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Title: Relatives' Experiences of Frontal-Variant Frontotemporal Dementia.

Publication year: 2013

Journal title: Qualitative Health Research.

Link to original published version: http://doi.org/10.1177/1049732312466294

Citation: Oyebode, J. R., Bradley, P. and Allen, J. L. (2013). Relatives' Experiences of Frontal-Variant Frontotemporal Dementia. Qualitative Health Research. Vol. 23, No. 2, pp. 156-166.

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Qualitative
Health Research

Relatives' Lived Experience of Frontal-variant Frontotemporal Dementia

Journal:	*Qualitative Health Research*
Manuscript ID:	Draft
Manuscript Type:	Research Article
Keywords:	caregivers / caregiving, dementia, families, caregiving, interpretative phenomenological analysis (IPA)

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Abstract

In this paper we examine the lived experiences of people who have a family member with frontal-variant frontotemporal dementia (fvFTD). Our analysis reflects the characteristics of fvFTD and the distinctive challenges of supporting a relative with fvFTD. Emergent themes related to the experience of becoming aware of fvFTD and entering the medical and social care system, coming to terms with a changing relationship, becoming a carer, and 'surviving it'. These caregivers live with specific behavioral challenges and personality changes including loss of empathy, socially embarrassing behaviour and lack of appreciation of risk by the person with fvFTD. The little known nature of the illness leads to lengthy periods of puzzlement and uncertainty for relatives and professional alike, and services cause frustration and distress as well as providing support. Family members adapt through giving up other aspects of life but also develop new skills and qualities of humour and acceptance.

Key words

Dementia; caregivers/caregiving; families, caregiving; interpretative phenomenological analysis.

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Frontotemporal dementia (FTD) is the fourth most common type of dementia affecting older people (Sjögren & Anderson 2006) and accounts for approximately 20% of cases (Snowden *et al.* 2002; Graham & Hodges, 2005). It is the second most common form amongst those under 65 years and is a more common cause of early onset dementia than was previously recognised (Ratnavalli *et al.*, 2002; Rosso *et al.*, 2003). Those with frontal variant FTD (fvFTD) present with changes in personality and behavior; interpersonal difficulties characterised by a lack of empathy or concern for others; disinhibition or other socially inappropriate behaviours; and a general lack of insight and apathy. Due to the prominence of behavioral symptoms fvFTD is also often termed behavioral variant FTD. In addition, the progressive social impairments together with executive deficits, which are also a prominent clinical feature, have led to the use of the term 'social and executive disorder' (SOC/EXEC; Eslinger *et al*., 2007). Some researchers report aggressive, socially disruptive, and antisocial behaviour (Miller *et al.,*1997) and these behavioral problems have been labelled as 'sociopathic' (Mendez *et al.*, 2005).

Dementia health services are currently predominantly focused on those dementias in which memory problems typically present early, hence, for example, the term 'memory clinics', and are also primarily older peoples' services. Although people with fvFTD eventually develop memory difficulties, this is typically much later on, so initially fvFTD is often misdiagnosed as an affective disorder, psychosis, or alcohol abuse (Sjögren & Anderson 2006). The unique and different symptom pattern experienced by a person living with fvFTD as opposed to more common forms of dementia, logically requires qualitatively different interventions or support structures. However, there is relatively little research about fvFTD that does not focus on clinico-pathological assessments or genetic investigations. One small study in Japan, of two people with fvFTD (Kumamoto *et al.*, 2004), looked at the problems that family caregivers encountered. They reported that the very specific behavioural

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3 symptoms typical to people with fvFTD created major problems and a heavy burden for family carers.

More information is needed about the impact on partners, spouses and other family members of those with of fvFTD, given that, at present, services are designed to respond to those whose dementia starts with memory problems. This research takes an in-depth look at the experiences of family members caring for someone with fvFTD. It utilises a qualitative approach with the aim of uncovering a broad and rich array of information that will add to knowledge. It is hoped that information from this study may inform the development of services that specifically respond to issues related to fvFTD.

