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BSc Hons MSc

EXPLORING THE EMOTIONAL IMPACT AND ADJUSTMENT
IN FRONTOTEMPORAL DEMENTIA FAMILY CARERS

Section A: Factors related to poor family carer wellbeing in
frontotemporal dementia: A review
Adjusted Word Count – 7933

Section B: Theorising the process of adjustment in family carers
of those with behavioural variant frontotemporal dementia
Adjusted Word Count – 9230

Overall Word Count – 17,163

A thesis submitted in partial fulfilment of the requirements of
Canterbury Christ Church University for the degree of
Doctor of Clinical Psychology

APRIL 2016

SALOMONS
CANTERBURY CHRIST CHURCH UNIVERSITY

Acknowledgements

Firstly I'd like to thank my supervisors, Dr Sue Holttum and Reinhard Guss, for their continuous support and guidance throughout this research process. I'm sure I wouldn't have managed this without the support of my wonderful family and friends, thank you all for your encouragement, for listening, cooking for me and just generally keeping me sane. I'm so lucky to have you. I would also like to thank my manager, Dr Rachel Terry, whose support has been priceless throughout this experience. A huge thank you to Jill Walton and the FTD network for allowing me to access participants through their group.

Last but by no means least, I can't put into words how grateful I am to my participants. I feel very privileged that they chose to share their stories with me and I hope I have managed to portray their experiences in this paper.

Summary of MRP

Section A

A review of the existing literature is presented which explores the factors related to poor emotional wellbeing in family members caring for someone with frontotemporal dementia. A total of 16 papers were reviewed. This review gives an overview of the findings of the studies and provides a critique of this research base. A range of factors were identified including patient and carer variables. Clinical implications and future research directions are discussed.

Section B

A grounded theory study exploring carer adjustment in behavioural variant frontotemporal dementia is presented. Semi-structured interviews were completed with 12 spousal carers. Results indicate a complex process of adjustment and acceptance in the context of an unusual progressive condition which is impacted by experiences prior to the onset of the condition and further complicated by the often lengthy and complicated route to diagnosis. Recommendations for services are discussed which may help to support the carers through this journey.

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Section A: Factors related to poor family carer wellbeing in frontotemporal dementia: A
review

Adjusted Word Count: 7933

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Abstract

Background - Research into carer wellbeing in dementia has indicated heightened levels of burden and distress than in other conditions. This has been particularly noted in those caring for someone with frontotemporal dementia (FTD). This rare form of dementia has an unusual presentation in the form of personality, behavioural and language changes.

Aim - This paper provides a narrative literature review exploring the possible factors associated with poor carer wellbeing in FTD.

Method and Results - Sixteen studies were reviewed. Generally findings varied, which may be due to a range of measures, variables and research methods used. Despite this, the findings suggest that factors related to both the person with dementia and their carer relate to poor carer wellbeing.

Carers of people with BV-FTD were more likely to have poorer wellbeing than other forms of FTD. Symptoms such as disinhibition, apathy and irritability displayed in the person with FTD were significant in considering the carer wellbeing. Time since diagnosis and living with the person with FTD was also associated with wellbeing. The coping style of the carer and the quality of the relationship between the carer and the person with FTD were also associated with wellbeing.

Conclusions - Findings from the studies varied, leaving it difficult to draw conclusions. The literature base is also methodologically limited. Despite these limitations, the review indicates that there are specific elements of FTD, particularly the behavioural variant, which make this a particularly difficult condition for carers to manage. The varied findings also imply an individual nature to experiences of carers. This confirms the complexity of the

Caring in FTD

condition and the need for targeted support for this particular population. Further research is recommended to explore the process of caring for someone with this complex condition.

Key words – frontotemporal dementia, carers, factors, wellbeing.

Introduction

A narrative literature review was conducted in order to investigate factors relating to carer wellbeing in frontotemporal dementia. An introduction to dementia, FTD and carer experiences is presented with a brief description of relevant policy documents and literature in order to provide a rationale for the review.

Search strategy and method are described followed by a summary of the results of the literature. A general critique of the literature base is presented, followed by suggestions for further research, clinical implications and concluding remarks.

Dementia: Definition

Dementia is an umbrella term referring to a range of conditions occurring as a result of structural or chemical changes in the brain. The condition is progressive in nature meaning further decline over time. The Diagnostic Statistical Manual – 5th edition (DSM; American Psychiatric Association, APA; 2013) classifies dementia as a neurocognitive disorder with clear diagnostic criteria including the presence of cognitive decline and decline in day to day functioning affecting the individual's independence. These must be present in the absence of delirium. This criteria has shifted over the years; the early version of the DSM (DSM-IV-TR; APA, 2000) stated that a person must display cognitive decline in two areas, one of which must be memory functioning. The later version does not highlight memory as an essential criterion. Although, dementia is more likely to affect people aged 65 and over, diagnoses of dementia are becoming more common in the younger population.

There are different subtypes of dementia, the most commonly diagnosed being Alzheimer's disease (AD), which accounts for 62% of cases (Prince et al., 2014). Alzheimer's disease is initially associated with the presentation of short-term memory difficulties and word finding in the early stages. However, other forms of dementia present

with different symptoms. Other forms include vascular dementia, Lewy body dementia and frontotemporal dementia. A Dementia UK report (Prince et al., 2014) states that there were 815,827 people in the UK with dementia in 2013. It is thought that this is set to increase to 1,142,677 by 2025.

A diagnosis of dementia is made by collating qualitative information from clinical interviews, brain imagery and cognitive testing. Memory clinics in the UK were introduced with the aim of making assessment, diagnosis and post-diagnostic support for dementia more accessible (NHS England, 2014) and were referred to as the ‘cornerstone of the diagnosis of dementia’ (p. 6). In 1990 there were 20 memory clinics. This has increased to 214 in 2013 demonstrating the increased diagnostic rate and therefore further need for support following the diagnosis. Despite this increase, dementia remains a priority within healthcare targets. The NHS mandate for 2016/ 2017 includes maintaining the diagnosis rate and improving support as objectives (Department of Health, DoH; 2015).

Caring in dementia

Of the current population of people with dementia it is reported that 493,639 (61%) currently live in the community (Prince et al., 2014). The Carer’s Trust report (2013) states that 670,000 family and friends are acting as the primary caregiver to someone with dementia. The impact of this on family members has been documented, for example the National Dementia Strategy (DoH; 2009) describes ‘profound, negative effects [of dementia]’ on family members (p. 7) and suggests early intervention as a way of improving family carer quality of life. The National Service Framework for Older People (DOH; 2001) highlighted the need for early diagnosis of dementia in order to support family members who may become ‘demoralised’ as a result of the circumstances. Research has described the impact of caring for someone with dementia. The Carer’s Trust (2013) describe that carers of people

with dementia report higher levels of stress and depression than other carers. They attribute this to the unique nature of the disease. This is consistent with the findings of Ory, Hoffman, Yee, Tennstedt and Schulz (1999) who report that carers of people with dementia were more negatively impacted than those caring for people with other conditions with respect to mental and physical health. The authors conclude that their findings imply that there is ‘something unique about caring in dementia’ (p.184).

There have been some attempts in the literature to explain the factors that may be associated with the above findings. Campbell et al. (2008) describe how certain dementia related symptoms and the level of need by the person with dementia amongst other factors were not associated with caregiver burden, however caregiver factors such as gender, overload, neuroticism and relationship quality were amongst factors accounting for 81.6% of the variance explaining caregiver burden.

Service guidance includes support for family members, for example National Institute for Health and Care Excellence (NICE, 2006) guidance recommends a carers assessment and consideration of carer’s psychological distress as an ongoing process. It is suggested that this be in the form of individual or group support, peer support and educational courses. Similarly, the Memory Services National Accreditation Programme (MSNAP) recommend improving training for healthcare professionals regarding the impact of diagnosis on family members and that carers should be offered an assessment and intervention, if appropriate (Royal College of Psychiatrists, 2015).

The above guidelines and research do not specify any subtype of dementia, nor does the guidance provide any further recommendations for supporting carers of the less common or more unusually presenting dementias.

Frontotemporal Dementia

Frontotemporal dementia (FTD) is a rare form of dementia. It is thought to account for only 2% of cases of the illness in the UK and is most common amongst men under 65 years of age (Prince et al., 2014). The term FTD refers to the parts of the brain which are affected in this subtype of dementia, namely the frontal and temporal lobes. FTD can present in different ways. In the DSM 5th Edition (DSM; American Psychiatric Association, 2013), FTD is identified by either the presence of behavioural symptoms such as disinhibition and apathy (namely behavioural variant, bv-FTD); or decline in language functioning, with ‘relative sparing of learning, memory and motor function’. There are two language variants, semantic dementia (SD) and progressive non-fluent aphasia (PNFA). Therefore, FTD is considered distinctly different to other forms of dementia in relation to symptomology and as a result the impact on the carers is thought to differ also. This, however is a new addition to the DSM, in the previous version (DSM-IV-TR, 2000), rarer forms of dementia such as FTD were considered ‘dementia due to other general medical conditions’, for diagnosis of which it was necessary to meet the more general criteria for dementia which included deficits in two or more cognitive domains, one of which had to have been a memory deficit. The more updated version of the DSM does not specify memory impairment as a criterion and therefore should enable more accurate diagnosis of FTD.

Dementia has been a frequently discussed topic of late with the guidance described above regarding rates of diagnosis and post diagnostic support, however rarer forms of the condition such as FTD are not mentioned in the guidance. The MSNAP document (Royal College of Psychiatrists, 2015) purely recommend that those presenting with ‘non-memory disorders’ should be referred to a neurologist or other specialist, which they state ‘may be memory service’. Therefore, guidance is rather generic and in practise is often still targeted towards memory problems.

Caring in FTD

Recent research has begun to explore the qualitative experiences of those caring for someone with a diagnosis of FTD. Relatives describe being faced with confusing and difficult situations including beginning to notice changes in a loved one (Oyebode, Bradley & Allen, 2013) and experiencing a range of difficult emotions such as isolation and anger in relation to changes in their loved one's behaviour (Massimo, Evans & Benner, 2013). Caregivers also describe the process of seeking medical support and diagnosis (Merrilees & Ketelle, 2010). As this is a rare form of dementia and as described above presents differently to more commonly understood forms, medical professionals can be unsure and a range of other diagnoses can be given prior to the FTD diagnosis (including mental health diagnoses such as depression).

Research has also focused on comparing the impact of caring in FTD with caring in the more common Alzheimer's disease. Differences were found between AD and bv-FTD carers on measures of strain, distress and perceived control (Wong & Wallhagan, 2012). Carer burden was found to be higher in caregivers of those with FTD than those caring for someone with AD (Boutoleau-Bretonniere, Vercelletto, Volteau, Renou & Lamy, 2008; De Vugt et al., 2006).

Despite these findings there are no specific guidelines recommended for supporting carers of FTD, therefore they often receive the same support as more common dementias such as AD. These findings have implications for clinical practice and suggest that FTD specific interventions may be appropriate. However, in order to tailor such interventions it would be helpful to know what unique factors, associated with FTD, lead to this increased burden. Therefore, specific interventions may be introduced to target these unique factors, in

order to reduce the possibility of mental and physical distress in this vulnerable group of carers.

Aim of current literature review

This literature review aims to answer the research question – What factors influence poor emotional and psychological wellbeing in relatives caring for someone with FTD?

This paper aims to review the empirical literature base which has looked specifically at factors associated with burden, strain and other psychological complaints in family carers of those with a diagnosis of FTD. The findings of this literature base will be presented in a narrative literature review describing the main factors identified as having an effect on this group of caregivers. A critique of the research base will be presented, along with consideration of future directions and clinical implications.

Methodology

A narrative literature review was conducted (Green, Johnson & Adams, 2006). This type of review has been described as presenting up to date knowledge on a topic from a contextual point of view. This type of review is not systemic in nature and it not designed to answer a specific quantitative research question (Rother, 2007). Due to the limited literature base in this area, this type of review enabled the opportunity to include research using a range of methodologies, measures and other variables in order to provide a broader understanding of the topic area.

Search strategy

A search of electronic databases was carried out. Databases included in this search were PsychInfo, ASSIA, OVID Medline and Cochrane. A set of predetermined search terms

were added into each database (see Table 1 for search terms and combinations). These terms were chosen to represent the review topic. It should be noted that FTD was previously referred to as Pick's disease and therefore both terms were included in the search. The aim of the review was to explore factors related to the psychological wellbeing of this group of carers, therefore a range of terms were selected to account for the general term 'wellbeing'.

Inclusion/ Exclusion

In order to answer the research question outlined above, studies were excluded if:-

- they were not written in English;
- FTD was not specified, for example the paper only mentioned early onset dementia;
- FTD was not analysed separately from other presentations;
- they were not original studies, for example literature reviews;
- the participants were not family caregivers, for example nursing staff;
- they were primarily concerned with Alzheimer's disease or other non FTD presentations;
- the paper was not deemed to specifically seek to answer the research question set out in this review.

See Figure 1 for a breakdown of the search results and exclusion process. This search strategy resulted in a total of 16 papers to be included in this review.

1. Frontotemporal dementia
2. FTD
3. Frontal lobe dementia
4. Pick's disease
5. Early onset dementia
6. Frontal lobe degeneration
7. Frontotemporal degeneration
8. Combination of above terms (1-7) using 'OR'
9. Caregivers
10. Carer*
11. Family
12. Relatives
13. Spouse
14. Combination of above terms (9- 13) using 'OR'
15. Combination of 8 'AND' 14
16. Burden
17. Quality of life
18. Distress
19. Depression
20. Stress
21. Psychological
22. Wellbeing
23. Combination of terms 16 – 22 using 'OR'
24. Search term 15 'AND' 23

Table 1. Search terms

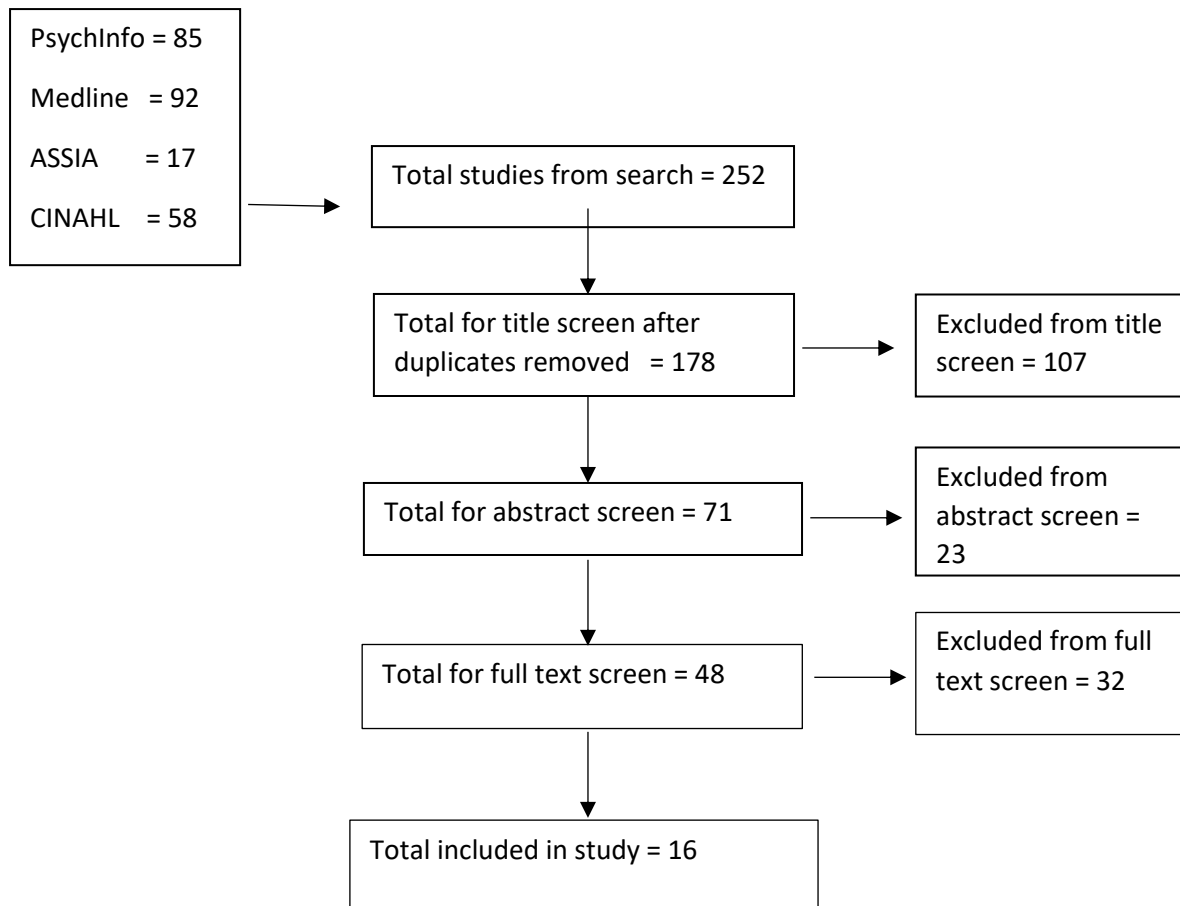


Figure 1 Flowchart of search results

Results

General Summary of Studies

As mentioned above 16 papers were included in this review. See Table 2 for a summary of each of the included studies.

<u>Reference</u>	<u>Country</u>	<u>Population</u>	<u>Measures</u>	<u>Analyses</u>	<u>Results</u>
Mourik, J.C., Rosso, S. M., Niermeijer, M. F., Duivenvoorden, H. J., van Swieten, J. C. & Tibben, A. 2004 Frontotemporal dementia: Behavioural Symptoms and Caregiver Distress.	Netherlands	63 patients, 34/63 caregivers were female, 81% were a spouse.	Neuropsychiatric Inventory (NPI) Global Deterioration Scale (GDS).	T-Tests Multiple Linear Regression Spearman's correlation	<p>Associated with caregiver burden</p> <p>Factors associated with carer burden were depression, irritability and aberrant motor behaviour in the person with FTD.</p> <p>Associated with caregiver distress</p> <p>NPI total scores and distress were strongly related ($r=0.70$; $p<0.001$)</p> <p>Delusions were highly distressing, although not commonly reported as a symptom.</p> <p>A significant relationship was found between GDS and caregiver distress, yet not strongly correlated ($r = -0.28$; $p< 0.03$)</p> <p>Female caregivers at home were more burdened than males.</p> <p>Symptoms such as delusions, hallucinations, irritability and agitation more predictive of caregiver distress ($p<0.001$), as were depression and anxiety in patient ($p<0.01$).</p>

					<p>Not associated Apathy, euphoria and disinhibition did not predict distress at the significant level (< 0.05).</p>
<p>Riedijk, S. R., De Vugt, M. E., Duivenvoorden, H.E., Niermeijer, M. F., Van Swieten, J. C., Verhey, F. R. J. & Tibben. A. (2006) Caregiver burden, health related quality of life and coping in dementia caregivers: A comparison of frontotemporal dementia and Alzheimer's disease.</p>	Netherlands	<p>63 patients FTD; 29 at home, 34 in nursing and 90 AD.</p> <p>Caregivers – 24-55% male depending on diagnosis 52-93% spouses (71-93% in FTD sample).</p>	<p>NPI Short form 36 health survey (mental and physical components); Utrecht coping list.</p>	<p>Regression analyses ANCOVA</p>	<p>Associated with caregiver burden</p> <p>Burden was predicted by the coping strategy of seeking distraction ($p<0.025$) and by passive coping style ($p<0.001$).</p> <p>Symptoms of anxiety ($p<0.01$) and disinhibition ($p<0.001$) also predicted burden.</p> <p>Those caregivers of patients living at home who had been diagnosed for a shorter duration were more heavily burdened.</p> <p>Associated with worse health related quality of life Worse mental health related quality of life was predicted by passive coping style ($p<0.001$) and shorter duration of illness ($p<0.05$).</p>
<p>Knutson, K. M., Zamboni, G., Tierney. M. C. & Grafman, G. 2008 Neural correlates</p>	U.S	<p>47 patients (25 fv-FTD; 22 CBS) and 14 controls.</p>	<p>Zarit burden inventory (ZBI); Mattis dementia rating scale (MDRS);</p>	<p>Pearson's correlation Multiple Regression</p>	<p>Associated with caregiver burden</p> <p>ZBI and NPI total scores were correlated ($r = 0.445$; $p=0.013$).</p>

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of caregiver burden in cortical basal syndrome and frontotemporal dementia			NPI.		Right orbital gyrus atrophy was correlated with ZBI scores ($p < 0.005$). Not associated Burden was not related to MDRS scores.
Riedijk, S., Duivenvoorden, H., Van Swieten, J.V., Niermeijer, M. & Tibben, A. (2009) Sense of competence in a Dutch sample of informal caregivers of frontotemporal dementia patients.	Netherlands	63 patients FTD and their caregivers; 46 caregivers included in follow up. 43% male, 78% spouse.	GDS; NPI; Sense of competence questionnaire; Symptom checklist-90-revised; Short form 36 health survey questionnaire (physical and mental components).	Pearson's correlation Multiple Regression	Sense of competence not related to dementia severity. Sense of competence had an effect on NPI and general burden. Sacrifice had an effect on physical (at $p = 0.001$) and mental quality of life ($p = 0.03$).
Mioshi, E., Bristow, M., Cook, R. & Hodges, J. R. (2009) Factors underlying caregiver stress in frontotemporal dementia and	UK	Patients – 79 FTD; 29 AD, (34 FTD in nursing home, 45 at home) Caregivers – 69-82% female.	NPI; Disability assessment of dementia (DAD); Perceived stress scale (PSS); Center for epidemiological studies	T-tests Logistic Regression	Female caregivers reported significantly higher levels of stress and depression than males ($p < 0.05$). Number of behavioural symptoms, severity of disease, activities of daily living, length of symptoms, social network or high contact roles were not associated with carer stress.

