

VOLUME I  
RESEARCH COMPONENT

by

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A thesis submitted to the University of Birmingham as partial fulfilment of  
the registration for the degree of Doctorate in Clinical Psychology

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## Overview

This thesis is submitted in partial fulfilment of the requirements for the degree of Doctorate in Clinical Psychology (D.Clin.Psy.) at the School of Psychology, University of Birmingham, UK. It comprises two volumes. Volume I consists of the research component whilst Volume II comprises the written clinical component based on work completed during training.

Volume I comprises two papers; a literature review and an empirical research paper. The literature review is a systematic review of the evidence for the efficacy of an errorless learning based strategy in aiding learning in people with Alzheimer's disease. The empirical research paper is a qualitative exploration of people's experiences of living with a diagnosis of behavioural-variant Frontal Temporal Dementia (bvFTD).

Volume II contains the five Clinical Practice Reports (CPR) completed during training. They are representative of work carried out whilst on placement within the Adult Forensic Service, Learning Disabilities, Older Adult, Child and Adolescent Mental Health and Working Age Dementia specialities. CPR1 presents cognitive behavioural and psychodynamic formulations in respect of an adult within a medium secure unit. He was referred due to concerns about violence towards NHS staff when he was acutely unwell. CPR2 was a service evaluation which sought to quantify the frequency and intensity of between-patient bullying and quantified patients' attitudes towards bullying behaviour and their social environment. Recommendations were made to help inform policy and to help ensure that the service manages the risks associated

with between-patient bullying in an effective way. CPR 3 presents the case of 32 year old male with a severe learning disability, communication difficulties and a sensory impairment who was referred due to an increased level of challenging behaviour. A single-case experimental design was used to evaluate the behavioural intervention. CPR 4 was a case study which reported the case of a 2 <sup>1/2</sup> year old girl with behavioural difficulties. Here the assessment, formulation and intervention were informed by the Solihull Approach (The Solihull Care Trust, 2006). CPR 5 was a dementia assessment conducted with a 64 year old lady reporting disorientation and forgetfulness. This CPR was presented orally and the abstract is included for reference.

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## LITERATURE REVIEW

IS AN ERRORLESS LEARNING BASED STRATEGY EFFICACIOUS  
IN AIDING LEARNING IN PEOPLE WITH ALZHEIMER'S  
DISEASE? : A SYSTEMATIC REVIEW

Word count: 8189

## **Abstract**

Developing interventions which support people with Alzheimer's disease (AD) is important in terms of promoting their independence and quality of life in line with person-centred approaches to care. Cognitive rehabilitation seeks to meet these aims by using individually tailored interventions aimed at addressing specific, personally relevant areas of difficulty, often by helping people to learn new skills, or re-learn previously held skills which have been lost. To do this appropriate teaching methods must be identified. Errorless learning (EL) seeks to eliminate, or at least minimise, errors during learning and has been purported to be an efficacious method for teaching people with AD. Therefore, this review systematically reviewed the evidence for this, by examining the evidence published from 2002 until July 2012. A systematic literature search was undertaken to identify all potentially relevant studies which were then reviewed against specific pre-defined inclusion and exclusion criteria. The studies identified were reviewed in a uniform way and assessed using appropriate quality assessment criteria. It was concluded that there was evidence for the efficacy of EL over trial and error learning methods for those with mild/moderate AD; but no clear evidence for its effectiveness over other effortful learning strategies. There was some evidence in favour of EL for procedural tasks in those with more severe levels of impairment. Limitations in the evidence base were identified and suggestions for further research were made. Limitations of the current review were also discussed.

## **Introduction**

Alzheimer's disease (AD) is the most commonly diagnosed form of dementia within the UK accounting for 60% of all diagnoses made (Alzheimer's Society, 2012). It is a neurological condition which results in a progressive loss of cognitive abilities and for which there is currently no cure (Buschert, Bokde, & Hampel, 2010; Thivierge, Simard, Jean, & Grandmaison, 2008). AD is characterised by an initial deficit in explicit or declarative memory, with implicit or non-declarative memory being relatively well preserved (Squires, Clarke, & Bayley, 2004; Squires, Stark, & Clark, 2004). This fits with the early neuropathology of AD which is focused on atrophy in the medial temporal lobe including both the hippocampus and entorhinal cortex (Braak & Braak, 1995); brain regions which are important for encoding and storing new memories (Squires et al., 2004). As the disease progresses this neuropathology spreads to neocortical areas including the parietal and frontal cortices (Braak & Braak, 1995) and this results in other areas of cognitive decline such as executive functioning, attention and visual-spatial abilities (Salmon & Bondi, 2009).