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Study Design and Methodology

We employed a qualitative approach, carrying out semi-structured interviews that we taped and transcribed verbatim, before carrying out interpretative phenomenological analysis (IPA; Smith *et al.*, 1999). Philosophically underpinning IPA is the view that meaning essentially occurs through understanding subjective experiences. It follows that in order to understand human experience it is necessary to explore the nature of that experience as closely as possible (McLeod, 2001). Using IPA we explored the individuals' views of having a relative with fvFTD and the meanings that they ascribed to this. These meanings together with our subsequent interpretations can be regarded as 'social constructions' rather than objective truths. We aimed to develop insight into the meaning of the experiences or events as a direct result of full immersion or engagement with the text.

Participants

Participants had to be a relative of a person who had received a diagnosis of fvFTD according to the Lund-Manchester criteria (Lund & Manchester Groups, 1994). They also needed to speak and understand English, as the methodology required direct communication and an

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analysis of that communication. In all, we interviewed six family members of people with fvFTD, comprising three husbands, a wife, a daughter and a brother, ranging in age from 23 to 67 years. We have given them pseudonyms in the account that follows.

Procedure

Professionals, in two specialist working age dementia services in the United Kingdom, raised the idea of taking part with their clients' family members. If they were interested the professional gave them introductory information. After a minimum of 24 hours the researcher (author 2) contacted them by telephone to answer any questions, gain consent and arrange to carry out the interview. I then met each participant on a one-to-one basis, though one participant chose to have the person diagnosed with fvFTD present. I arranged the interviews for a place that was convenient for participants, this being the participant's own home for all except one who I interviewed at the care facility in which his relative was resident. I obtained informed consent and brief demographic data and then interviewed the participant in depth about: health and history, diagnosis, living arrangements, day-to-day life, and the effects or consequences of the illness for the person, caregiver and relationship.

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Analysis

Analysis involved four basic stages (Smith *et al.* 1999). First of all we read the transcripts several times and recorded our initial thoughts and observations. Then we identified and labelled the emergent themes with the aim of capturing the essence of the accounts. At the third stage we attempted to create a structure out by looking for connections between these conceptual themes. We subjected each transcript to this process creating a preliminary list of themes (super-ordinate, main, and sub-) for each, with associated quotations. At the final stage we looked across the themes and produced a final super-ordinate structure. At this stage some themes were merged, dropped or raised to a higher level.

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Findings

Twelve salient sub-themes were derived and clustered into four main themes, which were further grouped under two super-ordinate themes. The main headings below reflect the two super-ordinate level and the sub-headings indicate the main themes. The material under each theme heading is grouped to represent each of the sub-themes. These are summarised in table 1.

TABLE 1 ABOUT HERE

Emergence and Realisation

This refers to the participants' experience of becoming aware of the difficulties with their relative, their increasing knowledge of the symptoms associated with fvFTD, and their initial interaction with statutory services.

The opening of the eyes (Becoming aware). All participants were unaware of the existence of fvFTD until their relative was diagnosed. They talked about noticing problems, sometimes many years before finally receiving a diagnosis, and explaining these away, even though they felt there was something that was not quite right. The issues that came to their attention were often minor, and innocuous explanations were brought into play. Mr Jones, for example, said:

> "I mean we were sort of thinking - oh you know, 'Mum's going through the change'. You know - it wasn't that bad you know what I mean… you noticed little subtle things." (Mr Jones.)

It is apparent in the participants' accounts that the changes that worried them embody some of the early signs of fvFTD, being connected for example, with matters as motivation and perseveration, rather than the incidents of forgetfulness that might be more typical in early

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Alzheimer's disease. Mr Jones described how his wife's motivation started to decrease, evidenced in her taking less care of her appearance:

> ...you know how women are - they like to look their best don't they…and she didn't seem bothered too much that way - you know what I mean - she wasn't bothered about her appearance…So things like that started suffering - you know what I mean. (Mr Jones.)

This gradual build up of experiences and incidents culminated in all participants experiencing more major incidents, or 'landmark events', which were so serious or unusual that they were not felt to be normal at all. Mr Mills, for example, described how his brother's driving behaviour and ability to plan and manage his journeys changed:

> But like I say it was the strange behaviour that came after it that really started and the truth is...really... initially... he rung me up one night, after we first realised that something was up, saying, "I need some money." "Oh why, what's the problem?" "Oh, I'm just standing in a petrol station, filled the car up with fuel and I've got no money." And he knew he'd done it and he knew that he hadn't got any money yet he proceeded to fill the car up. So, I sorted that out and then I spoke to my Mum and Dad two days later and he'd done the same again with them. (Pause). So that was strange. (Mr Mills.)