Alzheimers disease.			depression scale (CES-D); Social network index.		
Armstrong, N., Schupf, N., Grafman, J. & Huey, E. D. (2013) Caregiver burden in frontotemporal degeneration and corticobasal syndrome.	U.S	61 carers of FTD (52 with fv, 8 with PPA and 1 with SD)	Frontal Systems Behaviour Scale; ZBI	Logistic regression	Symptoms of frontal lobe dysfunction and apathy were associated with an increased possibility of caregiver burden than those without, further increased when both symptoms were present together.
Mioshi, E., Foxe, D., Leslie, F., Savage, S., Hsieh, S., Miller, L., Hodges, J. & Piguet, O. (2013) The impact of dementia severity on caregiver burden in Frontotemporal Dementia and Alzheimer's disease	Australia	Patients – 17 bv-FTD; 20 SD; 20 PNFA; 19 AD. Carers % women 70-79; spouse 65 – 90 % (Depending on diagnosis).	ZBI; DASS; Social network index, Intimate bond measure; Addingbrooke's Cognitive Examination-R, Frontotemporal dementia rating scale (FRS), Cambridge behavioural inventory revised (CBI-R).	ANOVA Multiple Regression	Carers in bv-FTD reported more burden and higher rates of depression than the SD and PNFA groups. Disease severity, caring relationship and caregiver depression were found to account for carer burden, but not apathy or 'abnormal behaviour'. Severity accounted for 48% alone. No differences were found between the different subtypes of FTD on scores of caregiver anxiety and stress.

<p>Cosseddu, M., Alberici, A., Pelizzola, S., Padovani, A. & Borroni, B. (2013) The other face of the coin: Caregiver burden in frontotemporal lobar degeneration</p>	<p>Not stated, authors are based in Italy.</p>	<p>56 FTLD patients (39 bv-FTD and 17 PPA; 64.3% male) Carers – 75% female; 73.2% spouses, 26.8% children; mean age 57.9.</p>	<p>Short form -36 health survey questionnaire (with mental component and physical component), CBI, Self-Rating Depression Scale, State Trait Anxiety Inventory, FBI.</p>	<p>Spearman's correlation Multiple regression</p>	<p>Associated with caregiver quality of life Older caregiver age, diagnosis of bv-FTD and patient's male gender were associated with worse physical health related quality of life. Severity of behavioural symptoms was associated with worse mental health related quality of life</p> <p>Associated with caregiver burden Burden was associated with bvFTD diagnosis and FBI total score yet in a multiple regression only severity (FBI score) was related to burden.</p> <p>Associated with caregiver depression Carer depression related to relationship with patient with spouses scoring higher than other caregivers</p> <p>Associated with anxiety Carer anxiety was associated with FBI scores.</p>
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<p>Merrilees, J., Dowling, G. A., Hubbard, E., Mastick, J., Ketelle, R. & Miller, B. L. (2013) Characterization of apathy in persons with Frontotemporal dementia and the impact on family caregivers</p>	<p>U.S</p>	<p>22 patient-caregiver dyads, 13 bv-FTD; 9 SD. All were spouse caregivers</p>	<p>Clinical Dementia Rating Scale, Mini Mental State Examination, NPI, Epworth sleepiness scale, Barthel Index (physical mobility). Sleep diary/ Actiwatch</p>	<p>T tests Spearman's correlation</p>	<p>Associated with caregiver distress Apathy was associated with distress for caregivers in all groups.</p>
<p>Hsieh, S., Irish, M., Daveson, N., Hodges, J. & Piguet, O. (2013) When one loses empathy: It's effect on carers of patients with dementia</p>	<p>Australia</p>	<p>31 patients (14 bv-ftd; 9 sd; 8 AD), 46 carers (18 bvftd; 14 SD; 14 AD) Spouses 84.4%; Female 76.1%.</p>	<p>Interpersonal Reactivity Index (IRI); ZBI; Intimate Bond Measure; Addenbrookes Cognitive Examination-Revised; Emotion selection task; CBI - R, clinical dementia rating; frontotemporal dementia rating scale.</p>	<p>ANOVA Spearman's correlation</p>	<p>Associated with caregiver burden Carers of the bv-FTD subtype reported greater burden than caregivers of SD Empathy scores on the IRI were associated with burden in the SD group. In bv-FTD burden was associated with lack of caring relationship.</p>

<p>Mekala, S., Alladi, S., Chandrasekar, K., Fathima, S., O'Connor, C. M. O., McKinnon, C., Hornberger, M., Piguet, O., Hodges, J. & Mioshi, E. (2013) Cultural differences are reflected in variables associated with carer burden in FTD. A comparison study between India and Australia.</p>	<p>India and Australia</p>	<p>69 patient=carer dyads from India (n= 31?) and Australia (n = 38).</p> <p>Unclear whether spouses only (definitely family caregivers)</p> <p>Female 61.3 (India) 78.9 (Australia)</p>	<p>ZBI; DASS 21, Frontotemporal dementia rating scale.</p>	<p>Mann-Whitney U Test</p> <p>Spearman Correlation</p>	<p>Anxiety was higher in Indian caregivers than Australian caregivers.</p> <p>In India and Australia, caregiver burden was associated with depression, anxiety and stress (p < 0.001).</p> <p>In neither India nor Australia, caregiver burden was not associated with dementia severity.</p>
<p>Diehl-Schmid, J., Schmidt, E-M., Nunnemann, S., Riedl, L., Kurz, A., Forstl, H., Wagenpfeil, S. & Cramer, B. (2013) Caregiver burden and needs in frontotemporal dementia.</p>	<p>Germany</p>	<p>94 caregivers considered for analysis; 87% bv-FTD and PPA in 13%</p> <p>80% spouses 72% female</p>	<p>Standardised questionnaire that addressed burden, problems and needs of FTD caregivers was developed. Caregiver strain index, Beck depression inventory.</p>	<p>Mann Whitney U test</p> <p>Kruskal-Wallis</p> <p>Spearman's correlation</p>	<p>Associated with caregiver burden</p> <p>Symptoms that were reported as burdensome for carers included changes in personality; aggression; misdemeanours; inflexibility; egocentric behaviour; lack of empathy, addictive behaviour; irritability; patient's dependency on the caregiver; and physical impairments.</p> <p>Associated with caregiver depression</p>

					<p>Associated with caregiver depression were; aggression; egocentric behaviour; addictions; and decreased sleep.</p> <p>Female gender, younger age at diagnosis and worsening of relationship were also associated with depression.</p> <p>Associated with caregiver strain The following symptoms were significantly associated with caregiver strain; difficulties with swallowing; misdemeanour; dependency; walking disability; speech problems; spatial disorientation; incontinence; apathy; and compulsive behaviours.</p> <p>Female gender caregivers and younger age at diagnosis were associated with strain. Financial difficulties also increased strain.</p> <p>Qualitative feedback</p> <p>What is the burdensome symptom? 45% said behavioural disturbances, 25% problems with communication, 30% need for care.</p>
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<p>Merrilees, J., Hubbard, E., Mastick, J., Miller, B. L. & Dowling, G. A. (2014) Sleep in persons with frontotemporal dementia and their family caregivers.</p>	<p>U.S</p>	<p>22 patient carer dyads; 13 bvFTD and 9 SD. Spouse caregivers. 14/22 female</p>	<p>Sleep and activity data using an Actiwatch Clinical Dementia Rating NPI Pittsburgh sleep quality index.</p>	<p>Spearman's correlation T-tests</p>	<p>Night time behaviours in patients with bvFTD associated with caregiver distress (p = 0.04).</p>
<p>Wong, C. C. & Wallhagen, M. I. (2012) Family caregivers of individuals with frontotemporal dementia: Exploring the relationship between coping and caregiver physical and mental health.</p>	<p>U.S</p>	<p>61 family caregivers of FTD</p>	<p>Brief coping orientations to problems experienced (Brief COPE) ; Couples Satisfaction Index NPI Short form health survey 12</p>	<p>Pearson's correlation T-tests Multiple regression</p>	<p>Associated with caregiver quality of life Problem focused coping was significantly associated with the physical component of quality of life. Caregiver age both contributed to the mental component of the quality of life scale.</p>
<p>Guevara, A. B., Knutson, K. M., Wassermann, E. M., Pulaski, S., Grafman, J. & Krueger, F. (2015) Theory of</p>	<p>U.S</p>	<p>bvFTD group 28; caregivers = 20; controls = 18 (FP task); controls = 14 (MRI).</p>	<p>MDRS; MRI scan; ZBI; National Adult Reading Test (NART);</p>	<p>T-tests Pearson's Correlation</p>	<p>Caregiver burden was associated with theory of mind impairment (p<0.01). Greater atrophy in lateral premotor cortex led to greater caregiver burden (p<0.05).</p>

<p>mind in patients with behavioural variant fronto-temporal dementia (bv-FTD) increases caregiver burden.</p>			<p>Faux pas (FP) task</p>		
<p>Roche, L., Croot, K., MacCann, C., Cramer, B. & Diehl-Schmid (2015) The role of coping strategies in psychological outcomes for frontotemporal dementia caregivers</p>	<p>Germany</p>	<p>94 caregivers included 68 were female; 79.8% were spouses. 87.2% bvFTD and 12.8% SD and PNFA</p>	<p>Packet of assessments including nursing care level, financial resources, caregiver strain index, Becks depression Inventory, Quality of life – Alzheimer’s disease, Brief COPE.</p>	<p>Mediation analyses Multiple regression</p>	<p>Associated with caregiver depression</p> <p>Nursing care level (NCL) did not predict caregiver depression. Strain predicted depression and dysfunctional coping</p> <p>Associated with caregiver quality of life</p> <p>NCL predicted caregiver quality of life. There was a significant relationship between caregiver strain and quality of life.</p> <p>Associated with caregiver strain</p> <p>Significant predictors of strain were found to be – dysfunctional coping, higher NCL, greater financial resources and an interaction between NCL and financial resources.</p>

Table 2 Summary of included studies

Sample

All of the studies identified use family carers as per the inclusion criteria. For each study which reported the type of carer relationship, the vast majority of participants were the spouse of the person with FTD. Armstrong, Schupf, Grafman and Huey (2013) report 'significant other' as the majority regarding relationship to the person with FTD. In all studies the majority of caregivers were female. However, not all studies report this information.

Measures

The studies included in this review investigate elements of caregiver wellbeing. These include caregiver distress, burden, stress, strain, depression, anxiety and quality of life. The studies use a range of measures to investigate the above, and these are listed in the summary of studies presented in table 2. It should be noted that whilst the aim of this review was to investigate psychological wellbeing, those using health related quality of life scales include both a physical and mental component and therefore both components are included in this review.

Methods of the studies

The methodology of the included studies varies. Some of the studies included in this review use a single group of FTD caregivers (Mourik et al., 2004) whereas others compare FTD subtypes, such as bv-FTD and SD (Mioshi et al., 2013; Cosseddu, Alberici, Pelizzola, Padovani & Borroni, 2013; Diehl-Schmid et al., 2013; Roche, Croot, MacCann, Cramer & Diehl-Schmid, 2015). Others include other diagnoses as a comparison group such as caregivers of people with Alzheimer's disease (Riedijk et al., 2006; Mioshi, Bristow, Cook & Hodges, 2009; Mioshi et al., 2013; Hsieh, Irish, Daveson, Hodges & Piguet, 2013). Some of

the studies use a control group as comparison (Knutson, Zamboni, Tierney & Grafman, 2008; Guevara et al., 2015).

Analyses

All the studies included are quantitative and use statistical analyses to explore the factors associated with elements of caregiver wellbeing. Most of the studies use an analysis of relationship, some use correlational analyses whilst others use regression analyses. Roche et al. (2015) additionally use a mediation analysis. Other studies use a method of comparison of groups, for example by using a t-test or ANOVA.

Summary of findings

The papers included in this review explore the effects of different variables in relation to caregiver wellbeing. This section will present a summary of the findings from the literature. Rather than presenting each study's findings individually, these findings will be presented according to the factors explored and are split into patient related and carer related variables.

Patient variables

The findings in this section relate to the effect of factors associated with the person with dementia on the caregiver's wellbeing. These included patient demographic factors such as age and residence; and factors associated with the disease such as behavioural symptoms and disease severity.

Age

Diehl-Schmid et al. (2013) found that the person with FTD's age, age at onset and age at diagnosis were all significantly negatively associated with caregiver strain, as measured by the caregiver strain index (Robinson, 1983). This suggests that when a person with FTD is

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younger, or diagnosed at a younger age, this is associated with higher levels of caregiver strain. However, these correlations were weak ($r = -.248; -.272$, respectively). Given that 80% of this sample were spousal caregivers, this finding may be due to factors related to the age of both the patient and the carer with regards to work commitments, financial pressures and family responsibilities in younger people as opposed to those who are diagnosed after retirement with older children. The authors also report higher strain in younger caregivers.

Interestingly, other findings suggest older caregivers reported worse scores for mental health related quality of life (Riedijk et al., 2006) and worse physical health related quality of life (Cosseddu, Alberici, Pelizzola, Padovani & Borroni, 2013), again the majority of whom were spousal caregivers in both of these studies. Therefore, there may be some difference between these groups with regards to the measures used.

Gender

Cosseddu et al. (2013) found a significant relationship between a person with FTD's gender and the physical component of health related quality of life in the carer, suggesting worse physical quality of life when a person with FTD is male. Given that 73.2% of the caregiver sample in this study were spouses, this is likely to be linked to findings described below which suggest female carers have worse levels of wellbeing than male carers.

Residence

Some of the included studies investigated whether there was any difference in caregiver wellbeing between those whose loved one remained at home and those whose loved one resided in a nursing home, and the findings varied. Mourik et al. (2004) found that caring for someone with FTD living at home predicted higher scores on the Neuropsychiatric Inventory (NPI; Cosseddu, Alberici, Pelizzola, Padovani and Borroni, 2013) distress scale than those with their relative in a nursing home.

Riedijk et al. (2006) reported mixed results. They found that those caring for a relative at home had worse scores on the mental component of health related quality of life. However in contrast, they reported no significant differences found between carers of those in care and living at home on physical health related quality of life. In addition, Diehl-Schmid et al. (2013) found no differences in caregiver depression between those with relatives living at home or in care.

Therefore, the above suggests that those caring for someone with FTD at home experience worse mental health related quality of life and distress than those with their relative in care, but this was not the case for physical health related quality of life or depression. Those at home may have found it more difficult to have independence and time away from the caring role, whereas those with a relative in care may have better quality time for self care.

It would be interesting to consider the distance of the residential care in respect to the carer's home, and whether this has an influence on physical health related quality of life, for example with regards to time spent commuting, or whether those closer to home feel more obliged to visit more often.

With regards to carer depression the findings reported suggest that the residence of the person with dementia is not significant and other factors may be considered relevant. Both groups were managing the situation of seeing a loved one with a progressive illness and whether they reside at home or in care, this is likely to impact on low mood.

Severity of dementia

Studies measuring the association between severity of the dementia and caregiver wellbeing also produced mixed results. Cosseddu et al. (2013) found that greater severity of behavioural symptoms were associated with worse mental health related quality of life, and

greater severity was also related to higher caregiver anxiety. Additionally, Mioshi et al. (2013) found that severity of the dementia accounted for 48% of the variance explaining caregiver burden when using a regression analysis. However, they do acknowledge that some caregivers reported high levels of burden at less severe stages, implying that other factors may also be important in understanding caregiver burden.

In contrast to Mioshi et al. (2013), Knutson et al. (2008) found that dementia severity as measured by the Mattis Dementia Rating Scale (MDRS; Jurica, Leitten & Mattis, 2001) was not related to scores on the Zarit burden scale (ZBI; Zarit, Orr & Zarit, 1985). Similarly, Mioshi et al. (2009) found that severity of dementia was not associated with caregiver stress and Mekala et al. (2013) also found that in both an Indian and Australian sample, dementia severity did not contribute to caregiver burden. In Mourik et al.'s (2004) multiple regression analysis they report no significant association between dementia severity and caregiver distress.

Therefore, it potentially not the severity of the dementia itself but other factors which influence carer wellbeing. The section below related to symptoms of dementia implies that it may be more likely that it is the type of symptoms rather than the severity which is significant.

Nursing care level

Nursing care level refers to the amount of time the person with FTD requires support with tasks such as personal care, feeding and mobility (Diehl-Schmidt et al., 2013). Diehl-Schmidt et al. (2013) found that the nursing care level of the person with dementia was not associated with depression but did contribute to higher strain. Therefore, those caring for someone with higher needs reported higher levels of strain. Similarly, Roche et al. (2015) found that nursing care level did not predict caregiver depression, however strain mediated

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the relationship between nursing care level and depression. Nursing care level did predict caregiver quality of life, and again strain mediated this relationship.

Diagnosis of the behavioural-variant subtype of FTD

Studies included compared elements of caregiver wellbeing between the various subtypes of FTD. The results indicate that those caring for someone with the behavioural variant of the disease had worse levels of wellbeing than those with the language based subtypes such as semantic dementia (SD), and progressive non-fluent aphasia (PNFA). Mioshi et al. (2013) found that those caregivers of people with the behavioural variant of bv-FTD had significantly higher levels of burden than those with SD and PNFA. Similarly, Cosseddu et al. (2013) and Hsieh, Irish, Daveson, Hodges and Piguet (2013) found that burden was associated with the diagnosis of bv-FTD. Armstrong et al. (2015) found carers of those with frontal lobe dysfunction had 13 times the odds of experiencing burden than those without, this was irrespective of the particular diagnosis. This links with the section below regarding symptoms associated with carer wellbeing, which suggests that symptoms more commonly experienced in the behavioural variant were found to be more difficult for caregivers to manage.

Duration of illness

Riedijk et al.'s (2006) findings indicate that those caregivers of someone with a newer diagnosis experience worse health related quality of life than those who had been diagnosed for longer. However, Mioshi et al. (2009) found that the duration of illness was not associated with caregiver stress. Similarly, Cosseddu et al. (2013) found no association between disease duration and caregiver burden. The differences in these findings, as with others in this review are likely due to the different constructs measured, for example health related quality of life, stress and burden. Research described below including Riedijk et al.,

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2006 suggests that the coping style of the caregiver has impact on their health related quality of life, therefore it may be worth considering whether coping styles of caregivers change over time, therefore impacting on their wellbeing.

Number of symptoms

Research investigated the association between the number of FTD symptoms experienced, as measured by the NPI and caregiver wellbeing. Mourik et al. (2004) found NPI total scores were significantly associated with distress in the caregiver. Knutson et al. (2008) supported this, finding that NPI total scores were correlated with scores on the ZBI, indicating that greater behavioural symptoms were related to higher caregiver burden.

