to be conducted. The research has been given Ethical Approval by the University Research Ethics Committee and the NRES Committee East Midlands- Nottingham 1 Research Ethics Committee

This research is being completed as part of a Doctorate of Professional Practice and as such each stage of the research process is supervised by a research supervisor to ensure the methods used are safe, rigorous and robust.

Contact for further information.

For further information please contact:
- email
- by phone on: ---------.
- by writing (or sending the consent form) to: Hilda Hayo
-

If at any time you have any concerns about the way in which the study has been conducted you can contact the Research Supervisor:
> Professor Judith Sixsmith
> School of Health- University of Northampton
> Park Campus, Northampton. NN2 7AL
> Email: Judith.sixsmith@northampton.ac.uk

> Or:
> Complaints Facilitator
> Northamptonshire Healthcare NHS Foundation Trust
> Sudborough House
> St Mary's Hospital

London Road
Kettering
NN15 7PW
Tel.No: 0800 917 7206

Thank you for taking time to read this information.

Appendix 8: Consent Form

Title of the Research Study:

The full title of the research is:

"How and why does social connectedness change in families living with the effects of bvFTD?"

Principal Researcher:

Hilda Hayo - Research Student on the Doctorate of Professional Practice

NB. This form should be read in conjunction with the information leaflet provided.

Please initial box

1. I confirm that I have read and understand the information sheet for the above study and have had the opportunity to ask questions. ☐

2. I understand that my participation is voluntary and that I am free to withdraw at any time, without giving reason. ☐

3. I understand that anonymised data collected during the study may be examined by my supervisors at the University of Northampton and research regulatory bodies and consent for these individuals to have access to this data. ☐

4. I understand that that any information given by me will remain totally confidential and will be stored securely. ☐

5. I understand that the interview will be recorded using a digital voice recorder. ☐

6. I agree to take part in the above study. ☐

Participant's Name: -----------------

Participant's Signature: ----------------- Date:-

I confirm that I have explained the nature of the study, as detailed in the information leaflet, in terms, which in my judgement are suited to the understanding of the participant.

Signature of Researcher……………………………………….

Date………………………………….

Appendix 9: Reflective Analysis

All researchers are influenced by their past experience (Stake, 2006). In qualitative research it is important for the researcher to recognise that their beliefs, background, and social identity could have an impact on the research process (Lathlean, 2010). Reflexivity is the process "where researchers engage in explicit, self-aware analysis of their own role" (Finlay 2002, p531). Reflexivity is also important to examine the researcher's place in the enquiry, the process of knowledge generation and the factors which have influenced it (Guillemin & Gillam, 2004).

Finlay (2003) identifies five variants of reflexivity and she suggests that researchers should select the type that most suits the research. My research is underpinned by social citizenship and the transformative paradigm with the focus on providing a new way of thinking about the status of people living with dementia to promote active involvement, equal rights and entitlements as the rest of society. The variant of reflexivity that most suits my research is "reflexivity as mutual collaboration" which suggests that a relationship develops between the participants and the researcher which helps to shape the research process and findings. The reflections of both participants and researcher are equally important during the research process.

The research study:

Assumptions

I assumed that I would be able to easily recruit participants in the Midlands, but this was not the case as prospective participants who were approached were either unwilling to engage in research, or had various life crises that prevented them from engaging at the time. I had to recruit nationally which increased the time spent travelling and also led to a delay in starting the research but it did enable me to get an insight into how different areas of the country supported families who were living with the effects of bvFTD.

I thought it would be easy to recruit identified others from the social network of the family and person living with the effects of bvFTD. It rapidly became apparent that the social networks had already significantly reduced by the time the family became involved in the research. In fact all the families said that the social connectedness had significantly reduced before the person was diagnosed with bvFTD. They said that this was one of the key things they noticed months, and in two cases, years before the diagnosis. When I discussed this with the families they said that in order to involve identified others, there would need to be an earlier diagnosis so the social contacts had not reduced so markedly.

Another issue I had not fully considered, was that the children in the families would be undergoing counselling in their own right and so it would be unethical to involve them in my research where I was asking them about their feelings, thoughts and experiences.

Although two families continued writing a diary after the first month, the other three families found it too upsetting to do so and so stopped writing. When asked why the family members said that writing down what was happening made it seem so hopeless and made them think of how very different their lives had become in such a short period of time. The family members also said that they could also see how things were getting progressively worse which they found distressing. All families were willing however to talk through the changes and situations concerning the issues of social connectedness they were experiencing. Of the two families writing their diaries one is now writing a book as keeping the diary has generated unarticulated thoughts and feelings that they wanted to share, to tell their story for the benefit of others in the same position.

Families spoke at length of their experiences, feelings and distress at having a loved one diagnosed with bvFTD and for all of the interviews the only input I had was to listen and on occasion gently focus the interview back to social connectedness. All of the family members interviewed spoke afterwards of how cathartic the interview had been as they were not able to speak to anyone else about how they thought and felt about the diagnosis and the effects that this had had on the family.

As a clinical practitioner I found the distress that the families were going through (without any advice and support from formal or informal sources)

difficult because I naturally wanted to take the families onto my caseload but this would not have been practical given the geographical spread. Nor would it have been ethical in that I was there in the role of a researcher not a counsellor or specialist nurse. I addressed this by referring the families to appropriate support services such as Admiral DIRECT (national helpline manned by specialist dementia nurses); Alzheimer's Society and other support and services in their local area.

I involved families in every stage of the research and once I had transcribed the recordings I sent them to the families for their comments and verification. Once the themes were identified I then shared these with the participants and they discussed these with me and we made necessary adjustments. Once the thesis is finished and examined I plan to prepare articles for publication and have offered to share these with the participants pre-publication.