So here, we see a lack of planning, probably caused by executive functioning problems, which is not only affecting the functioning of the person with fvFTD but is also beginning to impact on those around him. Mrs White recounted a story where this became even more

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salient as she feared for her husband's safety as a result of his impaired work standards in the face of an irate customer:

> This guy come over with a gang of his mates - wanted to punch my husband's lights out! Apparently it was a job that he'd done and he should have gone back to repair something, and I kept reminding him... I knew of the job and I kept reminding him and leaving little notes on the wall for him… but when he went out and started another job he forgets and this went on for a good long while – and this guy said "You're f-ing ripping me off!" and this, that, and the other, and he literally come right up to here with Brian. And I was pulling him off, pulling Brian off cos Brian wanted to bash him one! (Mrs White.)

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Not only did she describe feeling threatened but Mrs White was also aware that her husband's behaviour was inappropriate and risky perhaps reflecting a lack of self-awareness. Following the realisation that there was something wrong, participants described seeking information to try and explain these peculiar changes. This involved a search for explanations that eventually brought participants into contact with health and social care services. The unusual nature of the illness presentation and the difficulty making sense of the symptoms, together with their impact on the relationship, seemed to make the early stages of fvFTD a particularly distressing and difficult time for the participants.

The double-edged sword (Entering the system). For most, the search for a way of explaining the changes in their relative took them to an initial assessment with the General Practitioner followed by a referral to more specialist services. However, seeking help form services was not a straightforward process. This experience could be described as a 'double-edged sword', since the participants were in need of information, support and treatment but on entering the

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system, they often found limited knowledge and provision, long waits, uncertainty and lengthy bureaucracy. The contrasting feelings this provoked were: hope versus despair, isolation versus connection, loss of power versus power and uncertainty versus reassurance. There was a sense that in order to receive the benefits of the system, one needs to be at its mercy, giving up power and relying on others – and sometimes this seemed difficult for participants to accept.

For some the journey to the point of diagnosis was long and difficult. Mr Smith and his wife, for example, experienced a misdiagnosis and a period believing that she had Alzheimer's disease, before the diagnosis of fvFTD was given:

> We saw Dr Ahmed several times down at the Psychiatric Hospital. At first he sort of said it was Alzheimer's disease, and, I think for about 18 months, he thought it was Alzheimer's disease. And then the one visit we had with him he said, "I think that I might have got this wrong" and he organised a special sort of scan. I'm not sure what it was called but they injected Maggie with some sort of radioactive material, and then did this scan and then eventually he came to the house and he said, "Right, we have got the diagnosis now!" which was frontal temporal lobe dementia. So that's how we found out what she's got – but it took about 18 months to just over two years before we got there in the end. (Mr Smith)

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Mr Jones discussed how receiving a diagnosis of Pick's disease impacted on him at the time but also how having an explanation for the cause of the problem still affects his thinking now, again demonstrating the bittersweet nature of knowing:

> Well it was the consultant at the hospital, Mr Markum, and I mean he said to me like "Your wife's got Pick's disease." "What's Pick's disease then?" And urm, obviously its frontal lobal dementia and erm, there's no treatment and there's no cure. So like at

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> that point I was, I was in shock! I think you could have just done that [does action] and I'd have fallen over! I just didn't know how to take it – and I think from that point I've sort of been in a, well I don't know how to explain, urm, I mean it's what's been told to me you know, but it's like a sort of, well I can't take it in. Strange innit? (Mr Jones.)

The barriers encountered in trying to access services did not stop with diagnosis. It is almost as if each participant presenting with their relative to services is a test case that requires the service to then develop services around that person. At the time of interviewing, Mr Brown still had not received any social work support, several months after the initial diagnosis. He explained why this had occurred, again highlighting poignantly the double-edged nature of entering services:

> So of course we come back all enthusiastic [from a visit to a good suitable care home] "Yes, yes, yes, we're pleased with this", and then he [consultant] says, "Ah well, I can't get a social worker appointed to her until January." He says, "I could get a local social worker from the area – but they don't do the same things as us – well they're not under our umbrella (. . .), so I would rather ask you to wait until January, if you would, until we get the proper person appointed. (Mr Brown.)