However, in contrast to the above, in a regression analysis Mioshi et al. (2009) found that number of symptoms recorded on the NPI did not predict caregiver stress. In fact, the only associated variable with carer stress was carer depression.

Specific behavioural symptoms

Research also investigated the effects of individual symptoms associated with FTD on caregiver wellbeing. The results of the studies included in this review vary greatly and reflect the complex nature of FTD.

Mioshi et al. (2013) found no significant relationship between caregiver burden and a person with FTD's 'abnormal behaviours' (for example, disinhibited or impulsive behaviour) and 'stereotypical behaviours' (for example, repetitive behaviours) as measured by the Cambridge Behavioural Inventory – Revised (CBI-R; Wear et al., 2008).

Diehl-Schmid et al. (2013), however, explored which behavioural symptoms commonly associated with FTD were perceived as most burdensome for caregivers, using their own questionnaire which they report as standardised but provide no further information.

They found that more than 80% of their sample reported the most burdensome symptoms to be changes in personality, aggression, lack of manners, occurrence of misdemeanours, inflexibility, egocentric behaviour, irritability and gambling. They found that some of these behaviours such as aggression and egocentric behaviour were also related to caregiver depression.

Mourik et al. (2004) found that the presence of irritability in the person with FTD was significantly correlated with caregiver distress. Their regression analysis also indicated that irritability was amongst the cluster of behavioural symptoms which was most predictive of distress. Agitation was also amongst this cluster of symptoms.

They also found that whilst delusions were not as commonly present as other symptoms, when they were present they were significantly correlated with caregiver distress. Delusions along with hallucinations were also amongst the cluster of behaviours which was reported to be more predictive of caregiver distress. Aberrant motor behaviours and depression in the person with FTD were also found to be significantly correlated with distress.

Euphoria was least associated with caregiver distress in Mourik et al. (2004). Apathy was also found not to predict caregiver burden, and this was also supported by Mioshi et al. (2013). However, in contrast to this both Merrilees et al. (2013) and Armstrong et al. (2013) found that apathy was associated with both caregiver distress and burden.

Similarly, Mourik et al. (2004) found that disinhibition did not predict burden in their regression analysis, however Riedijk et al.'s (2006) findings contradicted this. Armstrong et al. (2013) also found that when a person with FTD presented with disinhibition, the caregiver had 11.3 times the odds of experiencing burden. However, the authors point out that

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disinhibition was rarely present without apathy, and most predictive were disinhibition and apathy together.

Both Mourik et al. (2004) and Riedijk et al. (2006) concluded that the presence of anxiety in the person with FTD predicted burden in the carer.

Merrilees, Hubbard, Mastick, Miller and Dowling (2014) found that in carers of people with bv-FTD, there was a significant correlation between night-time behaviours in the person with FTD and caregiver distress.

The results in this section vary, and it is noted that there were a range of measures used and a range of research method adopted. It may be that whilst certain symptoms were experienced negatively for some carers and not others, other factors are clearly impacting. This highlights the complex nature of the disease and the difficulty in managing extraneous variables when researching this condition. There is no mention as to whether the symptoms are new or long standing, which may be relevant to the impact this has on the carer, as it may imply whether or not the carer has had the opportunity to develop strategies to cope with the behaviours.

Daytime activity

Merrilees et al. (2013) concluded that ‘disruptions in daytime activity are associated with significant carer distress’ (page 5), however upon investigation this was not tested. The authors found that the number of immobility bouts in the person with dementia was related to apathy scores, and that apathy was associated with distress but did not investigate directly the relationship between immobility bouts and distress.

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Theory of Mind

Whilst FTD is more commonly related to the behavioural and language impairments described above, Guevara et al. (2015) found a significant correlation between a task used to measure theory of mind ability (the ability to understand that other's have their own perspective) in their sample of people with FTD and caregiver burden, indicating that impairment in theory of mind is associated with increased caregiver burden. This cognitive ability may link with some of the behaviours which are common in FTD such as disinhibition (not understanding why something is inappropriate) and therefore may be linked with research above which indicates this behavioural symptom as a source of burden in caregivers.

Activities of daily living

In addition to behavioural symptoms, Diehl-Schmidt et al. (2013) found that other aspects of the disease such as the need to supervise the patient at home, the patient's dependency on the caregiver and physical impairments were associated with increased strain for the caregiver. However, using the Disability Assessment of Dementia (Gélinas, Gauthier, McIntyre & Gauthier, 1999) Mioshi et al. (2009) found that patient difficulties in ADL's did not contribute to caregiver stress. Therefore, it may not necessarily be functional aspects of the disease which are related to caregiver wellbeing.

Neurological changes

Two of the included studies aimed to investigate whether there was any relationship to atrophy in the brain of the person with dementia and burden experienced by the carer. Knutson et al. (2008) initially reported no significant correlation between grey matter changes and caregiver burden, however when they adjusted the level of significance to a less stringent $p < 0.005$, they found scores on the burden assessment were significantly correlated with atrophy in the right orbital gyrus. Whilst this area of the brain was not found to be

associated with total NPI scores (number of behavioural symptoms), atrophy in this area was associated with apathy and aberrant motor behaviour in the person with dementia. As mentioned above, some of the research found an association between carer wellbeing and aberrant motor behaviour and apathy.

Guevara et al. (2015) also investigated the association between atrophy in the brain of the person with FTD and burden experienced by their caregiver. They found an association between burden and voxel density in the left lateral premotor cortex, suggesting that a deficit in this specific part of the brain was associated with higher levels of burden. They link this with theory of mind ability.

Caregiver variables

Studies included in this review aimed to establish if there was a relationship between caregiver variables and their wellbeing.

Age of Caregiver

Research described earlier investigated links between patient age and caregiver wellbeing, which suggested that younger patients resulted in poorer carer wellbeing. Given that the majority of the sample were spousal caregivers, it is likely that the caregivers were of similar ages to the person with FTD and therefore the age of the caregiver may be significant. However, Riedijk et al. (2006) found that the age of the caregiver was positively correlated with worse mental health related quality of life with older caregivers being more affected than those younger, however this was only for those cases where the dementia had been diagnosed for a shorter period of time. Additionally, Wong and Wallhagen (2012) found that caregiver age alone accounted for 32% of the variance of mental health related quality of life. Cossedu et al. (2013) found that older caregivers reported worse scores for the physical

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component of health related quality of life, however this may not be surprising and not necessarily related to caregiving and more due to general ageing.

Contradictory to these findings, Diehl-Schmid et al. (2013) found that strain was higher in younger caregivers. Again this may not be surprising given that those older caregivers are likely to have more physical health complaints, however strain refers to an excessive demand on an individual and may be more likely to affect those younger caregivers who have other pressures such as work commitments and young families.

Gender of caregiver

Mioshi et al. (2009) found that female caregivers reported significantly higher levels of stress and depression than males, although this was not purely the case for caregivers of someone with FTD but was also the case in their Alzheimer's disease carer sample. Diehl-Schmid et al. (2013) found that more female caregivers had depression and higher levels of strain than males. These findings would be consistent with earlier reported findings which suggested that carer wellbeing is lower when caring for males with FTD, given that the majority of caregivers in the research were spouses. Mourik et al. (2004) found that gender alone did not predict carer distress, however the interaction between caregiver gender and the residence of the person with FTD was a predictor of distress. The authors found that female carers, when living with the person with dementia, scored higher on the distress scale than males living with their relative.

Relationship to patient

As mentioned above, the majority of the caregivers in the research presented were spouses to someone with FTD and therefore research investigating whether this relationship was a factor in carer levels of wellbeing may be skewed by sample size. Cosseddu et al. (2013) found that spouses of those with FTD reported higher levels of depression than other

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family caregivers, however, Riedijk et al. (2006) found that the type of relationship between the caregiver and the patient was not associated with burden experienced by the caregivers. Similarly Diehl-Schmid. (2013) found no differences in depression between spouse or child carers. However, there was no information provided regarding other family support, or external support, which may also have an influence.

Sense of competence

Riedijk et al. (2009) investigated whether a caregiver's perception of self-competence could be related to their wellbeing. They found that self-competence in the caregiver was not related to the severity of the dementia and that self-competence did not have an effect on burden as measured by the NPI. It may be suggested that the construct of competence be dependent on other factors such as time since diagnosis, therefore when a dementia is more severe, the caregiver may have learnt ways to cope and/ or sought helpful support. The author found that 3 factors made up self-competence, namely 'sacrifice', 'emotions' and 'attributions'. One of these factors 'sacrifice' was found to have a significant relationship with both the physical component of health related quality of life ($p=0.001$), and the mental component ($p=0.03$), however, this seems to link with other factors such as level of dependency.

Coping strategies

Research has investigated whether the coping style of the caregivers of someone with FTD is associated with their wellbeing. Riedijk et al. (2006) found that burden in caregivers was predicted by the coping strategy of seeking distraction and passive coping style. A passive coping style (related to a feeling of helplessness in relation to the diagnosis, for example, avoidance) was also associated with worse scores on the mental component of health related quality of life. Roche et al. (2015) found that dysfunctional coping (such as

venting, self-blame and denial; Su et al., 2015) amongst other factors predicted caregiver strain, but also found that strain predicted dysfunctional coping. Therefore it is unclear how these two factors combine, and it may be that those who are experiencing more strain use more dysfunctional coping strategies as a consequence.

In contrast, however, Wong and Wallhagen (2012) found that neither dysfunctional coping nor emotion focused coping was related to either component of health related quality of life in caregivers, but did find that problem focused coping was related to the physical component, suggesting that those using problem focused coping reported better physical quality of life. Yet, despite these findings the authors report that in a multiple regression analysis emotion focused coping made a statistically significant contribution to greater mental health related quality of life.

It is likely, however, that coping style may not be stagnant and actually changes over time, therefore this research may be significant in considering early interventions in supporting the carers to develop more functional methods of coping.

Carer depression

Mioshi et al. (2009) found that caregiver depression alone accounted for 58.2% of caregiver stress. In a later study, Mioshi et al. (2013) found that caregiver depression was related to caregiver burden. Similarly, in both an Indian and Australian sample, depression and stress was associated with caregiver burden (Mekala et al., 2013). As with other literature included in this review, these are correlational studies and do not indicate direction. However, this research does indicate the necessity of support for this group of caregivers as it has been indicated that they experience more burden than other groups and therefore may be more prone to stress and depression.

Level of strain

Similarly to the previous section, Roche et al. (2015) found that strain predicted caregiver depression, and was significantly related to carer quality of life, again indicating the need for support.

Quality of the relationship between the carer and the person with dementia

Diehl-Schmid et al. (2013) found that 50% of their caregiver sample reported that their relationship with their loved one had worsened and that this was associated with carer depression. Other studies support this finding. Mioshi et al. (2013) found that ‘caring relationship’ scores on the Interpersonal Reactivity Index were related to carer burden but ‘controlling relationship’ was not, indicating that when a relationship was perceived as less caring, this was associated with increased burden in the carer. These findings may link with behavioural symptoms described earlier. Similarly, Hsieh et al. (2013) found that for behavioural variant FTD burden was associated with empathy loss and lack of a caring relationship. Additionally, Wong and Wallhagen (2012) found that scores on the couple’s satisfaction index, which measures relationship satisfaction, accounted for 11.6% of the variance for the physical component scale of the caregivers health related quality of life. This appears particularly significant given the type of symptoms that occur in FTD, for example communication difficulties, apathy, loss of motivation, disinhibition and loss of empathy.

Other factors

Other factors which were also explored in relation to poor carer wellbeing were social support, financial resources, cultural differences and quality of their relationship with the person with FTD.

Social support

Mioshi et al. (2009) reported that the carers perceived level of social support did not contribute to stress.

Financial resources

Diehl-Schmid et al. (2013) found that financial difficulties increased caregiver strain. Similarly, Roche et al. (2015) found that financial resources were a factor predicting strain in caregivers, and more so when combined with the patient's nursing care level.

Cultural differences

Mekala et al. (2013) was the only paper to compare burden, depression, stress and anxiety across different cultures. They found that only anxiety was significantly different between an Australian and Indian sample. Anxiety was higher in the Indian sample, but no other differences were found. The authors, however, do not refer to using culturally adapted measures.

General Critique

Each individual study was assessed using the Crowe Appraisal Tool (Crowe & Sheppard, 2011) and is presented in a table in Appendix A. A copy of the tool itself is also included in appendix B to illustrate the scoring system. Given that the papers included in this review used different design methodologies, this tool was selected as it can be used for all research designs unlike other appraisal tools. It was also simple to use and allowed for direct comparison between the different studies used for overall score as well as each individual domain. A more general critical evaluation of the literature base is provided below. It is

important to highlight that this literature base used a variety of different measures, methods and analyses, therefore it is difficult to form particular conclusions from the literature.

Population

One of the main limitations highlighted by the authors within the included studies is that of small sample sizes (Hsieh et al., 2013; Wong & Wallhagen, 2012), with numbers of caregivers of people with FTD in the studies varying from 22 to 94 (not including control or comparison groups). Whilst it is likely that these small sample sizes represent the rare incidence of FTD in the population itself, the result of such small sample studies should be interpreted with caution as the findings may not generalise to a larger population. Wong and Wallhagen (2012) describe attempts to account for this in their data analysis, whilst others do not.

In addition to the small sample sizes used, Wong and Wallhagen (2012) describe difficulty in generalisability of results due to other demographic features of the samples used. They describe a majority white sample and therefore explain that their results may not be generalisable to other cultural groups. Similarly, in all of the studies the majority of carer participants were female. Again this appears to be representative of this population but the implications of the results are therefore difficult to generalise to male caregivers.

Many participants in the samples described were sourced from FTD support groups, therefore as Roche et al. (2015) point out, it is interesting to consider how these results therefore appear to represent this particular sub-section of the overall population having already sought help. Therefore, the results of studies derived from a sample who are not in contact with services may paint a different picture. However, recruitment of such a sample would be difficult.

In Knutson et al. (2008), further information on demographics was not available, and this limits the possible interpretations of the findings.

Methodology

There was a range of methods used in the literature presented in this review. Comparison groups (for example, between different diagnoses) were used in 10 of the 16 studies included (Riedijk et al., 2006; Knutson et al., 2008; Mioshi et al., 2009; Mioshi et al. 2013; Cosseddu et al., 2013; Merrillees et al., 2013; Hsieh et al., 2013; Mekala et al., 2013; Diehl-Schmid et al., 2013; and Merrillees et al., 2014). Control groups were used in Knutson et al. (2008), Wong and Wallhagen (2012) and Guevara et al. (2015). The lack of such comparison groups was identified as a limitation in Merrillees et al. (2014).

The studies in this review use cross-sectional design to gather data. Again, some of the authors acknowledge this as a limitation (Riedijk et al. 2006; 2009; Mioshi et al., 2013). This design reduces the possibility to infer causality for findings presented above such as the effect of duration of illness on caregiver wellbeing, as this information is only taken at one snapshot of time. The use of a longitudinal design to assess change over time would be recommended.

It is likely that other confounding factors may explain some of the findings presented above. Guevara et al. (2015) point out that in their study, the association found between a person with FTD's theory of mind and the caregiver's wellbeing may be due to other variables which were not assessed.

Outcomes

There is a wide range of outcome measures used within the literature included in this review. It is important to consider whether the scales used actually fit the purpose of measuring what is claimed to be measured. All papers with the exception of Diehl-Schmid et al. (2013) used existing standardised measures. Diehl-Schmid et al. (2013) chose not to use these as they report that the use of their own measure allowed for less repetition in the data collection. However, this does make the results difficult to compare to other studies and brings into question the reliability and validity of the measure.

There are also some limitations in the use of the existing measures, for example Mourik et al. (2004) used the NPI to assess behavioural disturbances and as a measure of carer burden. Whilst this measure is reported to have adequate levels of validity and reliability (Cummings, 1997), it assesses distress in relation to behavioural symptoms within the patient and not necessarily general burden. Whilst the ZBI was used by Mioshi et al. (2013), they describe their use of this measure as unidimensional.

Additionally, limitations of other measures were highlighted. Mourik et al. (2004) point out that the NPI is an observer-rated measure. It is, therefore, interesting to consider how much objectivity is present, or whether scores indicating the severity of behavioural symptoms themselves may be affected by caregiver stress. For example, those caregivers who are experiencing higher levels of stress or burden may simply be reporting more behavioural symptoms and severity. Wong and Wallhagen (2012) describe the limited internal consistency with their choice of measure, the Brief COPE (Carver, 1997). Armstrong et al. (2013) describes the measure they used as 'narrow in scope' and that it does not assess particular areas which the NPI does. Hsieh et al. (2013) also describe limitations

to their use of a standardised measure for empathy and suggest that the additional inclusion of observation measures be a potential area for future research.

It is also important to consider whether the outcome measures themselves had been validated for this particular sample group, for example, Roche et al. (2015) explains that one of their measures (Caregiver Strain Index) was developed for more physical conditions and not dementia, although had been validated for dementia in one study. They also explain that the quality of life measure they use was not validated for caregivers, rather it was validated for those receiving care.

Whilst not suggesting a limitation in a particular measure, Roche et al. (2015) also indicate that using nursing care level as a measure for this group of patients may itself be a limitation. This may be more appropriate for other presentations and not necessarily an indication of dementia progression in a more behavioural or language centred illness.

Whilst Mekala et al. (2013) were the only study to compare groups from different cultural backgrounds, the authors highlight that some difficulty with using the same measures between two different cultures as the concepts explored (eg. anxiety) may be experienced differently in different cultures.

Analyses

Eleven of the 16 studies used t-tests or ANOVA analyses to make direct comparisons between groups (Mourik et al., 2004; Riedijk et al., 2006; Mioshi et al., 2009; 2013; Merrilees et al., 2013; Hsieh et al., 2013; Mekala et al., 2013; Diehl-Schmidt et al., 2013; Merrilees et al., 2014; and Wong & Wallhagen, 2012). All 16 studies present results of analyses of association (correlation and regression analyses). It is therefore important to indicate that a correlational analysis shows only a relationship between variables and does not imply causality. Closer inspection of the data indicates that the directions of the significant

relationships within selected studies are unclear. An example of this would be considering if stress levels are a result of or a risk factor to the use of dysfunctional coping (Wong & Wallhagen, 2012).

It should also be mentioned that in Knutson et al. (2008) a significant result is only present when the p value is adjusted, and studies report significance at varying levels (eg. $p < 0.05$ or $p < 0.001$).

In using t-tests and ANOVAs to make comparisons between groups, the authors are able to determine whether difference is present in association with the independent variable; in these studies often diagnosis. It is inferred that the groups are comparable with regard to other confounding variables. Even when comparing groups with regards to demographics there could be many other factors which are important to consider.

Discussion

Summary of results

Previous research has suggested that FTD is a particularly difficult experience for family members of those diagnosed (for example, Boutoleau-Bretonniere et al., 2008). This narrative literature review summarised 16 studies which looked specifically at the factors that relate to poorer aspects of wellbeing in family caregivers of those with FTD. The studies used a range of measures and variables when investigating this. The findings indicate that a mixture of patient and caregiver variables have been shown to be associated with psychological wellbeing of the caregiver. Whilst the findings varied, of significance was the caregiver's perception of the quality of their relationship. Where the relationship was perceived as less caring, caregiver wellbeing was impacted. This is particularly relevant to this type of dementia since the commonly presented symptoms in FTD including personality and behavioural changes inevitably impact on the relationship. Symptoms such as apathy,

disinhibition and stereotypical behaviours were also associated with caregiver wellbeing. In relation to this, those caring for someone with the behavioural variant of the condition were more likely to have poor wellbeing, than the other subtypes. Although findings varied, time since diagnosis, female gender of caregiver and spousal relationship to the patient were related to caregiver wellbeing.

The findings imply that there are a range of factors which impact the caregiver, and suggest that this is more of an individual experience than simply cause and effect in relation to one variable. Additionally, there appears to be a potential process occurring rather than purely a stagnant situation in which carers either experience difficulties or do not.

Regardless, it appears that there are particular elements of FTD and particularly bv-FTD which need further consideration and services required which understand the complexity of this condition. Whilst guidelines do emphasise the need for carer assessment and support (MSNAP, 2016; NICE, 2006), FTD or other rarer forms of dementia are not specified. The research in this review and other research indicate that this group of caregivers are particularly vulnerable to depression, burden and strain, and indicates a need for adapting current support for these needs.

Limitations of current review

There are limitations with this current review. In relation to search strategy used there were some areas of difficulty. Whilst it is felt that the method of selecting relevant search terms was a thoughtful process in the present review, some difficulty was experienced in relation to the terms and their constructs which were explored. Difficulty in defining ‘wellbeing’ has been documented in the literature (Dodge, Daly, Huyton, & Sanders, 2012). Whilst a range of search terms were used, it was possible that some relevant studies may have been missed as a result of the selected terms.

The databases searched were relevant to the current review, however, this was not a systematic review of the literature and therefore the use of these limited databases may have excluded studies which were unpublished, or held by different databases. Additionally, only studies written in English were included.

One of the main difficulties with this search was that there were many papers which included FTD but did not differentiate this group from their whole sample, therefore they were excluded from the study but may have provided some additional data.

Despite the above limitations it is suggested that this review achieved its aim of investigating the literature exploring the factors related to family caregiver wellbeing in FTD and provides an overview of the literature and a summary of overall findings.

Clinical Implications

Despite the range of results presented in this review, it is accepted that caring for a relative with FTD is a highly stressful experience. This should be considered in clinical settings. Systemic approaches to dementia care are recommended. NICE guidance (2006) recommends carer targeted care plans including carers' assessments, practical and psychosocial support such as therapeutic intervention, social groups and psychoeducation for carers of those with dementia. This guidance is recommended for dementia in general. The findings in the current study highlight the complexity of caring for a person with FTD and the many factors which may make this difficult for the caregiver. It is, therefore, important that services particularly consider this group of individuals and target more specific interventions and support.

Rahman (2014) discusses 'alzheimerisation' in his article. In this particular article the author is concerned that assessment and diagnosis of dementia often follows an Alzheimer's type dementia pathway related to memory problems and is not set up for other types of the

condition. It is suggested that post-diagnostic services have a similar issue and, due to vague guidance related to support for caregivers, this support is predominantly focused on Alzheimer's presentations. There are national support groups for FTD, and services should be aware of these and how to signpost carers to these groups. Additionally, further training to memory clinic teams, general practitioners and mental health teams relating to the complexities of FTD would increase understanding of the condition and the impact on carers. Whilst post-diagnostic groups may be helpful, given the rare rate of diagnosis it is likely that carers of FTD would be under represented and may not benefit as much as those caring for someone with AD. Therefore, individual sessions with FTD carers may be more helpful.

The content of post-diagnostic support for FTD carers also requires less emphasis on memory impairment and cognitive strategies to support these impairments, for example, life story work, and cognitive stimulation groups. It is suggested that FTD support provide an opportunity for carers to discuss management of complex behavioural symptoms and to explore changes within their relationship and how to cope with these. Clinical psychologists within memory clinic teams have a particular role when considering the level of distress experienced in relation to some of the FTD specific symptoms, and it is suggested that therapeutic sessions should be considered for this population.

Future areas of research

As alluded to above, the research to date has investigated factors related to poor carer wellbeing based on cross-sectional methods, and this makes it difficult to explore factors such as impact of disease severity and length of disease duration. It is therefore recommended that future research focuses on longitudinal approaches.

It is also important to consider future research using participants which were less represented in this research to date, as the majority of the included samples were female,

white and already in contact with support networks. It would therefore be useful to consider how these findings relate to more culturally and socially diverse groups.

Further research into psychosocial interventions for this group would also be recommended, particularly with an emphasis on implementing these into clinical settings.

Since the research to date includes very varied findings, there are clearly many factors which may be important when considering the wellbeing of these caregivers. Since some of the findings above suggested that spousal caregivers of people with the behavioural variant may be particularly at risk of poorer wellbeing, it is recommended that further research with this group of individuals may increase understanding about this unique experience.

The research above indicates that particular coping styles are associated with caregiver levels of wellbeing, yet also indicates that wellbeing does not deteriorate as the dementia progresses. It is also proposed that there is a process of learning to adjust to their situation. It would be of interest to explore how these attitudes to the condition and ways of coping develop within this population.

The range of findings in this review and the contradictions between findings confirm the complexity of this condition, particularly the behavioural variant of the condition. Further qualitative research into this area would allow for a more detailed understanding of carer perceptions of their situation and the process of adjusting, and how the factors included in this review appear to interact with each other and with other variables, which have not been explored in quantitative research.

Conclusions

FTD is a relatively under investigated area, and research to date has highlighted differences between this and more commonly understood forms of dementia. FTD presents in younger adults, mainly males, with behavioural and/ or language deficits. Research has

indicated that this disease is particularly difficult for those family members caring for the person with FTD. The current review summarised 16 research studies which investigated the factors associated with psychological wellbeing in family caregivers (mainly spouses). The studies highlight a range of potential factors, both patient variables such as disease severity and caregiver variables such as coping style. It appears from the literature that the type of behavioural symptoms rather than purely the severity of the dementia were associated with poorer caregiver wellbeing, symptoms such as apathy, disinhibition and depression in the person with FTD were found to have an impact on the caregiver's wellbeing. The behavioural variant of FTD was more associated with poor wellbeing in caregivers than the other subtypes. The caregiver's perception of the quality of the relationship and their coping style were also factors in their wellbeing. There were some suggestions that time since diagnosis, age and gender were factors, however, these results varied and were, therefore, inconclusive.

There are clear clinical implications such as dementia services specialist care and support to these particular carers. Future research should also be aimed at finding out more about caregiver perceptions of the process of caring in FTD, as this is likely to impact on the care they are able to provide for the person with FTD. A systemic approach to care would enable both the person with dementia and their carer to receive the best support available, along with interventions specifically targeted to FTD such as behaviour management.

References

- American Psychiatric Association (2013). Diagnostic and statistical manual of mental disorders (5th ed.). Arlington, VA: Author.
- American Psychiatric Association. (2000). Diagnostic and statistical manual of mental disorders DSM-IV-TR fourth edition (text revision). Arlington, VA: Author.
- Armstrong, N., Schupf, N., Grafman, J., & Huey, E. (2013). Caregiver burden in frontotemporal degeneration and corticobasal syndrome. *Dementia and Geriatric Cognitive Disorders*, 36, 310-318. doi: 10.1159/000351670
- Boutoleau-Brettonnière, C., Vercelletto, M., Volteau, C., Renou, P., & Lamy, E. (2008). Zarit burden inventory and activities of daily living in the behavioral variant of frontotemporal dementia. *Dementia and Geriatric Cognitive Disorders*, 25, 272-277. doi:10.1159/000117394
- Campbell, P., Wright, J., Oyebode, J., Job, D., Crome, P., Bentham, P., ... & Lendon, C. (2008). Determinants of burden in those who care for someone with dementia. *International Journal of Geriatric Psychiatry*, 23, 1078-1085. doi: 10.1002/gps.2071
- Carers Trust (2013). *A road less rocky: Supporting carers of people with dementia*. London:Carers Trust.
- Carver, C. S. (1997). You want to measure coping but your protocol's too long: Consider the

brief cope. *International Journal of Behavioral Medicine*, 4, 92-100.

doi:10.1207/s15327558ijbm0401_6

Cosseddu, M., Alberici, A., Pelizzola, S., Padovani, A., & Borroni, B. (2013). The other face of the coin: The caregiver burden in frontotemporal lobar degeneration. *International Journal of Geriatric Psychiatry*, 28, 655-657. DOI: 10.1002/gps.3892

Crowe, M., & Sheppard, L. (2011). A general critical appraisal tool: An evaluation of construct validity. *International Journal of Nursing Studies*, 48, 1505-1516.

<http://dx.doi.org/10.1016/j.ijnurstu.2011.06.004>

Cummings, J. L., Mega, M., Gray, K., Rosenberg-Thompson, S., Carusi, D. A., & Gornbein,

J. (1994). The neuropsychiatric inventory comprehensive assessment of psychopathology in dementia. *Neurology*, 44, 2308-2308. doi: <http://dx.doi.org/10.1212/WNL.44.12.2308>

de Vugt, M. E., Riedijk, S. R., Aalten, P., Tibben, A., van Swieten, J. C., & Verhey, F. R.

(2006). Impact of behavioural problems on spousal caregivers: a comparison between Alzheimer's disease and frontotemporal dementia. *Dementia and Geriatric Cognitive Disorders*, 22, 35-41. doi:10.1159/000093102

Department of Health (2001) National Service Framework for Older People.

https://www.gov.uk/government/uploads/system/uploads/attachment_data/file/198033/National_Service_Framework_for_Older_People.pdf

Department of Health (2009) Living well with dementia: A national dementia strategy.

https://www.gov.uk/government/uploads/system/uploads/attachment_data/file/168220/dh_094051.pdf.

Department of Health. (2015). *The government's mandate to NHS England for 2016 –2017*.

<https://www.gov.uk/government/publications/nhs-mandate-2016-to-2017>

Diehl-Schmid, J., Schmidt, E. M., Nunnemann, S., Riedl, L., Kurz, A., Förstl, H., ... &

Cramer, B. (2013). Caregiver burden and needs in frontotemporal dementia. *Journal of Geriatric Psychiatry and Neurology*. doi: 10.1177/0891988713498467

Dodge, R., Daly, A. P., Huyton, J., & Sanders, L. D. (2012). The challenge of defining

wellbeing. *International Journal of Wellbeing*, 2. Retrieved from:

<http://www.internationaljournalofwellbeing.org/index.php/ijow/article/view/89>

Rother, E. T. (2007). Systematic literature review X narrative review. *Acta Paulista de*

Enfermagem, 20(2), v-vi. doi: <http://dx.doi.org/10.1590/S0103-21002007000200001>

Gélinas, I., Gauthier, L., McIntyre, M., & Gauthier, S. (1999). Development of a functional

measure for persons with Alzheimer's disease: the disability assessment for dementia. *American Journal of Occupational Therapy*, 53, 471-481.

doi:10.5014/ajot.53.5.471

Green, B. N., Johnson, C. D., & Adams, A. (2006). Writing narrative literature reviews for

peer-reviewed journals: secrets of the trade. *Journal of Chiropractic Medicine*, 5, 101–117. [http://doi.org/10.1016/S0899-3467\(07\)60142-6](http://doi.org/10.1016/S0899-3467(07)60142-6)

Guevara, A. B., Knutson, K. M., Wassermann, E. M., Pulaski, S., Grafman, J., & Krueger, F.

(2015). Theory of mind impairment in patients with behavioural variant fronto-temporal dementia (bv-FTD) increases caregiver burden. *Age and Ageing*, 44, 891-895. doi: 10.1093/ageing/afv059

Hsieh, S., Irish, M., Daveson, N., Hodges, J. R., & Piguet, O. (2013). When one loses empathy its effect on carers of patients with dementia. *Journal of Geriatric Psychiatry and Neurology*, doi: 0891988713495448.

Jurica, P. J., Leitten, C. L., & Mattis, S. (2001). DRS-2 Dementia rating scale
2. Psychological Assessment Resources Eds. Odessa: FI.

Knutson, K. M., Zamboni, G., Tierney, M. C., & Grafman, J. (2008). Neural correlates of caregiver burden in cortical basal syndrome and frontotemporal dementia. *Dementia and Geriatric Cognitive Disorders*, 26, 467-474. doi:10.1159/000167268

Massimo, L., Evans, L. K., & Benner, P. (2013). Caring for loved ones with frontotemporal degeneration: The lived experiences of spouses. *Geriatric Nursing*, 34, 302-306.
<http://dx.doi.org/10.1016/j.gerinurse.2013.05.001>

Mekala, S., Alladi, S., Chandrasekar, K., Fathima, S., MO'Connor, C., McKinnon, C., ... & Mioshi, E. (2013). Cultural differences are reflected in variables associated with carer burden in FTD: A comparison study between India and Australia. *Dementia & Neuropsychologia*, 7, 104-109. Retrieved from
https://www.researchgate.net/profile/Olivier_Piguet2/publication/236136171_Cultural_differences_are_reflected_in_variables_associated_with_carer_burden_in_FTD_A_comparison_study_between_India_and_Australia/links/004635164c372a47c7000000.pdf

Merrilees, J., & Ketelle, R. (2010). Advanced practice nursing: meeting the caregiving challenges for families of persons with frontotemporal dementia. *Clinical Nurse Specialist CNS*, 24(5), 245. <http://www.ncbi.nlm.nih.gov/pmc/articles/PMC2943420/>

Merrilees, J., Dowling, G. A., Hubbard, E., Mastick, J., Ketelle, R., & Miller, B. L. (2013).

Characterization of apathy in persons with frontotemporal dementia and the impact on family caregivers. *Alzheimer Disease and Associated Disorders*, 27, 62.

doi:10.1097/WAD.0b013e3182471c54

Merrilees, J., Hubbard, E., Mastick, J., Miller, B. L., & Dowling, G. A. (2014). Sleep in persons with frontotemporal dementia and their family caregivers. *Nursing Research*, 63, 129-136. doi: 10.1097/NNR.0000000000000024

Mioshi, E., Bristow, M., Cook, R., & Hodges, J. R. (2009). Factors underlying caregiver stress in frontotemporal dementia and Alzheimer's disease. *Dementia and Geriatric Cognitive Disorders*, 27, 76-81. doi:10.1159/000193626

Mioshi, E., Foxe, D., Leslie, F., Savage, S., Hsieh, S., Miller, L., ... & Piguet, O. (2013). The impact of dementia severity on caregiver burden in frontotemporal dementia and Alzheimer disease. *Alzheimer Disease & Associated Disorders*, 27, 68-73. doi:10.1097/WAD.0b013e318247a0bc

Mourik, J. C., Rosso, S. M., Niermeijer, M. F., Duivenvoorden, H. J., Van Swieten, J. C., & Tibben, A. (2004). Frontotemporal dementia: Behavioral symptoms and caregiver distress. *Dementia and Geriatric Cognitive Disorders*, 18, 299-306. doi:10.1159/000080123

National Institute for Health and Care Excellence (2006) NICE guidelines [CG42];

Dementia: Supporting people with dementia and their carers in health and social care. <https://www.nice.org.uk/guidance/cg42>

NHS England (2014) Best practice in Memory Services: Learning from across

England. <https://www.england.nhs.uk/mentalhealth/resources/dementia/>

Ory, M. G., Hoffman, R. R., Yee, J. L., Tennstedt, S., & Schulz, R. (1999). Prevalence and impact of caregiving: A detailed comparison between dementia and non-dementia caregivers. *The Gerontologist*, 39(2), 177-186. doi: 10.1093/geront/39.2.177

Oyebode, J. R., Bradley, P., & Allen, J. L. (2013). Relatives' experiences of frontal-variant frontotemporal dementia. *Qualitative Health Research*, 23(2), 156-166. doi:10.1177/1049732312466294

Prince, M., Knapp, M., Guerchet, M., McCrone, P., Prina, M., Comas-Herrera, A., & Salimkumar, D. (2014). *Dementia UK: Update*. London: Alzheimer's Society. https://www.alzheimers.org.uk/site/scripts/download_info.php?fileID=2323

Rahman (2014, January 4) Not just medication but Alzheimerisation.

<https://livingwelldementia.org/2014/01/04/not-just-medicalisation-but-alzheimerisation/>

Riedijk, S. R., De Vugt, M. E., Duivenvoorden, H. J., Niermeijer, M. F., Van Swieten, J. C., Verhey, F. R. J., & Tibben, A. (2006). Caregiver burden, health-related quality of life and coping in dementia caregivers: a comparison of frontotemporal dementia and Alzheimer's disease. *Dementia and Geriatric Cognitive Disorders*, 22, 405-412. doi:10.1159/000095750

Riedijk, S., Duivenvoorden, H., Van Swieten, J., Niermeijer, M., & Tibben, A. (2009). Sense of competence in a Dutch sample of informal caregivers of frontotemporal dementia patients. *Dementia and Geriatric Cognitive Disorders*, 27, 337-343. doi:10.1159/000207447

Robinson, B. C. (1983). Validation of a caregiver strain index. *Journal of Gerontology*, 38,

344-348. doi: 10.1093/geronj/38.3.344

Roche, L. D., Croot, K., MacCann, C., Cramer, B., & Diehl-Schmid, J. (2015). The role of coping strategies in psychological outcomes for frontotemporal dementia caregivers. *Journal of Geriatric Psychiatry and Neurology*, 28, 218-228. doi:10.1177/0891988715588830

Royal College of Psychiatrists (2015) Memory Services National Accreditation Programme (MSNAP): Standards for Memory Services.

<http://www.rcpsych.ac.uk/PDF/MSNAP%20standards%205th%20edition.pdf>

Su, X., Lau, J. T., Mak, W. W., Choi, K., Feng, T., Chen, X., ... Cheng, J. (2015). A preliminary validation of the Brief COPE instrument for assessing coping strategies among people living with HIV in China. *Infectious Diseases of Poverty*, 4, 41. <http://doi.org/10.1186/s40249-015-0074-9>

Wear, H. J., Wedderburn, C. J., Mioshi, E., Williams-Gray, C. H., Mason, S. L., Barker, R. A., & Hodges, J. R. (2008). The Cambridge behavioural inventory revised. *Dementia and Neuropsychologia*, 2(2), 102-107. <http://www.demneuropsych.com.br/imageBank/PDF/dnv02n02a04.pdf>

Wong, C. C., & Wallhagen, M. I. (2012). Family caregivers of individuals with frontotemporal dementia: Examining the relationship between coping and caregiver physical and mental health. *Journal of Gerontological Nursing*, 40, 30-40. doi: 10.3928/00989134-20130827-05

Zarit, S., Orr, N. K., & Zarit, J. M. (1985). *The hidden victims of Alzheimer's disease: Families under stress*. NYU Press.

Section B: Theorising the Process of Adjustment in Family Carers of Those with Behavioural
Variant Frontotemporal Dementia.

Adjusted Word Count: 9230

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Abstract

Background: Behavioural variant frontotemporal dementia manifests in personality and behavioural changes. This study aimed to explore carer's perceptions of the condition, with an emphasis on how these carers learn to adjust and accept their situation.

Design: Grounded theory methodology was used to analyse 12 semi-structured interviews.

Results: A theoretical model is proposed to describe the carer journey and adjustment process. The early stages in the process are significant, often a time of confusion and difficult emotional responses. With time carers begin to find helpful strategies in managing.

Conclusion: There is a continuous process of adjustment for these carers. These findings add to the existing research base by considering different stages of the adjustment process for a better understanding of the experiences.

Key words: frontotemporal dementia, carers, spouses, adjustment, process.

Introduction

Frontotemporal Dementia

Frontotemporal dementia (FTD) is a rare form of the progressive condition and is thought to account for only 2% of dementia cases in the UK (Alzheimer's Society, 2016). There are approximately 16,000 people in the UK with a diagnosis of FTD (NHS Choices, 2015). Unlike other forms of dementia, FTD is predominantly characterised by symptoms relating to behavioural, affective and language deterioration. These symptoms are caused by atrophy in the brain, predominantly within the frontal and/ or anterior temporal lobes (Englund et al., 1994). There are three subtypes of FTD; behavioural variant (bv-FTD), semantic dementia and progressive non-fluent aphasia. The latter two refer to predominantly language impairment, whereas bv-FTD refers to changes in behaviour, affect and personality.

FTD in the Healthcare Context

Memory clinics were introduced in the UK as a means of improving dementia diagnosis rates, which has been part of a government initiative (Department of Health, 2015). The American Psychiatric Association (APA) recently updated their diagnostic criteria for dementia. Prior to this change, dementia was characterised by decline in two areas of cognitive functioning, one of which had to be memory impairment as well as a reduction in functioning in daily living (APA, 2000). Cognitive decline does now not have to include memory (APA, 2013).

Despite these changes, early presentations of FTD may not always fit a cognitive criteria and are often more associated with personality and behaviour change than the more accepted memory decline seen in Alzheimer's disease (Lindau et al, 2000). Therefore, referral to a memory clinic may not always be the obvious choice. Many of those with FTD

are seen by mental health practitioners and misdiagnosis is often common (Mendez, Shapira, McMurtray, Licht & Miller, 2007).