Another barrier experienced by several participants, that caused frustration, was the lack of age-appropriate services. Some participants experienced a dilemma related to envisaging their relative in a care home for the elderly, as Mr Mills puts it:

> We certainly didn't want him in an elderly home, you know. You can imagine it can't you. You can't stick a forty year old in a room full of people like that - through

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> no fault of their own, dementia patients, but for a forty year old to live in an eighty year olds' care home. (Mr Mills.)

Others were rejected by services because they were under the age of 65:

> They've tried to fit Maggie up with another sort of day care centre, and basically the one, she spent the day there you know and they said she was ok, you know, and that she would be acceptable. And then they said, "We can't sorry, she's under the age of 65!" And this is what you find all the time. (Mr Smith.)

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All participants had some experience of research, and described this as giving them a sense of hope, that was often paired with frustration or anxiety. Miss Green, whose relative was eventually offered trial medication, tries to describe her complex thoughts and feelings around this, conveying her gratitude overall for the opportunity to try something:

> She's started some drugs now. That's been another thing, I think in the back of your mind you sort of think that she's gonna get better, and maybe that's what keeps us going. The fact that she's taking these tablets you think that one day she might get better, or she's staying at the same stage because of the tablets (…). We've been told that it won't make her better and that there's only a, I think it was a 20%, chance that it could keep her where she is or slow things down. We couldn't have **not** given her the tablets knowing that there's something out there, you know. We've got to try it haven't we. (Miss Green.)

Once the initial medical and social assessment had been undertaken and a suitable service provided then it seems that participants felt more supported, and stress reduced. This protracted struggle can be contrasted with the experience of those with early Alzheimer's

disease or vascular dementia, who nowadays would be referred to a memory clinic or equivalent as soon as the family doctor realised there were signs of progressive memory impairment.

Life Adjustment and Coping

This theme refers to the pressures placed on the participants to respond and make changes both outwardly and inwardly to the demands of their situation.

The adaptation (Becoming a Carer). In all cases the participants expressed the transition to becoming a caregiver, against the backdrop of the shift in relationship that necessarily occurred as the person with fvFTD functionally declined and exhibited more behaviours that required understanding, management and acceptance. Participants either talked openly about their experience of becoming more distanced in their relationship, or it was evident in more subtle ways. Mrs White explained her experience of losing her connection with her husband quite graphically and emotively, indicating how she was struggling with the change in role. She said:

> I'm a carer, a mother, a nurse. I'm treating him like a child three quarters of the time and then I've got to reverse my roles to be wife again and a lover and one thing and another. Erm I have to keep my eyes closed 'cos if I open my eyes and see his face, I'm seeing this face that isn't him, and it just doesn't feel right to be doing the sex bit. (Mrs White)

In transition from the role of close family member to carer, individuals began to recognise and accept their new status. Mr Jones said that this was brought this home to him in a consultation with his GP:

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> The GP she said, "Have you had your flu jab?" And I said, "Well, no, I'm not eligible am I? You have to be over 65, don't you?" And she said, "Oh no, you're a carer, you can have your jab now" which I did. So you see stuff like that. (Mr Jones)

For some, acceptance of their situation was indicated by the sense that somehow it comes to be experienced as normal, as expressed by Mr Smith:

> She does all these strange things you know. I've become sort of used to it, accustomed to it, and I don't draw attention to it you know. She'll put two night dresses on to go to bed and a work's overall from when she used to go to work, but you don't make a big thing out of these issues, you know. I do tell her, but I don't sort of argue about it – there's not much point. I've learned to sort of cope with these things. Well she says "Well it won't hurt, it's up to me what I wear." So I say, "Ok fine, if that's what you want to go to bed in that's fine" (Mr Smith)

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This degree of acceptance, however, had usually been achieved as a result of adjustment and adaptation in day-to-day life. Part of Mr Brown's adjustment had involved him taking the lead responsibility for domestic chores:

> I cook everything, I think I'm a dab hand now. (Laughs.) I cook everything and weekends I try to do as much housework as possible but it is difficult I mean, I tend, if I've got the downstairs here pretty clean, the upstairs suffers, you know. We have to keep it - it's difficult to do everything. Err, all the washing, all the ironing, I do all that. So, err, yeah, I've adapted! (Mr Brown)

Others talked about how they had had to give up leisure pursuits, facing the reality that they do not have enough time to commit to them. Mr Smith talked about his experience:

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> Most of my time ,when Maggie is here, is taken up with Maggie all day long you know. Well I used to have fish in the garden, I haven't got that no more now. The garden's over run now. I don't get so much time to do what I used to do in the garden. Even in the house you know it's difficult. I used to have big Koi carp out there, big ones, and well I gave everything away about two year ago now – all the fish, all the equipment, and I'd kept fish for about 25 years. I just couldn't get the time to sort of do what I needed to do like to look after them. I just couldn't do it. (Mr Smith)

The issue of employment also featured heavily, some participants taking early retirement, but others not able to afford this and finding themselves juggling work and home care commitments. This is an important area since people with fvFTD are generally of working age and correspondingly their partners and carers also. Mr Jones was able to fortunately take early retirement:

> I was in engineering. So I was like a supervisor in engineering.I'd been with the company for 38 years, so I err obviously I err well somebody had to look after her so I retired early. […] So from doing urm, well sometimes weekend working like shifts and working all my life – all of a sudden just stopping. (Mr Jones)

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Another aspect of life as a carer is the possibility that one may need to develop personal qualities that perhaps had not been so important before. Mrs White, for example, described how she had been able to draw out a sense of humour in order to best manage the difficult situations in which she found herself with her husband:

> Brian's always been the laughey, jokey, barmy type. He's always the clown. He can get everyone laughing you know, and I was always the shy one sitting in the background. […] And I hadn't got a very good sense of humour at all, but I don't

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know how or why I did it but with the things that he was doing or saying and still does now, what choices I said to myself have I got? I'm not a shouty yawpy telly offy type person. I don't like conflict and things like that. I can never belittle him and put him down and I can't keep telling him, "No, you've done that wrong", "No, don't do that." So somehow or another out the blue it come, that if he was doing something that was wrong or whatever, I would turn it round as a laughing joke, and then we'd both end up laughing about it! And I don't know how I started doing it or what gave me the insight to be able do it but that way works best for both of us. (Mrs White)

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Thus this theme draws on the experiences of participants in how they came to accept their new role, by recognising the pressures inherent in it, not only through the loss of their usual way of relating, but also in needing to live with the uncertainties. All participants described how they had made adjustments to adapt to their new role, which may have meant giving things up or developing new skills or qualities. There was also a sense that the situation eventually comes to be experienced as normal.

The maintenance (Surviving it). This theme details participants' methods of coping and surviving from day to day. Participants talked about symptoms and behaviors that they experienced regularly, and the repertoire of techniques and methods that they had learned to manage these.

A major part of caring for someone with fvFTD involves supervision, and Mr Jones described his need to prompt his wife to carry out activities due to a decline in her self-motivation, he said:

Researcher: You sort of prompt her to do things then?

Mr Jones: Yeah. I mean like, "Brush your teeth", "Put the toothpaste on the brush". I hand her the toothbrush and she'll brush her teeth, and I give her a brush to comb her

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> hair, and she'll have a little go at it. But everything is half-hearted you know. She won't do them properly. She just loses interest. There's no drive there. (Mr Jones)

Another reason for supervising the person with fvFTD on a daily basis was related to safety. Miss Green talked about how, in the earlier stages of the disease, her mother had left the house on a few occasions and driven off in the car causing some concern. In response to this the family had increased their supervision of her:

> My Dad can still go and do gardening whilst she wanders around the house as long as the doors are locked and the keys are hidden. Then he can still do things like that but obviously you've got to be with her all the time. You couldn't, sort of, you know we couldn't leave her in here and go shopping, you know cos you've got to, cos she has got to have somebody with her all the time. (Miss Green)

Mrs White talked about how she had needed to develop an anticipatory strategy while carrying out daily tasks and activities in order to avoid mishaps:

> And the clumsiness he's had that right from the start but it's getting a darn site worse now. It's like having a child – mmm (. . .). I know it's not big but if he's going to get a glass of water and I'm doing a slice of toast, and we're both coming the same way then I have to side step, I have to watch, you know. I have to keep out of his way you know, cos he'll have me over. If he's got a cup of tea I have to be very careful because he'd spill the cup of tea, and it could go on me (. . .). You have to anticipate what's going to happen, yeah, you do have to have forethought exactly. (Mrs White)

Both Mr Mills and Mrs White talked about how they had to employ a certain amount of manipulation, as in Mrs White's account:

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> "I'm having a bike! I don't care what you say, I'm getting a bike!" And, oh my god a bloody bike! I mean he's bad enough in a car, a bike's gonna be even worse and I thought, "Oh well he's put his foot down now and he's getting stroppy, let him get it". I can always find ways of wheedling round him, you know "Don't go out on it you know it's raining" or "John's gonna pick you up" or "Your son's gonna pick you up or..." So all right yeah he went and got this bike and he got the crash hat. I was just dreading the first time he went out on it. I watched him going down the road on it like this, and I was just like, "Oh my god!" So erm the next time he went out on it, I kept making excuses from then on in, any excuse I could think of. Whenever he said he was gonna use the bike I would come up with a better answer for why he shouldn't. I can't remember all the little lies I told him but fingers crossed and touch wood he hasn't been on it for a month or two now. (Mrs White)

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In addition to managing the supervision of the person with fvFTD on a daily basis, participants spoke of taking some responsibility for others' emotions. Mrs White described trying to manage her children's experience of their father:

> To start with when he first got bad I was telling them most of the progress. Like, we've seen a doctor and they've said he should have that test, and then I'll say, "Ooh your dad wasn't very good today, erm he was erm forgetting more words" or, "He was breaking things up", thinking that it's their dad and they should be knowing, you know. But I've stopped doing that now. My daughter especially, you know, "Every time you say anything to me it's doom and gloom!". (. . .) I know they love us but especially me daughter she's a business woman, very very busy (. . .). It's obvious she doesn't want to know. She says, "You'll tell me anything I need to know" and that's it. (Mrs White)

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Similarly, Mr Mills felt responsible for his parents' emotional experiences:

> I come down and have meetings with the staff and erm, I convey the facts that I get, in a watered down form to my Mum and Dad. I try to protect them as much as I can. I protect them really as much as I can because they don't need to know the ins and outs of what John's doing. (Mr Mills)

Participants were also aware of the need to manage situations with friends who may not have any notion of fvFTD and the way it impacts upon a person. Mr Smith described his wife's behavior:

> When I'm out and about with her now, because she's so jolly and she looks physically well, people always say, "Well she looks fine to me, there doesn't look anything wrong with her" and I says "Well it doesn't show on the outside you know." So err, when they say to her, "How are you Maggie?" and then all of a sudden she grabs hold of them, you know, by the shoulders, in a friendly sort of way. But they're a bit sort of, "Well what's happening here", sort of thing! And she'll say to them (and she'll say the same thing over and over again you know), "What I say" she says "Carry on regardless! Look on the bright side!" So they all say "That's right Maggie - that's right!" But it's when she grabs hold of them you know. She would never do this before you know, but now it could be a complete stranger. (Mr Smith)

In this story Maggie's behaviour showed some lack of understanding about other people's feelings, and diminished awareness of personal space, which may be a result of deterioration in her understanding of theory of mind and indicates a lack of empathy or understanding on her part, which then has to be smoothed over with others.

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This sub-theme also includes the carers managing the impact of fvFTD on themselves. On the whole participants seemed able to recognise their own emotional state and difficult feelings related to their situation. When asked about the most difficult aspect of his life at the time of the interview Mr Jones said:

> Looking back. If I do - then that brings me down too much. But if I just take each day and just work with what you've got then it seems alright. But occasionally, like if you're on your own, sometimes I start to look back and that's difficult because then you can actually see how far you've come. And that's, well I don't like doing that really too much you know, looking at photographs if you like or, well I wouldn't say that I don't like doing it, but it depends. I know it's not going to do my morale that much good really. (Mr Jones)

In order to manage their emotions, participants seemed to have developed some strategies. Not dwelling on the problems and challenges that they encounter, and getting on with daily life, seemed to be a prominent method. Miss Green said:

> And like me and my Mum were really close. We used to do a lot of things together. Um, and it sort of meant we stopped all that because we couldn't really do those things anymore. You know. Now it's me who's taking my Mum out, you know, we're not going out together, you know I'm like, I'm like, looking after her you know it's difficult, but you just have to get on with things. (Miss Green)

Developing a sense of humour also seemed important for some. Mrs White's development of a sense of humour has already been documented, there follows an example:

> We were going into a supermarket shopping the other week and just as we were going into the door he'll think of summat, erm, and he'll say erm, "Underspray!" Right. "I