The Royal College of Psychiatrists (RCP) introduced national standards for memory clinics (MSNAP). In the latest edition of the guidance FTD is mentioned only once, however on the final page of the document. Here it briefly mentioned rarer forms of dementia and recommends neurological input (RCP, 2015).

Post diagnostic support is recommended by MSNAP (RCP, 2015) and the National Institute of Health and Care Excellence (NICE, 2006). In practice there is no clear guidance as to what this involves, often including groups such as cognitive stimulation and reminiscence; post diagnostic guidance for carers including memory strategies; and more practical support such as applying for lasting power of attorney. However, this may not be appropriate for FTD patients and carers. Additionally, there are medications prescribed in early Alzheimer's disease which allow the patient and their carer to be followed up for medication review. As this is not the case for FTD, this results in discharge from the memory service. Memory clinics within the National Health Service are often linked with older adults community mental health teams. Since FTD is often diagnosed as a young onset dementia, there currently appears to be a lack of specific services to support this younger population, therefore it is unclear which service would be appropriate for this group.

Carer burden and distress in FTD

Research on caregiver burden in dementia indicates those caring for someone with a diagnosis of FTD experience higher need (Nicolaou, Egan, Gasson & Kane, 2010), levels of stress and depression (Mioshi, Bristow, Cook & Hodges, 2009) and levels of burden (Riedijk et al., 2006) than carers of those with a diagnosis of Alzheimer's type dementia.

Various factors are thought to account for these findings, including the complex presentation, loss of personality and affect in the person diagnosed as well as the younger onset of this form of dementia (Englund et al., 1994). Wong and Wallhagen (2012) investigated how these presentations affect carers. They concluded that carer distress was particularly associated with symptoms of apathy, indifference and loss of insight in the person cared for. Diehl-Schmid et al. (2013) indicated that changes in personality were the most burdensome for carers. Other studies have found that carer variables such as gender account for higher levels of stress (Mioshi et al., 2009). Interestingly, despite the progressive nature of the condition, levels of burden for this group of carers have been shown to decrease or remain stable (Riedijk et al., 2008). These findings would imply that some process of adjustment to the carer's experiences occurs.

Experiences of FTD carers

In relation to the above, it appears necessary to gain more of an understanding of the carer's actual experiences of FTD. Although limited to date, some qualitative research has been carried out to further explore the experiences of these carers.

Kumamoto et al. (2004) used data from the family carers of two people with FTD. They reported themes emerging relating to behavioural symptoms common in FTD, such as over eating, distractibility and impulsivity, which make the carer's role difficult with regards to carrying out day to day activities. Additionally, Massimo, Evans and Benner (2013) interviewed two spousal caregivers. They described difficulties in managing their situation related to accepting the new role of being a carer; feeling isolated; experiencing anger in relation to some of the common FTD symptoms such as lack of empathy; and adjusting from a position of blame to one of understanding the behaviours in the context of FTD.

Oyebode, Bradley and Allen (2013) describe similar findings from interviews with six carers, such as adapting to their new role as a carer and the impact this has on their relationship with the person with FTD. They also discuss particular challenges in relation to FTD symptoms such as personality and behavioural changes, lack of empathy and socially inappropriate behaviour. Additionally they provide some context to the situation and highlight difficulties participants have experienced with health care professionals and the route to diagnosis. They begin to present information regarding coming to terms with the changes and strategies the carers have adapted in order to manage, such as supervising the person with FTD and anticipating behaviours. Similarly, Kindell, Sage, Wilkinson and Keady (2014) examined qualitative experiences of one family of a person with semantic dementia, finding that the emerging themes related to strategies and approaches to the experience. This family described the use of routines; policing (which is similar to supervising described by Oyebode et al, 2013); finding new ways of communicating; and being adaptive in relation to the changes in the person with SD's likes and dislikes.

Models of adjustment in dementia

Both quantitative and qualitative research described above has begun to imply that there is an adjustment process occurring for the family carers of people with FTD. There has been some attempt to link existing models of adjustment to dementia. Theories of loss have been linked with the experience of living with dementia. The dual process model (Stroebe & Schut, 2010) of grief has been applied to the process of adjustment in dementia care. This model proposed that in bereavement an individual oscillates between 'loss-orientation', processing the loss of a loved one, and 'restoration-orientation', focussing on what remains and adapting to life without the loved one. Robinson, Clare and Evans (2005) highlight that in dementia, it is important to consider a 'family systems perspective' which uses the couple's experiences and perceptions of adjusting rather than purely the carer alone. They

adapted the dual process model to include adjustment in couples to a diagnosis of dementia. Similarly, Merrick, Camic and O'Shaughnessy (2013) also investigated adjustment in dementia from a relational perspective. From interviews with couples, they concluded that the person with dementia tended to be able to reflect on their situation and empathise with the carer. This allowed the couple to process their circumstances as a couple. They also found that the person with dementia was able to speak about past experiences, therefore enabling the oscillation process between what was loss and restoration. This work had implications for clinical practice and the authors recommend a solution-focused approach to healthcare in order to enable resilience. The ability to reflect by the person with dementia may also support the use of reminiscence type approaches in which the family can spend time together talking about shared experiences from the past.

Whilst in agreement that there is a process of grief in caring for a family member with dementia, Silverberg (2007) reflects on personal experience and suggests that healthcare is based on practical approaches but does not support the grieving process. She suggests further ongoing psychosocial support to carers to normalise and process the grief, and to encourage the use of respite.

The above literature has mainly focused on Alzheimer's disease and vascular dementia, although Merrick et al (2013) include one couple with FTD. It is suggested that due to the unusual presentation in FTD, such as personality and behavioural changes, that this process may differ. Given common difficulties related to lack of empathy, carers of those with FTD may experience feeling more isolated and less understood by the person with FTD. Additionally, as services tend to be set up with memory difficulties in mind, how helpful they are for this different presentation, for example a reminiscence group where the individual with FTD has socially inappropriate behaviours, is questionable. There is little research

linking models of adjustment specifically to the experience of those caring for a family member with FTD.

Rationale for current research

There is a small research base relating to the qualitative exploration of carer experiences of FTD. Although these produced themes, they are currently distinct themes that are not interlinked. Whilst most of the literature to date has focused purely on the problems faced by these carers, recent research by Oyebode et al. (2013) and Kindell et al. (2014) have reported a process of adjustment for these carers, yet theoretical models have not been applied. Therefore this study intends to add to the limited research base and aims to tie together some of these themes in order to create a theoretical model of adjustment for this set of carers. Since literature indicates that the most burdensome and distressing symptoms of FTD are behavioural, this study was focused only on bv-FTD.

Research Questions

- a. What are participant's perceptions of caring for a person diagnosed with bv-FTD?
- b. How do participants explain their responses to the diagnosis?
- c. What are carer perceptions of the process of coming to terms with the condition?
- d. How do participants describe their ways of managing their circumstances?

Methodology

Design

A non-experimental qualitative design was used in the study. Semi-structured interviews were carried out (see appendix C for interview schedule). Constructivist grounded theory methodology was adopted, as described by Charmaz (2014). As recommended by Charmaz (2014) the interviews were not kept strictly to the initial schedule, some participants required little prompting and were able to share their experiences, whereas others found it easier to answer the specific questions. This interview schedule was also adapted as the data collection process continued, in order to check out emerging themes.

All interviews were conducted face to face, and all, bar one, were one to one. In one interview, the daughter of the interviewee was also present and contributed to the discussion. Interviews were carried out at a place which was convenient for the participant; most were at the participant's home, one was in a café and another at the participant's place of work.

Participants

In this study 12 participants were interviewed. Participants were recruited via a national support network for FTD. A presentation was given by the researcher at one of the group meetings, and an advertisement of the study was included in the network newsletter along with the researcher's contact details. Participants were asked to make contact if they were interested.

Of the 12 participants, only two were male. All of those interviewed were a spouse of the person with a diagnosis of bv-FTD. Ten participants lived with their partner, the partners of the other two participants had passed away not more than six months prior to the study. All the participants were of a white British or white Irish ethnicity and their ages ranged from 61 to 68. Participants were located in various parts of England, some in central London,

others in more rural localities. The diagnoses were received between 6 months and 10 years prior to the interview. See table 3 for demographic information.

Participant code	Age	Gender	Relationship to person with FTD	Time since diagnosis	Ethnicity
P1	66	Female	Wife	10 years	White British
P2	68	Female	Wife	4 years	White British
P3	63	Female	Wife	5 years	White Irish
P4	61	Female	Wife	4 years	White British
P5	68	Female	Wife	2 years	White British
P6	65	Male	Husband	6 months	White British
P7	68	Female	Wife	18 months	White British
P8	65	Female	Wife	2 years	White British
P9	67	Female	Wife	3 years	White British
P10	61	Female	Wife	6 years	White British
P11	67	Male	Husband	1 year	White British
P12	69	Female	Wife	5 years	White British

Table 3 Participant Demographic Information

Procedure

Prior to data collection a pilot interview was carried out with a member of the FTD support group used for recruitment whose partner had passed away some years before. As a result of this interview the interview schedule was cut down significantly as the interview ran over 2.5 hours duration. It was clear during the interview that a lot of the questions were being covered during conversation, therefore less emphasis was placed on the interview questions themselves and instead more follow up from participant comments was prioritised. One of

the questions referred to carer perceptions of the future, it was fed back from the pilot interview that this may not be a helpful question and may cause unnecessary distress, therefore this question was dropped and only asked if the carers had made reference to the future in conversation. Once a participant had made contact, they were sent the information sheet (see appendix D) and a consent form (see appendix E). Participants were asked to read over the information and contact the researcher should they still be interested. If there was no response, it was assumed they no longer wished to participate and were not contacted. Those who were still interested at this point were contacted to arrange a suitable time and place for their interview.

On the day of the interview, participants were given another copy of the information sheet and asked if they had any questions. The researcher then highlighted certain aspects of the information sheet regarding confidentiality, the possible emotional effects of the interview and ensured the participants knew they could terminate the interview at any point should they wish to do so. They were then given a consent form and asked to sign this prior to the interview.

Once the participants had given their consent, the interview was carried out. As mentioned above, the full interview schedule was not always followed completely depending on the participant's responses. Interview duration ranged from 38 minutes to 126 minutes. Interviews were recorded and were transcribed by the researcher. Following the interview, participants were asked how they were feeling and were debriefed.

Quality Assurance

Guidelines were used to maximise quality throughout this study (Mays & Pope, 2000). A bracketing interview was completed prior to any data collection to explore the researcher's assumptions at this early stage, no changes were necessarily made as a result of

this interview since the aim was to consider how these assumptions may influence the interpretation of the data (see appendix F for list of key assumptions discussed). It was clear that the researchers previous knowledge of memory clinics may influence some of the questions, therefore a more curious approach to the development of questions was adopted. A reflective account was also written by the researcher throughout the study, which allowed the researcher to continue to notice assumptions (see appendix G for an extract of this account). There were points at which assumptions were made relating to the meaning of the content which were clearly influenced by the researchers own beliefs and experience. This was acknowledged during the process and checked out in further interviews, for example, whether the carers felt listened to.

Using the grounded theory method of ‘constant comparison’ (Corbin & Strauss, 2008) allowed for a curious approach to be taken towards the data. This meant analysing interviews during the data collection, which continued to inform further interviews and encouraged the researcher to continuously check out emerging themes with participants. This allowed for saturation of the data, as further interviews were used to check out emerging themes, the results of which immediately influenced the emerging themes and connections between themes. The researcher also arranged meetings with their supervisor to discuss emerging themes, findings and how these fit together. This led to a shift in the visual representation of the model, as well as adaptations to terminology throughout. A peer of the researcher was also asked to check the emerging codes against an anonymised extract of the raw data. This did not result in any changes in the data.

Additionally, the researcher carried out ‘member checking’ (Saldana, 2015) at certain points throughout the study in order to check emerging themes with the participants. After drawing out the categories and themes, the researcher reviewed the interviews to ensure these

themes fit with the raw data. Again, this allowed for saturation of the data as no further themes were emerging from the final interviews.

Ethical Considerations

This study received ethical approval from the NRES committee London - Fulham (see appendix I for approval letter). The BPS code of conduct (2010) was also followed.

Data Analysis

Line by line in vivo coding was administered initially; these participant quotes were then used to form categories. Axial coding was then used to further develop categories and themes that seemed to capture the raw data. See appendix J for an example transcript with codes. This allowed for the data from the various interviews to be combined and links between the categories to begin to emerge. The method of ‘constant comparison’ was adopted, with data collection and analysis being carried out simultaneously in the research journey. Throughout the process, memo writing and diagramming were an important part of developing the theory.

Results

Analysis of the 12 interviews resulted in the development of 5 core components, subcomponents were also identified. See table 4 for a more detailed breakdown of each component. The main components identified were as follows:

1. Life before FTD
2. Characteristics of FTD
3. Carer journey – The road to FTD diagnosis

4. Carer journey - 'Living Bereavement', Coming to terms with the diagnosis of FTD.
5. Carer journey – Coping with FTD.

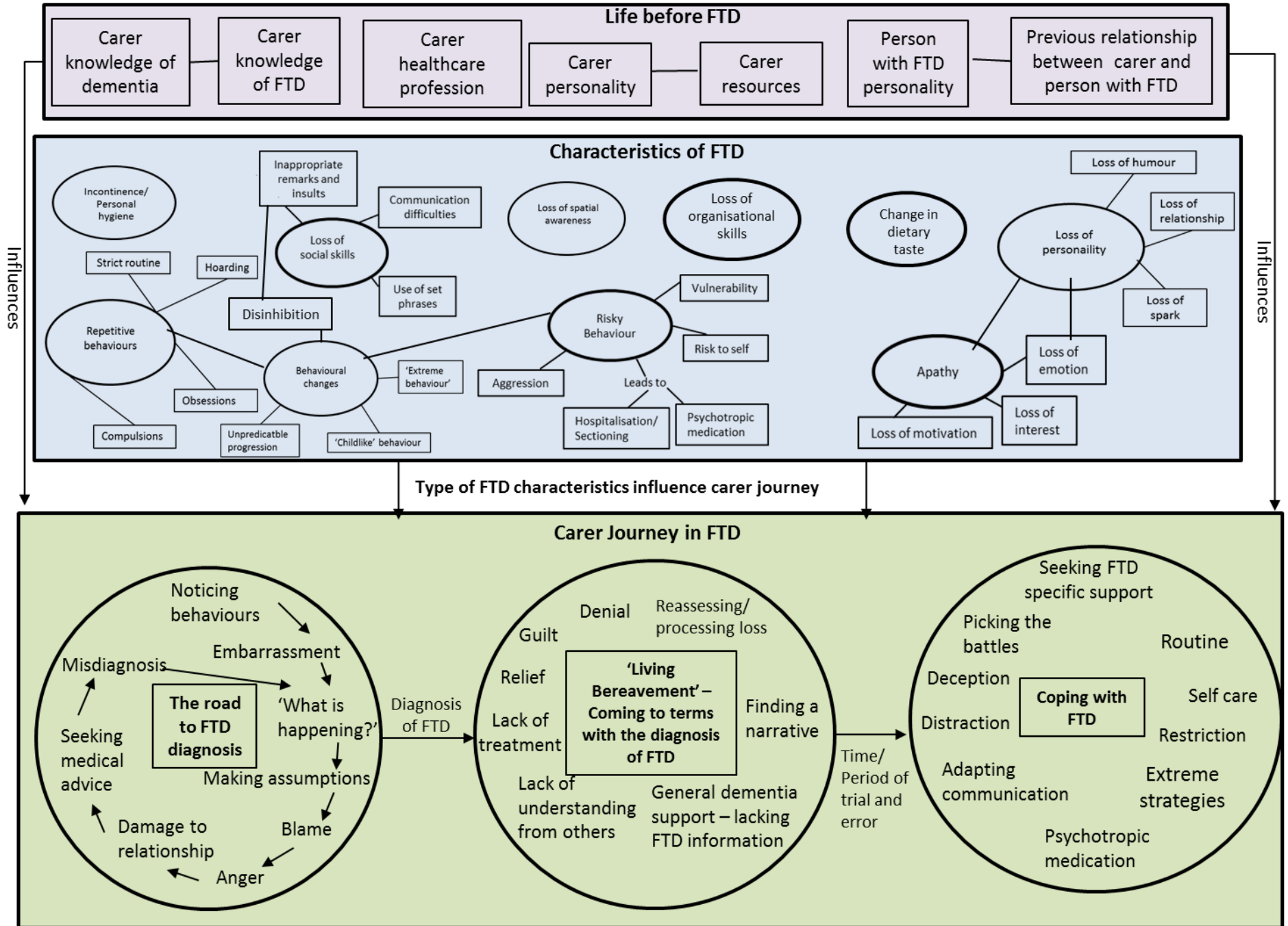
A theoretical model was developed to describe how these separate categories interlink. A visual representation of the theoretical model is presented in figure 1. This model and its components are discussed below with example quotations from the interviews (see appendix H for further example quotations for each category).

Category 1: Life before FTD

The aim of the research was to focus on carer life since the diagnosis of bv-FTD. In the interviews, it appeared important to understand elements of the carer's past in order to understand their perceptions of bv-FTD and their current situation. Therefore, the overall model of adjustment in bv-FTD does not exist solely within the context of the disease itself but is shaped by the people affected, their personalities, knowledge, resources and relationships prior to the onset of the condition.

Given the unusual presentation of bv-FTD, carers reported attributing the noticed changes down to personal factors in the first instance. However, those with a previous knowledge of

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dementia or FTD were able to query a disease process or dementia as a possibility. Whilst not all participants had experience of dementia, some participants reported using links with friends with this knowledge to explore the 'making assumptions' stage. In this early stage, 'road to FTD diagnosis' may include less blame and damage to the relationship in these cases. However, this did not always result in a quicker diagnosis of bv-FTD.

'The first time I thought of any type of dementia was...when [friend] said to me 'you do realise [husband] is in the first stages of dementia'and I went to the doctor....and he said 'no, he can still play bridge, his short term memory is excellent, he does not have dementia'.' (P8).

Additionally, experience of dementia appeared to impact on the carer views of the future.

For example one participant said;

'My children would say 'well you know what dad is going to do next, remember what grandma used to do'.' (P1)

Additionally, those participants who reported previous experience in healthcare settings reported feeling as though they were able to be more assertive with professionals in order to ask for help from services and manage day to day life in general. A retired nurse stated;

'It's alright for me, I have inside information. I have experience of the medical world, so I know more or less, but even I can become a cropper.....There's no way if I didn't know what I know that we could handle this here.' (P3)

The reported symptoms of bv-FTD as well as carer perceptions of relationship difficulties were also impacted by their past experiences. One participant described how he and his wife were always independent but, due to the condition, she had become dependent.

'...in a sense that was an early symbol because in the past she had wanted to do that herself but she wanted me to be with her.' (P11)

The previous relationship between the carer and the person with bv-FTD and the personality of the person with the diagnosis prior to the onset of the condition is particularly significant given the commonly reported symptoms of loss of personality and apathy. The previous relationship is impacted upon and a new relationship and roles within the relationship are to be adjusted to.

'There wasn't any affection' (P6)

'We used to have great conversations, we don't now.....It's not mates anymore' (P8)

One carer explicitly made this link between the past experiences of relationships and how the current situation may be perceived.

"It depends where the relationship starts as to how it's going to change" (P2)

Carers found themselves drawing on their existing knowledge, personality and resources prior to the onset of the condition in the management of the condition.

"I'm intelligent...I find it easier to cope with organisations" (P11)

"I'm the eldest of 8, so I'm used to having to fix it from all angles and sides" (P3)

Category 2: Characteristics of bv-FTD

It was evident within the interviews that it is difficult to understand the carer journey without first understanding the complexity of the condition itself. Participants described details of bv-FTD symptoms as a way of portraying the difficult nature of caring for someone with this condition. This is indicated in the model presented, the carer model is very much influenced by the presenting symptoms.

Apathy

Apathy was commonly reported in the research interviews. This was perceived as not caring, leading to blame towards the person with FTD and resulting in further strain on the relationship, therefore illustrating the impact of the type of symptoms on the carer journey itself.

“He is entirely apathetic...he just can't be bothered, it's quite frustrating” (P2)

Personality Change/Loss

Similarly, another commonly reported symptom is personality change and/or loss and was often noticed at early stages. Since this is unusual and varies from other types of dementia in the early stages, it is often initially attributed to personal factors rather than a disease process. Therefore, this impacts on the ‘route to diagnosis’ and ‘living bereavement’ stages as the carer may not seek medical support initially and as mentioned above is required to come to terms with the loss of the person and their relationship.

“The one longstanding thing for me is the brightness of his eye, that went and has never come back” P12

“It is like having [name] here but not here, if you know what I mean” P9

Participants described a loss of emotional response in their loved ones, which was often linked with distress for the carer.

“He doesn't have sympathy or empathy” P11.

One participant described how this differs from other forms of dementia.

“At least with Alzheimer's, from my experience, often you get somebody who their memory might be atrocious, but they are still loving” P5.

This loss of personality is also a factor in why the condition is difficult to diagnose, as many of the early signs are subtle changes that only those closest to the person would notice. This links to the common experience of misdiagnosis discussed below.

Behavioural Changes

Behavioural changes are characteristic of bv-FTD and appeared to impact on the journey for the carer. Behaviour was described as ‘extreme’ and ‘childlike’ with unpredictable progression.

“He just doesn’t know how to behave...he told an old man with 2 grandchildren.....” P7

Disinhibition was common and would lead to embarrassment for the carer.

“He just says whatever comes into his head...A lot of sexual comments have been made” P12

Social situations were significantly affected by these behaviours, therefore limiting opportunities for social activities.

“He could say awful things to people” (P12)

“You can’t go to a restaurant with someone with this [FTD]” (P2)

Participants often perceived negative views by others with regards to these behaviours.

“He intimidates people” (P7)

“We are still having an issue with other people being around him and not just me. He looks very...it kind of scares people” (P3).

The behavioural symptoms, particularly more extreme behaviours increase risk. Participants reported vulnerability and risk to self and others, but more commonly reported aggressive behaviour by the person with bv-FTD.

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“I had him screaming that he was going to kill me” (P1)

“He threatened to punch an old lady” (P12)

“The children can’t be with him on their own, if there is an emergency he hasn’t a clue what to do” (P4).

In many cases these behaviours have resulted in police involvement, hospitalisation and the use of psychotropic medication. These unusual behaviours can often be the reason for the delay of a diagnosis of bv-FTD and recurrent misdiagnoses.

The behavioural symptoms were described by carers as unpredictable.

“Every day you wake up in fear, because you do not know what is going to happen today” (P12).

Participants also reported the repetitive behaviours.

“One of his compulsions has been dressing and undressing. It would go on for 2 hours” P12

The participants which reported this behaviour also reported routine as an extremely helpful strategy. Therefore, the type of strategies the carers adopt to help them cope are dependent on the presenting symptoms of the condition.

Reduction in social skills

In addition to disinhibition in relation to social skills, communication skills were also reported as affected, particularly with regards to loss of language and the adoption of set phrases.

“You can’t sustain a conversation with [husband] anymore” (P8)

“I could write the sentences he uses on a postcard” (P7)

Other cognitive symptoms

Other cognitive symptoms reported by participants were related to loss of organisational skills, slowed information processing and loss of spatial awareness. These changes have an impact on the role of the carer within the relationship, as they reported now finding themselves responsible for household tasks their spouse would previously have handled or make joint day to day tasks more of an effort.

“I had screamed several times when we’d be driving, all of that sort of decision making had gone” (P1)

“If you have to react quickly, that’s when he gets really slow, like slow motion” (P4)

“He bumps into you the whole time...whereas before we just sort of work around each other” (P4).

Category 3: Carer Journey – the road to diagnosis

The period from noticing initial changes to receiving a diagnosis of bv-FTD was particularly relevant for the participants interviewed, and illustrates the complexity of this form of dementia. The sections above relating to presenting symptoms and past experiences impact on this stage. Participants described the difficult period prior to any medical investigation in which they were first noticed changes in their loved one’s presentation and behaviours. The duration of these early symptoms prior to the diagnosis varied, but were often some years before any investigation or diagnosis was received.

“Looking back it was 10 years of change” (P7)

During this period, participants report difficult feelings such as embarrassment – often related to behavioural symptoms such as disinhibition. Following a period of bewilderment,

participants began to make their own assumptions as to the potential causes of the behaviours. A minority of participants had some understanding of this being a condition such as dementia, however, due to unusual presentation of the condition, others reported not understanding what was happening and therefore not attributing the changes to a medical condition. As described above, this was influenced by the sort of symptoms being noticed as well as carer experience and knowledge.

“I still didn’t understand that this was illness” (P5)

“I put it all down to alcohol” (P7)

This pre-diagnosis period of symptoms resulted in a range of carer responses both towards their loved one and in general. Some participants reported blaming their loved one and becoming frustrated with the behaviours, others reported this early experience as impacting significantly on their own mood. This was often due to the carer not understanding that the presentation of their loved one was down to a medical condition.

“I was getting very angry with him” (P12)

“It was before he was diagnosed that I was on that antidepressant” (P10)

This period often began to have a significant impact on the relationship often due to symptoms such as personality change and lack of empathy and the carer attributing these to personal factors rather than a disease process.

“There were a lot of rows before [the diagnosis]” (P7)

“I did have a lover for a while, before the diagnosis” (P7)

Therefore, the damage to the relationship and emotional impact was evident for these carers often long before any medical advice was sought. Some carers were initially reluctant to seek medical advice due to the unusual presentation of the condition.

“You can’t take your husband to a doctor and say this man doesn’t love me anymore”. (P5)

Once medical advice was sought, a common experience amongst the participants was misdiagnosis, in some cases on multiple occasions. In some cases no diagnosis was given initially and the symptoms were put down to personal causes such as relationship difficulties, despite these difficulties perhaps being a result of earlier FTD related changes.

“We were told to go to marriage counselling” (P2)

In other cases, initial mental health diagnoses were given.

“They would say [husband] was just playing up, it was depression” (P12)

Once the diagnosis of bv-FTD was received, it is proposed that the carers then move into the next stage of the process ‘Living Bereavement’.

Category 4 Carer Journey – ‘Living Bereavement’ Coming to terms with the diagnosis of BV-FTD

Following the diagnosis, the carers begin coming to terms with it. This was described by one participant as a ‘living bereavement’. Early descriptions of feelings following diagnosis for most participants were related to relief that the symptoms they had been reporting were finally confirmed.

“It meant there was a reason for all this awful behaviour” (P9)

“I’m glad it wasn’t just me being paranoid” (P5)

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Participants also reported a change in their perceptions of the presenting symptoms and for some this drew up feelings of guilt related to blaming their spouse for their presentation in the initial stages.

“....we got the diagnosis and my attitude changed....I felt more kindly towards him, because it is not his fault” (P7)

“I became a really awful person towards him, just to get a reaction for him, I didn't understand this illness” (P5)

As with other long term conditions, participants were required to reassess their role, relationship and expectations for the future.

“It is hard when you've had a partner for years and now you are looking after their side of the decision making” (P4)

“I can't help but feel angry at the life we have lost” (P11).

Unlike other forms of dementia, there is no prescribed medication for FTD. Therefore, in Alzheimer's type dementia for example, the diagnosis may open up options for medication and other forms of support. One participant stated

“I would say don't get the diagnosis, why bother? There is no medication” (P8)

Similarly participants described a lack of support being offered to them following the diagnosis, particularly specific support relating to FTD. Carers reported receiving generic dementia support but felt that this was not always relevant to their circumstances.

“We go to dementia suppers, dementia lunches, of course they are all Alzheimer's, nobody like him at all” (P5)

Participants described a general lack of understanding of the condition, even within services.

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“His brother will have nothing to do with him, they just find him so unpleasant” (P7)

In one carer’s experience, a local dementia service refused to work with her husband because of his behavioural symptoms.

“They dropped him because he is too much for them” (P9)

For some carers this led to a feeling of isolation. Other carers struggled to find a narrative to describe their situation and were required to develop ways of discussing the condition with others.

“If I had a pound for every time somebody said ‘oh my grandmother had Alzheimer’s...sometimes I just say ‘well it’s a different disease’” (P4)

“I had these little cards to show her what was wrong with him” (P9)

Category 5: Carer Journey – Learning to cope with FTD

With time and some trial and error, participants described developing strategies to help them cope with their situation.

Seeking FTD support

Of importance in the interviews was seeking support specifically related to FTD.

“I joined the FTD forum, that was my lifeline mainly” (P6)

“That’s [meeting another FTD carer] been incredibly useful because his wife had almost exactly the same thing so he could tell me many things” (P11)

Pick your battles

Participants described confronting behaviours in the early stages, yet after a period of adjustment began to prioritise which behaviours to confront and which to let go.

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“He had decided to lie in bed with his shoes on, I said it’s not worth having a fight to take them off” (P1)

Taking time out

Participants described caring in bv-FTD as ‘exhausting’, ‘draining’ and ‘full on’, often in relation to behavioural symptoms, therefore they reported that time out is essential.

“I would pray that she would not wake up and I would have a bit more time on my own” (P6)

Routine

Routine was commonly described as a helpful strategy.

“Repetitiveness is part of the disease, so he responds well to repetitive behaviour” (P5)

Deception

Participants also described the use of deception as a strategy they use.

“My son was shaving his father, to save a fuss, this would be under the guise of learning how to shave” (P1)

Boundaries

Participants described putting boundaries in place to manage the difficult behaviours.

“You have to say ‘no’ a lot of times” (P12)

Others described the practical use of boundaries.

“[Husband] went through a stage of eating nonstop so I put locks on the fridges” (P8)

Distraction

One participant described distracting his wife as helpful in stopping aggressive behaviour.

“I found one of the things which distracted her was to sing a song. The singing would distract her from digging her nails in” (P6)

Communication

The most commonly reported strategy related to adapting communication style with the person with bv-FTD, yet the approaches varied. Carers generally reported that clear and concise language was favourable, with less questions and options. Some carers described taking an assertive approach.

“I’m quite bossy with him” (P4)

While others found that instructions were not helpful.

“Directing her to do something wasn’t effective, it had to be very much one of encouragement” (P6)

One carer described using communication to resolve difficult situations.

“If things are a problem, all I have to do is say something really stupid or childish, just to get him to laugh” (P5)

Timing was also considered important.

“I have to filter information. I couldn’t say tomorrow we are going to do such a thing, otherwise at midnight he’d be up, and at 1, and at 2...so now I have to stop saying we are doing such a thing until maybe half an hour before we do it” (P3)

Caring in FTD

Another participant described how it is not necessarily the content of what is said but how it is said that is important.

“He responds a lot to the tone of voice, so the way you say things is much more important [than before]” (P8).

Managing risk

Carers reported using extreme measures in order to manage risky behaviours displayed by their loved ones, such as police involvement and the use of prescribed psychotropic medication.

As the FTD progresses, new symptoms emerge. As mentioned earlier, these symptoms are often described as unpredictable. Therefore, the development of strategies and learning to cope with FTD is a continuous battle.

Discussion

The aim of the study was to explore the experiences of those caring for a relative with bv-FTD in relation to their process of adjustment and coping. Twelve interviews were completed with spousal caregivers. Whilst it was anticipated that the content of the interviews would focus on time since the diagnosis, it was clear during the interviews that this process could only be understood in the context of the person’s history and the presentation of the FTD symptoms. A model is presented to consider five core categories and how these categories interlink in relation to the carer’s journey of adjustment.

The participants spoke about their past personality traits and experiences; their relationship with their spouse; and their spouses premorbid personality. These factors influenced their perceptions of the early symptoms and the health system, for example prior experience as a nurse enabled more assertive communication with professionals. These

previous experiences also shaped the impact of the change in their partner and their relationship and influenced their emotional responses to these changes. Therefore, this process is individual, the model presented uses common factors to understand the process.

Additionally, it was clear that the type of symptoms specific to FTD influenced the carer process. This is in keeping with the literature, which describes particular FTD-related symptoms to be associated factors in carer distress, burden and poor wellbeing (Armstrong, Schupf, Grafman & Huey, 2013). An example from this research is lack of empathy in the person with bv-FTD was described as difficult to understand and often led to the carer attributing blame and feeling frustrated towards their spouse; these feelings were also described in Massimo et al., (2013). The range of symptoms also made it difficult to understand the changes as a medical condition. This may be particularly relevant in FTD than in the more common Alzheimer's dementia in which memory decline is well understood.

The road to diagnosis was often complicated, as highlighted in previous research by Oyeboode et al, 2013. In keeping with the literature, participants reported a lack of understanding by professionals leading to feeling isolated and misdiagnoses leading to confusion and further emotional impact. Mendez et al. (2007) also described the common misdiagnoses in FTD including mental health diagnoses and Alzheimer's disease. This period prior to diagnosis seems particularly significant with regards to the impact of the condition on the relationship of the spouse. This research indicated that the carer perception of the relationship as controlling or less caring impacted on carer wellbeing in SD.

Once a diagnosis of bv-FTD was received this too led to a complexity of emotions, including guilt (relating to earlier blame and anger) and relief (of finally knowing what was happening). During this time, the carers are making sense of the diagnosis; finding a

narrative (a way of telling others); attempting to engage in dementia support, which was often described as inappropriate to their needs; and making sense of the change in their relationship and role as a carer (this is also documented in Oyebode et al., 2013). This stage was described as ‘living bereavement’ and there are commonalities with grief models such as the dual process model in which an individual oscillates between acknowledging loss and focussing on what remains, as presented by Stroebe and Schut (1999). As described earlier, this model has been applied to dementia care (Robinson et al., 2005; Merrick et al., 2013), in particular with regards to a relational process in which the person with dementia can reflect and empathise with the carer. This may not fit as well to FTD presentation, given that there is a lack of empathy commonly reported. The change in personality also means that the couple may now not share the same interests in order to reflect. Apathy may also be of significance in that the person with dementia may come across as not engaging or interested. In addition, carers have described communication difficulties, and a lack of conversational skills. One woman described showing her husband a clip of a film he was previous keen on, but he did not show any interest. Therefore, individual models may be more appropriate to this group. There was certainly a sense of loss amongst the participants, Boss (1999) describes ‘ambiguous loss’ which seems extremely relevant to this group in which the person is still physically present but absent in terms of personality, emotion and cognition.

After a period of trial and error, the carers begin to develop ways of managing their circumstances, some of which relate to specific behavioural strategies; others to taking time out; and others to their attitude towards their situation, eg. picking your battles. In the current study the participants also described the use of extreme behaviour management at times such as police involvement and sectioning.

It is proposed in the model there is a continuous, cyclical process between the ‘living bereavement’ stage and the ‘coping’ stage. Since the condition itself is progressive in nature,

the carer is required to adapt their coping as new challenges emerge. These challenges may relate to the condition itself as well as other factors such as ageing, family coping mechanisms and environmental factors. This is in keeping with Lazarus and Folkman's transactional model (1984) which states that coping is not static and is dependent on the relationship between the individual and their environment.

Whilst elements of the content of the presented model are consistent with previous research, this model ties together elements of the process and includes the context of historical factors and the FTD symptoms in understanding this process further.

Limitations of study

Whilst the current study included more participants than other qualitative studies in the area of carer experiences of FTD, it is still a small sample, and this has implications as to the transferability of the findings to others. Similarly the participants were all either from a white British or white Irish ethnicity, therefore it remains unclear whether these findings would relate to other ethnic groups. Whilst not emergent from the data in the current study, future research may indicate that culture may be an additional mediating factor.

The sample was also mainly women (10 of the 12 participants) and therefore the transferability to male carers may be questioned. It was, however, not felt that the experiences reported by the males in this study were overly different to that of the women. Further research would be advised.

The participants were recruited from a support network, and whilst this allowed for a range in terms of location and time since diagnosis, it is important to highlight that these participants had all sought support and therefore other carers who do not seek support or have not begun to seek support were not present in the sample. This was suggested by one participant as to

why there were so few males showing interest in the study (*'men aren't good at coming into a group'*, P6).

Despite these limitations, this research adds to the current literature base. Emerging themes are tied together to create a process model related to this specific group of carers. Whilst it was a UK based study, participants were sourced from different parts of England, including Central London, rural areas and Northern towns. This allowed for the results to be more transferable as the resources and networks differ greatly between these areas.

Another positive in relation to this research was including two participants whose loved ones had passed away, this enabled the author to give an indication of various stages of the process, which would not have been possible if participants were recruited purely in the early stages. However, it would have added to the findings if there were more participants at an early stage.

The use of qualitative research is open to bias, however quality assurance techniques were used to reduce the impact of this. The use of qualitative methodology allowed for an increased depth of data and analysis. The use of grounded theory methodology in particular allowed for theory building and links to be made between components.

Future research

The experiences of minority ethnic groups has not been explored in the current study, nor in the further literature. Therefore, it would be interesting to explore the experiences of other ethnic groups within Britain and elsewhere to give an indication of how relevant these findings are to other groups.

Similarly, since there were only two male participants in the current study, it would be helpful to explore the experiences of male carers further to explore the generalisability of the current findings.

The current study only included spousal carers, therefore the model may not be relevant to other caring relationships, and it would be interesting to explore the adjustment process for other family members such as sons and daughters or even parents of those with FTD, given the early onset of the condition.

In the current study, relational models of grief were not incorporated in the method. It was assumed that those with FTD symptoms such as loss of empathy would not be able to engage in these conversations, however, this was not explored. Therefore it would be interesting to give people with FTD more of a voice by including them in the research.

Since the current research and other qualitative research in this area has only focused on one time point regarding participant interviews, longitudinal research is needed to further clarify elements of the proposed process.

Clinical Implications

Guidance by the National Institute of Health and Care Excellence (NICE, 2006) for psychosocial support for dementia is generalised to all subtypes. It is suggested general guidance be offered to bv-FTD carers such as practicalities as in applying for lasting power of attorney and carer assessments. Whilst some general support may be helpful, participants in the current study found support to be relevant to Alzheimer's dementia and not appropriate for FTD. It is important for services to acknowledge the particularly challenging experiences of bv-FTD carers and offer support accordingly. Psychoeducational support including information specific to FTD such as common symptoms and challenges may be of benefit.

Carers may also benefit from being able to engage with discussions about helpful strategies for behaviour and risk management rather than memory strategies.

Since no medication is prescribed for FTD, carers often find themselves discharged from memory services without ongoing support. This needs to be reviewed as there isn't currently a specific service to provide ongoing psychosocial support for these carers who experience unique stressors.

There was also a perception amongst participants that those involved in memory clinics did not know much about FTD and are therefore unable to provide FTD specific support.

Therefore staff training would be recommended to enhance knowledge of the condition and carer experiences.

Psychological input could also be considered with regards to processing loss, relationship change and complex emotions such as guilt. Silverberg (2007) also recommends psychosocial support relating to grief and loss. Additionally, Whitebird et al. (2012) found that an 8-week mindfulness programme reduced stress and depression and improved mental health in a sample of 78 dementia family carers. This was not specific to FTD carers but may be of benefit.

Peer support is also recommended by the NICE guidance (2006), and was highlighted as an important element in the adjustment process. Given that FTD is a relatively rare diagnosis, it is likely that services will have difficulty providing peer support groups specifically for this condition. However, making professionals aware of groups such as the FTD network would allow them to signpost accordingly.

In the current study one aspect of the adjustment process was the importance of the carer making time for themselves. Silverberg (2007) suggests that carers are more likely to take up

respite if recommended by healthcare professionals as this reduces feelings of guilt in relation to this.

Conclusion

The current study provides a theoretical model to describe the experiences of adjustment in those caring for a spouse with a diagnosis of bv-FTD. This model includes a range of stages in the adjustment process and attempts to link together these stages in order to capture and portray the overall complexity of the process and day to day experience of this group. Of importance in understanding this process are the carer's history and the particular presentation of FTD which make this journey individualised. Complex emotions and relationship strain are present throughout the process, even prior to the diagnosis of FTD.