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> need underspray, I want underspray, I need underspray!" And then cos, well, it's hurting in here, but we joke. I've learned to get a sense of humour which I never had before ,and I say "What you on about you daft sod, underspray?" "Is it underspray for the car? Is it underspray for the bed?" or well you know I was being barmey you know. No, no, and he got it then you know, under ARM spray. (Mrs White)

Finally, another way of managing emotions seemed to be for participants to see themselves as being fortunate by comparing themselves to others. For example Mr Jones talked about another family who had a genetic form of FTD:

> But I mean their plight was far worse than ours really, you know. Cos she had lost her mum when she was something like about 4 years old, and because her family was young, her sister had to look after them and she later died of the Pick's disease. And like then her brother's just gone as well, and I thought, "Well that's devastating that isn't it." So straight away, I mean, my situation is nothing compared to hers. (Mr Jones)

On a more practical level, participants talked about a range of activities they could draw on to help them recharge their batteries or keep them motivated. Mrs White was thankful for being able to retire to a nearby holiday caravan each weekend to unwind with her husband, and Mr Smith talked about music being a great stress reliever:

> I've always said that I love me music, you know. I always have done. All sorts of music you know. Many many times I still do, I did last night. Err, when I'm feeling sort of really stressed out, I'll say to her "I'm going to leave you watching television" and I sit where you are now and I put me headphones on, put a CD on and listen to it,

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> and that way I feel myself coming down. Yeah, I find music a great stress reliever. (Mr Smith)

Time away from the person with fvFTD also featured prominently. Mr Smith had been able to benefit from respite care for his wife but he described it at first as being a difficult adjustment to make, having spent many years with his wife:

> I mean when she first started going in the first couple of times I found it quite stressful myself you know, cos we've been married for such a long time, you know, and all of a sudden that person is, as I say, the way she is terrible at times you still miss her. But the last time she went in it didn't bother me at all and I felt really relaxed last time. Saw my brothers, and they phones and says "Well, we'll come and pick you up, spend the day over with us" like. So they came, took me over to their area and we'd go and have a drink and a pub lunch, something like that, which is something I didn't really get to do, and have a walk around somewhere, like you know, and I haven't got to worry about getting back. Yeah, it was really relaxing the last time she was in hmm yeah. (Mr Smith)

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Finally receiving support from others seemed important, particularly from other family members. However there was a mixed response from participants on this issue. While some reported difficulties and distancing within family relationships, others reported experiencing good support. Mr Brown was pleased with the help that he had received from his own children, and his wife's from a previous relationship:

> He's the eldest and he's been very supportive, hasn't he? He tries to come down every weekend. At one time he used to come midweek from work but of course it's such a long distance to come. So I think he couldn't keep that up. He usually invites us up

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> on a weekend or he comes down at the weekend. So yeah. And the other one, he comes, err, fairly regularly, our Michael, he will keep, he always phones anyway to see how you are. He's always there if you need any help anyway and, and err, his wife, err, she, Mary, she err, she takes you to have yer hair done, don't she, every month? And she's gonna take you to have yer manicure soon, come Christmas. So they're very supportive. Yeah, yeah. And my own son and his girlfriend err, they took her for a meal the other day, they're supportive as well aren't they? So, the family's being supportive. (Mr Brown)

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Discussion

In this study we have explored family members' experiences in caring for someone with fvFTD. Four main themes have emerged from the data, each containing examples of behaviors that are very distinctively related to fvFTD. All participants had taken on the role of caregiver (except 'Mr Mills' whose brother was in a specialist unit) and were potentially subject to carer stress. They highlighted the many symptoms associated with fvFTD, which create risk and social embarrassment. Participants' individuality, the nature of the relationship and the availability of support, seemed to affect stress levels. Adams *et al.* (2008) proposed a model that places relationship factors at the centre of the stress process. They stated that the loss of intimate exchange, the change in the quality of the relationship, and an associated loss of 'sense of self' has a pervasive and important effect on caring for a loved one with dementia, and that this may exacerbate carer burden. This seems especially salient in this study given the specific nature of fvFTD and the impact reported by all participants of decline in the quality of their relationships from an early stage.