References

- Alzheimer's Society (2016, 25th April) What is frontotemporal dementia? Retrieved from https://www.alzheimers.org.uk/site/scripts/documents_info.php?documentID=167
- American Psychiatric Association (2013). Diagnostic and statistical manual of mental disorders (5th ed.). Arlington, VA: Author.
- American Psychiatric Association. (2000). Diagnostic and statistical manual of mental disorders DSM-IV-TR fourth edition (text revision). Arlington, VA: Author.
- Armstrong, N., Schupf, N., Grafman, J., & Huey, E. (2013). Caregiver burden in frontotemporal degeneration and corticobasal syndrome. *Dementia and Geriatric Cognitive Disorders*, 36, 310-318. doi: 10.1159/000351670
- Boss, P. (1999) *Ambiguous loss: Learning to live with unresolved grief*. MA: Harvard University Press.
- British Psychological Society (2010) *Code of human research ethics*. Leicester: BPS.
http://www.bps.org.uk/sites/default/files/documents/code_of_human_research_ethic.
- Charmaz, K. (2014). *Constructing grounded theory*. London: Sage publications.
- Corbin, J., & Strauss, A. (2008). *Basics of qualitative research: Techniques and procedures for developing grounded theory*. London: Sage publications.
- Department of Health. (2015). *The government's mandate to NHS England for 2016 –2017*.
<https://www.gov.uk/government/publications/nhs-mandate-2016-to-2017>
- Diehl-Schmid, J., Schmidt, E. M., Nunnemann, S., Riedl, L., Kurz, A., Förstl, H., . & Cramer, B. (2013). Caregiver burden and needs in frontotemporal dementia. *Journal of Geriatric Psychiatry and Neurology*, 26(4), 221-229 doi: 10.1177/0891988713498467

Englund, B., Brun, A., Gustafson, L., Passant, U., Mann, D. M. A., Neary, D., & Snowden, J.

S. (1994). Clinical and neuropathological criteria for frontotemporal dementia. *Journal of Neurology, Neurosurgery & Psychiatry*, 57, 416-418. Retrieved from <http://www.ncbi.nlm.nih.gov/pmc/articles/PMC1072868/>

Kindell, J., Sage, K., Wilkinson, R., & Keady, J. (2014). Living with semantic dementia. A case study of one family's experience. *Qualitative Health Research*, doi:1049732314521900.

Kumamoto, K., Arai, Y., Hashimoto, N., Ikeda, M., Mizuno, Y., & Washio, M. (2004). Problems family caregivers encounter in home care of patients with frontotemporal lobar degeneration. *Psychogeriatrics*, 4(2), 33-39

Lazarus, R. S., & Folkman, S. (1984). *Stress, appraisal, and coping*. Springer publishing Company:NY.

Lindau, M., Almkvist, O., Kushi, J., Boone, K., Johansson, S. E., Wahlund, L. O., ... & Miller, B. L. (2000). First symptoms—frontotemporal dementia versus Alzheimer's disease. *Dementia and geriatric cognitive disorders*, 11(5), 286-293.
DOI:10.1159/000017251

Massimo, L., Evans, L. K., & Benner, P. (2013). Caring for loved ones with frontotemporal degeneration: The lived experiences of spouses. *Geriatric Nursing*, 34, 302-306. doi: <http://dx.doi.org/10.1016/j.gerinurse.2013.05.001>

Mays, N., & Pope, C. (2000). Assessing quality in qualitative research. *British Medical Journal*, 320, 50. doi: <http://dx.doi.org/10.1016/j.gerinurse.2013.05.001>

Mendez, M. F., McMurtray, A., Licht, E., Shapira, J. S., Saul, R. E., & Miller, B. L. (2006). The scale for emotional blunting in patients with frontotemporal dementia. *Neurocase*, 12, 242-246. doi:10.1001/archneur.64.6.830.

Merrick, K., Camic, P. M., & O'Shaughnessy, M. (2013). Couples constructing their experiences of dementia: A relational perspective. *Dementia*, 00, 1-17 DOI:

10.1177/1471301213513029

Mioshi, E., Bristow, M., Cook, R., & Hodges, J. R. (2009). Factors underlying caregiver stress in frontotemporal dementia and Alzheimer's disease. *Dementia and Geriatric Cognitive Disorders*, 27, 76-81. <http://dx.doi.org/10.1016/j.gerinurse.2013.05.001>

National Institute for Health and Care Excellence (2006) NICE guidelines [CG42];

Dementia: Supporting people with dementia and their carers in health and social care. <https://www.nice.org.uk/guidance/cg42>

NHS Choices (2015) Frontotemporal dementia. Retrieved from

<http://www.nhs.uk/Conditions/frontotemporal-dementia/Pages/Introduction.aspx>

Nicolaou, P. L., Egan, S. J., Gasson, N., & Kane, R. T. (2010). Identifying needs, burden, and distress of carers of people with Frontotemporal dementia compared to Alzheimer's disease. *Dementia*. doi: 10.1177/1471301209354024

Nolan, M., Ryan, T., Enderby, P., & Reid, D. (2002). Towards a more inclusive vision of dementia care practice and research. *Dementia*, 1, 193-21. doi:

10.1177/147130120200100206

Oyebode, J. R., Bradley, P., & Allen, J. L. (2013). Relatives' experiences of frontal-variant frontotemporal dementia. *Qualitative Health Research*, 23(2), 156-166. doi:

10.1177/1049732312466294

Riedijk, S. R., De Vugt, M. E., Duivenvoorden, H. J., Niermeijer, M. F., Van Swieten, J. C., Verhey, F. R. J., & Tibben, A. (2006). Caregiver burden, health-related quality of life and coping in dementia caregivers: a comparison of frontotemporal dementia and

Alzheimer's disease. *Dementia and Geriatric Cognitive Disorders*, 22, 405-412,

DOI:10.1159/000095750doi:10.1159/000095750

Riedijk, S., Duivenvoorden, H., Rosso, S., Van Swieten, J., Niermeijer, M., & Tibben, A.

(2008). Frontotemporal dementia: change of familial caregiver burden and partner relation in a Dutch cohort of 63 patients. *Dementia and Geriatric Cognitive Disorders*, 26(5), 398-406, doi: 10.1159/000164276

Disorders, 26(5), 398-406, doi: 10.1159/000164276

Robinson, L., Clare, L., & Evans, K. (2005). Making sense of dementia and adjusting to loss:

psychological reactions to a diagnosis of dementia in couples. *Aging & Mental Health*, 9(4), 337-347. doi:10.1080/1360786050011455

Health, 9(4), 337-347. doi:10.1080/1360786050011455

Royal College of Psychiatrists (2015) Memory Services National Accreditation Programme

(MSNAP): Standards for Memory Services.

<http://www.rcpsych.ac.uk/PDF/MSNAP%20standards%205th%20edition.pdf>

Saldaña, J. (2015). *The coding manual for qualitative researchers*. London: Sage

publications.

Silverberg, E. (2007). Introducing the 3-A grief intervention model for dementia caregivers:

acknowledge, assess and assist. *OMEGA-Journal of Death and Dying*, 54, 215-235.

doi: 10.2190/D1R5-0473-1922-4N70

Stroebe, M., & Schut, H. (2010). The dual process model of coping with bereavement: A

decade on. *Journal of Death and Dying*, 61, 273-289. doi: 10.2190/OM.61.4.b

Vernooij-Dassen, M., Derksen, E., Scheltens, P., & Moniz-Cook, E. (2006). Receiving a

diagnosis of dementia: The experience over time. *Dementia*, 5, 397-410. doi:

10.1177/1471301206067114

Whitebird, R. R., Kreitzer, M., Crain, A. L., Lewis, B. A., Hanson, L. R., & Enstad, C. J.

(2012). Mindfulness-based stress reduction for family caregivers: a randomized

controlled trial. *The Gerontologist*, gns126. doi: 10.1093/geront/gns126

Wong, C. C., & Wallhagen, M. I. (2012). Frontotemporal dementia: The impact of patient behavioral symptoms on the physical and mental health of family caregivers.

Dementia and Geriatric Cognitive Disorders Extra, 2, 516-528, doi:

10.1159/000345082

Section C:

Appendices of supporting information

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Salomons Centre for Applied Psychology

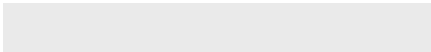
Canterbury Christ Church University

Appendix A: Crowe appraisal table

<u>Reference</u>	<u>Preliminaries</u>	<u>Introduction</u>	<u>Design</u>	<u>Sampling</u>	<u>Data collection</u>	<u>Ethical Matters</u>	<u>Results</u>	<u>Discussion</u>	<u>Total</u>
	/5	/5	/5	/5	/5	/5	/5	/5	/40
Mourik, J.C., Rosso, S. M., Niermeijer, M. F., Duivenvoorden, H. J., van Swieten, J. C. & Tibben, A.	4	2	3	4	2	4	4	3	27
Riedijk, S. R., De Vugt, M. E., Duivenvoorden, H.E., Niermeijer, M. F., Van Swieten, J. C., Verhey, F. R. J. & Tibben. A. (2006).	5	5	2	3	2	4	4	4	29
Knutson, K. M., Zamboni, G., Tierney. M. C. & Grafman, G. 2008	4	5	4	3	2	3	5	3	29
Riedijk, S., Duivenvoorden, H., Van Swieten, J.V., Niermeijer, M. & Tibben, A. (2009)	4	5	3	4	4	5	5	4	34
Mioshi, E., Bristow, M., Cook, R. & Hodges, J. R. (2009).	5	5	2	3	4	4	4	5	32
Armstrong, N., Schupf, N., Grafman, J. & Huey, E. D. (2013).	4	4	2	3	1	5	5	3	27
Mioshi, E., Foxe, D., Leslie, F., Savage, S., Hsieh, S., Miller, L., Hodges, J. & Piguet, O. (2013)	3	3	2	3	1	3	3	3	21
Cosseddu, M., Alberici, A., Pelizzola, S., Padovani, A. & Borroni, B. (2013)	2	0	1	1	0	1	3	3	11
Merrilees, J., Dowling, G. A., Hubbard, E., Mastick, J., Ketelle, R. & Miller, B. L. (2013)	3	3	3	2	2	1	3	3	20
Hsieh, S., Irish, M., Daveson, N., Hodges, J. & Piguet, O. (2013)	4	5	4	4	4	5	5	5	36

Caring in FTD

Mekala, S., Alladi, S., Chandrasekar, K., Fathima, S., O'Connor, C. M. O., McKinnon, C., Hornberger, M., Piguet, O., Hodges, J. & Mioshi, E. (2013)	5	4	3	4	5	5	5	5	36
Diehl-Schmid, J., Schmidt, E-M., Nunnemann, S., Riedl, L., Kurz, A., Forstl, H., Wagenpfeil, S. & Cramer, B. (2013)	4	5	2	3	4	4	5	4	31
Merrilees, J., Hubbard, E., Mastick, J., Miller, B. L. & Dowling, G. A. (2014).	3	4	2	4	3	2	3	5	26
Wong, C. C. & Wallhagen, M. I. (2012).	5	5	3	5	4	5	5	5	37
Guevara, A. B., Knutson, K. M., Wassermann, E. M., Pulaski, S., Grafman, J. & Krueger, F. (2015).	3	4	3	2	2	4	4	5	27
Roche, L., Croot, K., MacCann, C., Cramer, B. & Diehl-Schmid (2015)	4	5	3	4	4	3	4	5	32



Appendix B:
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Appendix C: Interview Schedule

Check they have read the info sheet – give another copy

Any questions

Quick recap – don't want to cause any distress but might be upsetting, can stop at any time.

- Confidential
- If want to withdraw – they can at any time

Consent form signed

Questions before we begin?

Interview will concentrate on after the diagnosis, I have a few questions but we'll just play it by ear really and see where the conversation takes us. Ok?

Can you tell me about your experience since the diagnosis?

How has the diagnosis changed your life?

How would you describe yourself? Do you think you have changed?

Has there been anything that you have found helpful or unhelpful?

Can you tell me how you manage the difficult times?

Do you have any strategies you have developed?

Is there anything you do differently now, that you didn't do at the time of diagnosis?

Can you tell me about your relationship with? How has this changed?

How do you look after yourself?

How do you view the future – might not ask

Is there anything that you would share with someone in the early stages of the experience?

Appendix D: Information Sheet

Information about the research

Understanding the experiences of carers of people with frontotemporal dementia (FTD).

Hello. My name is and I am a trainee clinical psychologist at Canterbury Christ Church University. I would like to invite you to take part in a research study. Before you decide, it is important that you understand why the research is being done and what it would involve for you.

Talk to others about the study if you wish.

(Part 1 tells you the purpose of this study and what will happen to you if you take part.

Part 2 gives you more detailed information about the conduct of the study).

What is the purpose of the study?

The purpose of the study is to gain more of an understanding of the day to day experiences of being an informal carer for someone with a diagnosis of FTD. There is a lack of research which has looked in detail at how people manage this experience everyday. This will give you an opportunity to tell your story and this information would be useful for dementia services to provide more specialist support for those diagnosed with FTD and their families/ carers.

Why have I been invited?

You have been invited to participate in the study as a result of your response to my advertisement in the FTD network newsletter (or expressed an interest via the memory service – contingency plan).

Do I have to take part?

It is up to you to decide whether to join the study. If you agree to take part, I will then ask you to sign a consent form. You are free to withdraw at any time, without having to give a reason.

What will happen to me if I take part?

Once you have returned the attached demographic questionnaire, I will contact you to arrange a date for an interview at a time and location which is appropriate for you. The interviews can be conducted at your home if this is the most convenient for you. This will be a one-off interview and is likely to take 60-90 minutes. The interview will be recorded by audiotape. It will be important that we have a private space, so if the person you are a carer for lives with you, you may need to make arrangements so that he or she will not be present. It is fine to discuss your participation with him or her if this seems appropriate, but the interview is about **your** experience.

Following the interview we will have an opportunity for you to ask me any further questions you have about the study that may have occurred to you.

If you wish to receive information on the results of the study you will be given the option to opt in and this information will be sent out to you once collated. There will also be an option to comment on a summary of the findings by telephone after you have been sent it to read.

Expenses and payments

You will be reimbursed for your travel if you are interviewed outside of your home, up to £10.

What will I have to do?

Should you wish to participate in the study, you will be required to complete the attached brief questionnaire and return it in the prepaid envelope. You will then be contacted to arrange the interview. At the time of interview you will be asked to sign a consent form. During the interview I will ask you questions about your experiences as an informal carer. You are only expected to share what you feel comfortable with and can end the interview at any time should you wish to do so.

What are the possible disadvantages and risks of taking part

Given the nature of the interview, it may be that difficult or emotive topics may arise, however, as mentioned above you are only expected to share as much as you feel comfortable with.

What are the possible benefits of taking part?

We cannot promise the study will help you but the information we get from this study may help improve the treatment of people with FTD and the type of support that would be helpful for carers.

What if there is a problem?

Any complaint about the way you have been dealt with during the study or any possible harm you might suffer will be addressed. The detailed information on this is given in Part 2.

Will my taking part in the study be kept confidential?

Yes. We will follow ethical and legal practice and all information about you will be handled in confidence. The details are included in Part 2.

This completes part 1.

If the information in Part 1 has interested you and you are considering participation, please read the additional information in Part 2 before making any decision.

Part 2 of the information sheet

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

Should you wish to withdraw from the study, your interview will not be included in the study and any information about you will be destroyed.

What if there is a problem?

If you have a concern about any aspect of this study, you should ask to speak to me in the first instance and I will do my best to address the issue [contact number or email].

If you remain unhappy and wish to complain formally, you can do this by contacting Professor Paul Camic at Canterbury Christ Church University on.....

Will my taking part in this study be kept confidential?

Yes, any electronic information will be kept on a password protected memory stick. Paper based information will be stored securely. The only person who will have access to identifiable data will be the researcher (myself), for the purpose of contacting you. This information will be retained until the submission of the study to the University and it will then be disposed of securely.

All information which is collected about you during the course of the research will be kept strictly confidential, and any information about you will have your name and address removed so that you cannot be recognised. When the interview recording is transcribed to written text, names of people and places will be changed to protect anonymity. Participants have the right to check the accuracy of data held about them and correct any errors.

What will happen to the results of the research study?

It is intended that the results will be published and will be made available to participants via the FTD network. You will not be identified in any report or publication, as anonymised quotes from your interviews will be used in the report.

Who is organising and funding the research?

Canterbury Christ Church University is the organising body for this study.

Who has reviewed the study?

All research is looked at by an independent group of people, called a Research Ethics Committee, to protect your interests. This study has been reviewed and given favourable opinion by the NHS Research Ethics Committee.

Further information and contact details

If you would like to speak to me and find out more about the study or if have questions, you can leave a message for me on Please say that the message is for me,, and leave a contact number so that I can get back to you.

Appendix E: Consent Form

CONSENT FORM Version 1 (04/05/12)

Title of Project: Understanding the experiences of carers of people with frontotemporal dementia (FTD).

Name of Researcher:

Please initial box

1. I confirm that I have read and understand the information sheet dated 4/12/14 (version 1) for the above study. I have had the opportunity to consider the information, ask questions and have had these answered satisfactorily.

2. I understand that my participation is voluntary and that I am free to withdraw at any time without giving any reason.

3. I understand that my interview will be audio-recorded and agree to this.

4. I agree that anonymous quotes from my interview may be used in published reports of the study findings

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

Name of Participant _____ Date _____

Signature _____

Name of Person taking consent _____ Date _____

Signature _____

Appendix F: Summary of themes from bracketing interview

Assumptions from Bracketing Interview

My experience working in memory clinics has made me already aware of some of the views of people I have worked with so far.

Anticipated findings –

- **They learn and develop strategies over time**
- **Sense of humour is important**
- **Stop challenging things**
- **Change of relationship**
- **Change of role**
- **Stop work**
- **Adjust to their new life**
- **Lack of FTD support**
- **Conversation re diagnosis taking too long**
- **Services aimed at AD**
-

Expecting that the interviews will be easy as carers love to chat. Met some potential participants at the FTD network annual seminar.

Other factors putting pressure on – want to pass the course, and want it to get published.

Appendix G: Extracts from reflective diary

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Appendix H: Categories and example quotes

<u>Key Constructs</u>	<u>Main categories</u>	<u>Example Participant Quotes</u>
<p>Life before bv-FTD</p>		<p>P2 'We lived very parallel lives' P3 'We had such a good life' P5 'We had a lot of things in common' P8 'It's always been [him] and me' P10 '...such a good relationship' P11 'We used to go to so many fantastic places' P12 'We always did everything together'</p> <p>P4 'I've always looked on the positive' P4 'I've been a sort of forceful, strong...doing new things' P5 'I was a very quiet and private person' P6 'Might be something about my philosophy as a Christian' P8 'I'm not really a people person if I'm honest', 'I'm happy enough to be independent' P10 'I was a softy and I was happy go lucky' P11 'I'm intelligent...I find it easier to cope with organisations'</p> <p>P3 'He had such a fine brain' P4 '...was quiet, very modest and thoughtful' P5 '...was a restrained sort of person' P10 'He was a highly intelligent man' P11 'She used to do everything' P1 'I was the perfect daughter in law, not now' P1 'We'd been the perfect family before' P3 'I'm the eldest of 8 so I'm used to having to fix it from all angles and sides' P5 'There's no children, it's just us.' P3 'My career (nurse) had also enormously helped me' P6 'I trained as a counsellor....been helpful'</p>

		<p>P8 'We were both head teachers so you do learn to be on your own'</p> <p>P10 'My experience as a nurse.....got me through'</p> <p>P1 'His mother was very similar'</p> <p>P4 'And their mum.....had.....FTD'</p> <p>P7 'His father had some sort of dementia'</p> <p>P8 '[His] father...had this condition'</p>
Characteristics of bv-FTD	Incontinence/ Personal hygiene	<p>'Awful incontinence issues' P5</p> <p>'He is doubly incontinent' P2</p> <p>'Incontinence was getting worse' P6</p> <p>'We had moved from continent to incontinent in that one space' P3</p> <p>'He is very dirty' P7</p> <p>'He can go 3 months without having a wash' P12</p> <p>'He was smelly and horrible' P9</p>
	Repetitive Behaviours	<p>P4 'Routine is very rigid'</p> <p>P9 'It was his routine, it was compulsive'</p> <p>P1 'He was pacing'</p> <p>P12 '[Husband] was hoarding'</p> <p>P12 'He would chant my name all night'</p> <p>P12 'One of his compulsions has been dressing and undressing. It would go on for 2 hours'</p> <p>P9 'He shows many OCD traits'</p>
	Behavioural changes	<p>Childlike behaviour</p> <p>P5 'It's almost as if I have adopted a child'</p> <p>P2 'Child 2 years olds and you are not far off'</p> <p>P1 'I now had 3 children'</p> <p>P3 'Once an adult twice a child'</p> <p>P4 'He's regressing'</p> <p>P6 'Regressing and being childlike'</p>

		<p>P7 'Odd things that are childlike' P9 'He was just like another child'</p> <hr/> <p>Extreme behaviour</p> <p>P7 'He has been banned from 6 places in [place]' P1 '...call from the police about a man that was urinating in someone's garden' P6 '...would just masturbate just totally unaware of what was going on' P5 'He is beginning to pick up a slight case of shoplifting.' P7 'It's extreme personality' P11 'It's real extreme behaviour' P2 'He was sort of a public nuisance'</p> <hr/> <p>Unpredictability</p> <p>P12 'Every day you wake up in fear because you don't know what is going to happen today' P4 'You have no idea what's going to happen next' P5 'You never know what is going to happen next' P7 'Those things were a surprise to me' P8 'You can't quite tell how quickly the deterioration will be' P11 '...can just come out of the blue' P11 'Be prepared for changes in behaviour which are not constant' P12 ' No two days are the same'</p> <hr/> <p>Disinhibition</p> <p>P5 'Lost all his inhibitions' P7 'He just doesn't know how to behave' P11 'It is inappropriate' P5 'He will just go up a complete stranger if they are laughing and just join in' P12 'He just says whatever comes into his head'</p>
--	--	--

		<p>P9 'Inappropriate behaviour. He'd pull the girl's hair and touch them'</p> <p>P11 'She's do all kinds of things in public that she would never have done'</p>
	Loss of social skills	<p>Inappropriate remarks and insults</p> <p>P11 '[She] repeats the joke to everyone, no idea that it is inappropriate at all'. P9 'He was insulting people' P12 'He could say awful things to people' P7 'He told a man with 2 grandchildren...oh you must be a paedophile' P12 'A lot of sexual comments have been made'</p> <p>Communication difficulties</p> <p>P4 'His language is going away' P4 'He often hasn't got a clue what we are talking about' P8 'He doesn't have much conversation really'</p> <p>Use of set phrases</p> <p>P7 'I could write the sentences he uses on a postcard' P4 'He has got this list of wonderful words that he uses'</p>
	Risky behaviour	<p>Aggression</p> <p>P3 'We are getting more and more agitated' P6 '...beginning to be aggressive' P1 'I had him screaming that he was going to kill me' P1 'He went from aggressive to violent' P9 'He would get nasty' P12 'He threatened to punch an old lady'</p>

		<p>Vulnerability P4 'He is very vulnerable'</p> <p>Risk to self</p> <p>P1 'We had 4 fires' P4 'If there is an emergency, he hasn't got a clue what to do'</p> <p>Police Involvement P7 'We had the police here five times that year'</p> <p>Sectioning/ Hospitalisation P1 'It was best to admit him' P2 'We had 'husband' sectioned at one stage' P11 'She had to be admitted to the mental health ward'</p> <p>Psychotropic Medication P1 'They put him on olanzapine' P7 'He's on sertraline'</p>
	<p>Loss of organisational skills</p>	<p>P1 'I had screamed several times when we'd be driving, all of that sort of decision making had gone' P7 'The concept of choice was too difficult' P4 'If you have to react quickly, that's when he gets really slow, like slow motion'</p>
	<p>Loss of spatial awareness</p>	<p>P4 'He bumps into you the whole time....whereas before we'd just sort of work around each other' P8 'I can see his navigational skills are decreasing'</p>

Caring in FTD

	Changes in dietary taste	<p>P7 'There are food obsessions'</p> <p>P11 'Drinking became w huge problem'</p> <p>P6 'He has developed a really sweet tooth'</p>
	Loss of personality	<p>P12 'He's no personality'</p> <p>P8 'He has withdrawn'</p> <p>P8 'He has become a much more timid person'</p> <p>P5 'He changed so much, a totally different person'</p> <p>P4 'He is just sort of drifting away'</p> <p>P5 'The personality goes first'</p> <p>P6 '...beginning to show signs of anxiety and loss of confidence'</p> <p>Loss of emotion</p> <p>P5 'Becoming cold and uncaring and selfish'</p> <p>P5 'He doesn't have sympathy or empathy for other people'</p> <p>P12 'He's never happy, he's never sad'</p> <p>P11 'She rarely smiles, but laughing and crying have almost stopped'</p> <p>P11 'She just couldn't see that I was ill at all, no sympathy, no concern'</p> <p>P5 'The sort of tangents of emotion are all gone'</p> <p>P2 'He just doesn't care, I find that difficult to understand'</p>
	Apathy	<p>P2 'He is entirely apathetic'</p> <p>P12 'He's no motivation'</p> <p>P9 'He just doesn't do anything'</p> <p>P12 'He just can't be bothered'</p>

The road to FTD diagnosis	Noticing changes	P7 'Looking back it was 10 years of change' P8 'Noticed things for quite a few years' P9 'The change had been over so long' P10 'The past few years had been horrendous' P2 'He was behaving very peculiarly' P2 'I knew something was wrong' P7 'His behaviour was beginning to change' P5 'He kept disappearing' P2 'Things were not as they should be'
	Embarrassment	P5 'I would get very embarrassed' P7 'Extreme embarrassment' P9 'It was just so embarrassing'
	'What is happening?'/ Making Assumptions	P5 'I still didn't understand that this was illness' P7 'I put it all down to alcohol' P9 '...maybe it's just me' P12 'Before the diagnosis I was saying, don't be silly' P4 'I knew there was something wrong and it was driving me mad' P5 'I was convinced he was meeting some other woman' P3 'I had several things in mind' P7 'I believed he had Korsakoff's syndrome'
	Blame	P7 'I had blamed him, I thought everything was his fault'
	Anger	P5 'I became a really awful person' P12 'I was getting angry with him' P6 'I lost my temper' P7 'I lost my temper, I screamed and threw a bottle of wine at him'
	Impact on the relationship	P5 'The illness did a lot of damage to my marriage' P7 'I did have a lover for a while' P7 'There were a lot of rows before [the diagnosis]'

	Seeking medical advice	P3 'It took me about 6 months to persuade anyone to get a referral' P5 'You can't take your husband to a doctor and say this man doesn't love me anymore'
	Misdiagnosis	P1 'They said it was arsenic poisoning' P3 'They thought it was post-operative stress' P2 'we were told to go to marriage counselling' P12 'They would say [husband] is just playing up, it was depression'
'Living bereavement'	Denial/ Resistance	P2 'I tried to let it interfere with me as little as possible' P3 'I was so resistant to it' P8 'I thought I could manage it all' P10 'We tried to get on with our lives' P11 'I remember first of all trying to battle...' P12 'I didn't want to think it was as bad as it was'
	Guilt	P7 'After the diagnosis I felt it's not his fault' P4 'I had been so horrible'
	Relief	P5 'The first effect of the diagnosis was relief' P9 'It meant there was a reason for all this awful behaviour'
	Finding a narrative	P1 'I couldn't tell anyone, I thought I was just not coping' P8 'For a long time we just avoided talking about it' P9 'It was like a big dark secret and I was holding it in' P4 'If I had a pound for every time somebody said oh my grandmother had Alzheimers...sometimes I just say I know, well it's such a different disease' P9 'I had these little cards to show her what was wrong with him'

	<p>Reassessing/Processing loss</p>	<p>P3 'You think it will last forever and it doesn't' P4 'We have given up everything that is part of everyday life' P7 '[Relationship]....we don't have one' P9 'Our relationship doesn't exist anymore' P10 'Everything that had gone before wasn't anymore' P12 'Our life together is no more' P3 'I lost my sense of being carefree' P1 'You lose them' P3 'He was going into this black hole' P12 'He's not my husband anymore' P1 'I could no longer rely on him' P2 'You have to reassess what they are like' P6 'not able to share food' P8 'It's not mates anymore' P10 'I expect different things out of life now' P5 'This is what my life will be now' P5 'In a way I'm being two people. I'm having to do everything he used to' P6 'It's totally me as the carer' P5 'I thought of all the things we wouldn't be able to do' P11 'I can't help being angry at the life we've lost' P11 'All my expectations have all gone'</p>
	<p>Lack of appropriate support</p>	<p>General dementia support</p> <p>P8 'The psychiatrist said he'd given us some cognitive exercises of something, I don't consider the suggestion of using a diary much good at all' P9 '[Local dementia service] dropped him because he is too much for them' P12 'We had some input by the mental health team, they didn't know much about FTD' P5 'We go to dementia suppers, lunches, of course they are all Alzheimer's, nobody like him at all' The Alzheimer's society is not so helpful....these are the elderly and people with loss of mobility. T1 We were offered a whole lot of things that were absolutely useless T2</p>

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		Lack of treatment P8 'What's the point [of diagnosis], there's no treatment.'
	Lack of understanding from others	P1 'My sister in law told me it was a socially unacceptable diagnosis' P9 'One woman in a shop told me she knew why she left her husband, she thought I was being abused' P7 'His brother will have nothing to do with him, they just find him so unpleasant'
Coping with FTD	Seeking FTD specific support	The picks support group were a great support for me, they give valuable information. T12 There was another lady on the course whose husband had FTD T7 It's [A.S guidance] not that helpful because there is nobody with FTD there. T7 I joined the FTD forum, that was my lifeline mainly. T6 The FTD support group is very good at listening T1 One is more likely to bond more if they have this in common. T2 I was lucky enough to find a woman who did understand FTD. T12 Being able to talk about it without being embarrassed is useful T2 The FTD clinic I went to....that was really brilliant....there were lots of other people T8
	Restricting	I have my boundaries and that works T12 You have to say 'no' a lot of times T12 [Husband] went through a stage of eating nonstop so I put locks on the fridges. T2
	Self care	I have a room to go into when he is whistling his head off and I can't stand it. T9 He'd shout me all the time, I took him to the office, put the TV on and went to have a shower. T12 I've had to learn to move out of the room if he is offending me too much. T12 I get up early in order to have an hour on my own T5
	Extreme strategies	It's extreme behaviour, and you often have to take extreme things to deal with it. T12

		<p>I horrified myself by spanking him T2 There were occasions when I hit her on the hand when she was digging her nails in T6 I sent the children to boarding school T1 I managed to get the phone and call 999 T1 The only response is to call the police T3</p>
	Dsitraction	<p>I found one of the things which distracted her was to sing a song. The singing would distract her from digging her nails in. T6</p> <p>Distraction has got to be it T6</p>
	Routine	<p>Routine is the most important, everything is glued to that T4 The routine is rigid, if we can stick to that routine he is much, much better that if he doesn't. T4 He's very worked up if it goes past 7:30 [for dinner] His routine is about 3 hours non stop. T12 We have absolute routine, even things like the way he gets dressed. T12</p>
	Adapting communication	<p>It's probably making me more bossy T8 Don't suggest anything because he wont go along with it. T7 I try not to ask questions T7 I don't give him a choice T7 I'm quite bossy with him T4 There's no point in talking about it [behaviour to the PwD. T7 He responds a lot to the tone of voice, so the way you say things is much more important T8 When he is clearly frightened or upset... I would be a kinder person than I naturally feel inclined to be. T8 If things are a problem all I have to do is say something really stupid or childish, just to get him to laugh. T5 I have to filter information. I couldn't say tomorrow we are going to such a thing, otherwise at midnight he'll be up, and at 1, and 2...so now I have to stop saying we are doing such a thing until maybe half an hour before we do it. T3 Directing her to do something wasn't effective, it had to be very much one of encouragement. T6</p>

Caring in FTD

	Deception	<p>I started 'losing' the keys, the only way I could stop him driving. T1</p> <p>There is a certain amount of deception involved T8</p> <p>He eats a lot of sweet stuff, I hide it, I do hide chocolate. T12</p> <p>I creep into his bedroom and remove the jumper and replace it with a clean jumper before he wakes up in the morning T7</p> <p>My son was shaving his father, to save a fuss, this would be under the guise of learning how to shave. T1</p>
	Picking the battles	<p>At first I tried to battle her saying 'it's gone', and then it's embarrassing but I'm going to change things by telling her not to do it. T11</p> <p>It's almost impossible to stop that behaviour, I just mitigate really. I do that so I can have a bit of peace. T11</p> <p>You'd be amazed at what we've let go and don't worry about. T4</p> <p>He had decided to lie in bed with his shoes on, I said it's not worth having a fight to take them off. T1</p>

Appendix I: Ethical Approval

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Appendix J: Transcript extract with interpretation

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Appendix K: Feedback to participants and ethics panel

Qualitative exploration of the experiences of adjustment in family carers of those with behavioural variant frontotemporal dementia

Thank you for taking the time to be interviewed as part of this study. I am in the process of writing up and will be submitting the study in partial fulfilment of the Canterbury Christ Church University Doctorate in Clinical Psychology. As part of my write-up, I would like to hear your views on the research findings and how well they capture your experience of the work. Below is a summary of the study and its findings – please send any feedback you have to z.rigg16@canterbury.ac.uk.

Aim

The current study aimed to further explore these experiences with an emphasis on how these carers adjust and accept their situation.

Method

Semi-structured interviews were completed with 12 participants (2 male). All participants were the spouse of someone with bv-FTD. Interviews were transcribed and analysed using a grounded theory methodology.

Results

Analysis of the data resulted in 5 main components and a number of sub-components which were linked together to form a theoretical model (enclosed on a separate sheet).

Participants described a journey to adjustment and acceptance, whether this was in relation to accepting the diagnosis, individual symptoms or general acceptance. This is named 'Carer Journey'. This process included 3 components; 'The road to diagnosis', 'Living Bereavement' and 'Coping'.

It was clear during the interviews that the carer journey begins prior to the diagnosis, with the carers interviewed reporting that changes in their loved one were noticed years before diagnosis. The road to diagnosis is often rocky due to the unusual presentation of symptoms. Carers often don't realise initially that the changes are due to an illness and this can result in blame, anger and damage to the relationship. Since this condition is relatively rare, it is often misunderstood by professionals and leads to misdiagnosis. These early experiences shape the rest of the carer journey.

Life following the (eventual) diagnosis of FTD was described and this tended to include aspects of loss; change; a range of emotions (such as relief and guilt); trying to find support, which was often not helpful; and finding a way of talking about the condition. This is named 'living bereavement' as a direct quote taken from an interview. This was used to indicate that experiences of living with FTD were of a continuous nature with certain aspects becoming easier or more difficult and therefore experiences changing over time.

With some time and trial and error, it was clear that the people interviewed had developed coping strategies. Some of these were recommended for other's to try. These included seeking support, asking for help, the importance of talking to others about the experiences. People interviewed described taking each day at a time and not trying to predict what might come next. They also described learning to 'pick your battles' and that not everything can be challenged. It was also clear that a lot of the strategies are individualised and differ to those used for someone with a different

type of diagnosis, for example Alzheimer's dementia. These strategies included restriction, deception, different communication styles and, at times, extreme strategies (such as police involvement). As the disease progresses, the carer is likely to move between the 'living bereavement' stage and the 'coping' stage as they begin to process further symptoms and develop strategies accordingly.

Every stage of the proposed process is influenced by a number of factors, meaning every individual's journey is likely to differ in some way. One factor was the past experiences of the carer, for example, their personality, their relationship with their spouse prior to the FTD, their job, their family values, etc. This seemed to be an important factor in how people experienced elements of this process and in how they learn to cope.

There was great variability in experiences with health services. Again, it appeared that negative experiences with professionals and services made this process of adjustment even more difficult. The importance of good quality services, with staff that understand FTD and continuous support for this group was emphasised. These positive experiences can encourage a carer to utilise existing strategies and develop new ones.

Participants spoke of the complex nature of the bv-FTD in terms of symptoms, level of dependency and progression. As indicated in the diagram, the entire carer journey takes place within the context of the constantly changeable and progressive condition that is bv-FTD. The range of symptoms was clear, and it was evident that these symptoms differ somewhat from the more understood forms of dementia, for example memory loss in Alzheimer's dementia.

The process is highly individualised and dependent on the carer experience, personality and resources prior to the onset of symptoms. The journey and impact on the carer also depended on the relationship between the carer and the person with FTD prior to the condition, for example, shared experiences and roles.

There are a number of clinical implications which can be drawn from these interviews. The interviews suggest that carers adopt a range of strategies to help them adjust and cope with their experiences. Therefore it is important for services to not only consider what is missing from services but also how they might help these carers to make the most of what they are already doing. Guidance by the National Institute of Health and Care Excellence (NICE, 2006) for psychosocial support for dementia is generalised to all subtypes. However, it does suggest that those carers experiencing higher levels of stress and depression be offered psychological therapy. It is important for services to acknowledge the particularly challenging experiences of bv-FTD carers and offer support accordingly. General guidance should also be followed, for example carer's assessments and psychoeducation. It would be helpful for FTD carers to receive specialised support to help repair and rethink the damage to the relationship, as well as the difficult feelings related to anger and guilt. Support regarding dementia strategies is often aimed at memory strategies, however for this group it may be more helpful to consider communication as a strategy as well as normalising some of the extreme strategies which are often necessary.

It was also suggested by some of the carers interviewed that groups and available support are often targeted at those with Alzheimer's type dementia, and that this does not always feel relevant to FTD and the professionals involved do not always seem to understand FTD. Therefore it is important for services to increase awareness amongst staff members of FTD, and training sessions should be encouraged. Services should also consider a flexible approach to care, in which those with or caring for someone with FTD can be offered appropriate support. Carers interviewed also described the usefulness of practical support, such as being informed about lasting power of attorney.

Peer support is recommended by the NICE guidance, and was highlighted as an important element in the adjustment process. Given that FTD is a relatively rare diagnosis, it is likely that services will have difficulty providing peer support groups specifically for this condition. However, professionals should be aware of groups such as the FTD network and signpost accordingly.

Additionally, carers included in this study felt that services were very separate and felt that it would be helpful for them to be more linked together. In a clinical setting it is important for professionals to understand the various levels of services and networks and for them to make some attempt to work collaboratively.

Any thoughts you have about these results, particularly whether or not this fits with your experience and ideas about the work, would be greatly appreciated. Thank you again for your involvement in this project!

Appendix L: Journal Guidance



Submission

Two types of manuscripts will be considered and should be submitted online:

1. Original Research Articles. An Abstract, Introduction, Materials and Methods, Results, and Discussion sections are required.

2. Review Articles in which a specific field is reviewed through an exhaustive literature survey. An Abstract is required and should be divided into Background, Summary and Key Messages. Review Articles should consist of a maximum of 4,000 words.

Names, postal and e-mail addresses of 6 experts in the appropriate area of research should accompany each manuscript. Referees suggested should not be from the same institution or be research collaborators of the author(s).

Should you experience any problems with your submission, please contact:

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Arrangement

All pages should be consecutively beginning numbered with the title page, then the text, acknowledgements, references and legends to figures.

Title page: The first page of each paper should indicate the title, the authors' names, the institute where the work was conducted, and a short title for use as running head.

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Abstracts of Review Articles: Should be divided into the following subsections: Background, Summary and Key Messages. The Background should provide a brief clinical context for the review and is followed by the Summary, which should include a concise description of the main topics covered in the text. The Key Messages encapsulate the main conclusions of the review.

Abstracts of Original Research Articles: The first page of the text should include an abstract of up to 10 lines. It should be structured as follows:

Background/Aims: What is the major problem that prompted the study?

Methods: How was the study carried out?

Results: Most important findings?

Conclusion: Most important conclusion?

Footnotes: Avoid footnotes.

Abbreviations: Abbreviations should not be used excessively in the text. Only standard abbreviations should be used. Nonstandard abbreviations of terms that are used frequently in the text should be explained by the term written out completely and followed immediately by the abbreviation in parentheses, for example: 'increase in norepinephrine (NE) content ...'.

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(b) Papers published only with DOI numbers:

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