Leventhal *et al.'s* (1984) self regulatory model (SRM) attempts to describe and explain how people represent and respond to health threats. It assumes that when faced with

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an illness people are motivated to define and control it. Central to this model is the idea that an individual actively constructs a cognitive representation of the health threat and regulates their coping accordingly. The SRM has been applied to understanding a wide range of chronic health conditions in recent years (Hale *et al,*. 2007) including dementia (Clare *et al.,* 2006), and has helped in understanding people's responses to the illness and developing suitable interventions. We think that the SRM could potentially be used to evaluate how family caregivers of people with fvFTD construe the condition, and how this relates to their coping strategies and well being. Particularly salient in the case of fvFTD, is the relative rarity of the condition and the lack of knowledge and awareness both in professional and lay circles. Carers have no reference point or information to guide their illness representations and hence their coping responses. This creates an extensive period of time prior to diagnosis in relative ignorance; a time in which they experience a sense of helplessness, frustration, and essentially an increased burden of care. Linked to the ignorance of fvFTD, we also saw many examples of services proving to be a double-edged sword as their benefits were balanced with deficits. Wuest and Hodgkins (2011), summarising key points from two decades of their own research on caregiving, put forward a theory of precarious ordering, in which they express how important it is that care is 'connected' (i.e. timely and appropriate) rather than 'disconnected'. The experiences of our participants seemed to be of finding services to be helpful and unhelpful both at the same time, with the unhelpful aspects being seen as an almost inevitable corollary of gaining the beneficial aspects. Thus, to improve services, we may need to look at how to minimise unintended psychosocial costs that come as inherent side-effects.

We also saw in this study, how family members adapt to the challenging changes wrought in their lives by the development of fvFTD in their relative. Two of our themes, The Adaptation (Becoming a carer) and The Maintenance (Surviving it) seem to echo those found

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by Healey-Ogden and Austin (2011) in their recent account of the lived experience of well-being in 40-60 year-olds in which they found that turning towards a new identity and finding spaces for nature and for play seem to be central to gaining a sense of well-being.

This study highlights the need for improving dementia care services specifically to tackle the idiosyncratic symptoms and behaviours evidenced in people with fvFTD. Initiatives to raise awareness of fvFTD could be directed at primary care workers initially and clearer care pathways could be developed, to reduce frustration and distress. Interventions could also be developed and evaluated, including approaches based on neuropsychological rehabilitation, those aimed at bolstering family caregivers' efficacy and self-confidence and those which help carers to increase the use of emotion-focussed coping (e.g. acceptance). Finally, mental health care policies should be designed to promote equality of access and avoid age-segregated services that may unfairly limit access to effective and appropriate specialist care.

Our study has some limitations in having a small and varied sample of participants, although they were united in their experience of having a relative with fvFTD and the small number allowed for deep idiographic analysis. Being from the United Kingdom, the service and family context differs to some extent from that in the USA and other countries. There may also be biases in our interpretations. Excerpts of the transcripts and initial analyses were discussed between all three authors, and we have also shared our themes and conclusions with wider audiences of staff who work with people with FTD in order to check for plausibility. The results are not generalisable but offer a valuable insight into a sensitive and little known subject area, grounded in the participants' own words.

Through the presentation of an organised and coherent structure we hope to shed light on the issues facing family caregivers of people with fvFTD. However, it must be stressed that our intention was not to develop a linear model of becoming a carer. Whilst there are

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some aspects that might logically follow on from others, the experience of discovering that your relative has fvFTD is neither straightforward nor organised. It would be more fitting to consider the themes highlighted as occurring simultaneously, and dependent on individual circumstances. Future research could be designed to further consider burden of care issues in response to the specific symptom profile of fvFTD. It may also be valuable to examine more closely the underlying mechanisms effecting the changes in relating (e.g. in social cognition) and to draw out the process of acceptance in family caregivers in response to this loss of relationship.

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Table 1: Summary of super-ordinate themes, main and sub-themes

Super-ordinate themes	Main Themes	Sub-themes
Emergence & Realisation	'The opening of the eyes' (Becoming aware)	1. Noticing changes: What raised suspicion... 2. Recounting landmark stories: What was serious... 3. Understanding: What might be going on...
	'The double-edged sword' (Entering the system)	4. Getting labelled: What it is... 5. Getting help: What is out there... 6. Getting researched: What might help...
Life Adjustment & Coping	'The adaptation' (Becoming a carer)	7. Reassessing relationship: What is lost... 8. Accepting: What is and what could be... 9. Readjusting: What needs to change...
	'The maintenance' (Surviving it)	10. Managing daily life: What needs to be done... 11. Managing other's emotions: What they feel... 12. Managing self: What about me...

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