Whilst, in the earlier stages of the disease, a decline in cognitive abilities is typically the most prominent change observed, AD is also associated with a decline in global functioning, e.g. the ability to carry out everyday tasks such as managing finances and self-care (Bucks, Ashworth, Wilcock, & Siegfried, 1996). Indeed, to make a diagnosis of dementia there must be evidence that the decline in cognitive function interferes with the individual's ability to undertake their usual activities (McKhann et al., 2011). Often this loss of functional abilities results in people requiring assistance from statutory services. The cost to the NHS and Social Services

of caring for people with dementia is high being estimated at £1.17 billion and £2.13 billion respectively in 2007 (The National Audit Office, 2007). Furthermore, this cost is expected to rise significantly since, although dementia can occur throughout adulthood, the biggest risk factor for developing dementia is ageing with estimated prevalence rates of 1 in 6 in those aged over 80 (Alzheimer's Society, 2012). Given that people are tending to live longer, it is expected that the number of people being diagnosed with dementia and therefore AD will increase, and it is estimated that there will be one million people living with dementia in the UK by 2021 (Alzheimer's Society, 2012). Hence developing interventions which support people with AD is of fundamental importance in terms of promoting their independence and quality of life in line with person-centred approaches to care (e.g. Kitwood, 1997). One type of intervention that seeks to meet these aims is cognitive rehabilitation (Clare, 2007).

Within the research literature the terms cognitive rehabilitation, cognitive training and cognitive stimulation are sometimes used interchangeably. However, there are differences in the aims and outcomes that each of these types of intervention seeks to achieve (Clare & Woods, 2008). Cognitive rehabilitation, which is the focus of this review, involves individually tailored interventions aimed at addressing specific, personally relevant areas of difficulty which are related to day-to-day activities and the individual's level of cognitive impairment (Clare, 2007; Clare & Woods, 2003). So, here the aim is specificity in terms of achieving the target of the intervention thus supporting independence and well-being, rather than seeking to achieve a generalised improvement in one or more areas of cognitive functioning. Therefore in studies of cognitive rehabilitation, outcome measures are tailored to the specific intervention used, e.g. if relearning steps to complete an instrumental activity of daily living (IADL) was the goal for intervention, an appropriate outcome measure

would be the number of steps correctly completed post-intervention compared to pre-intervention. In comparison, cognitive stimulation refers to a standardised group-based intervention which seeks to improve an individual's overall level of cognitive functioning (Woods, Aguire, Spector, & Orrell, 2012). In a similar vein, cognitive training uses standardised tasks with the aim of improving specific area(s) of cognitive functioning, either on a group or individual basis (Clare & Woods, 2008). For both these approaches, outcomes are measured using standardised neuropsychological tests and the aim is that the intervention will generalise to situations not directly linked to the intervention provided (Clare & Woods, 2008; Woods et al., 2012).

Whether the intention is to teach new or previously held skills, it is important to choose the most appropriate teaching method. In healthy individuals, who are able to make effective use of episodic memory, trial and error or effortful learning (EF) is effective since the active processing of information supports both encoding and subsequent retrieval from memory (Middleton & Schwartz, 2012). However, it has been hypothesised that this may not be the most effective learning method for people with specific deficits in episodic memory such as those with AD. Here, it has been purported that techniques which support implicit memory may be more efficacious since explicit and implicit memory processes are underpinned by different brain regions; with implicit memory being relatively preserved in AD (Squires et al., 2004).

Errorless Learning (EL) seeks to eliminate or, at least minimise, errors during learning (DeVreese, Neri, Fioravanti, Belloi, & Zanetti, 2001; Ehlhardt et al., 2008; Grandmaison & Simard, 2003). Developed from the animal learning literature (Skinner, 1965; Terrace, 1963), EL is based on the premise that eliminating or minimising errors during learning, decreases competing memory traces thereby

increasing the likelihood that a correct response will be accessed when the information is subsequently retrieved (Clare & Jones, 2008; Haslam, Moss, & Hodder, 2010). Two explanations for this have been proposed: first, those with episodic memory deficits rely on implicit memory processes which typically occur outside of awareness based on the strength of the memory trace without specific recollection of the learning context. This means that errors during learning will tend to be replicated with the effect increasing each time the error is produced (Anderson & Craik, 2006; Baddeley & Wilson, 1994). Hence, reduce training errors and the strongest memory trace would be the correct response. The second explanation purports that EL supports residual episodic memory since those with episodic memory difficulties are particularly susceptible to repeating previous errors since their ability to monitor errors is compromised, i.e. they struggle to learn that a response was erroneous (e.g. Tailby & Haslam, 2003). Therefore eliminating, or minimising, errors during learning supports their remaining explicit memory resources since they only have to remember their previous response without the added complication of having to remember the feedback associated with that response.

Systematic reviews aim to identify all studies which are relevant to the research question posed, using specific pre-defined criteria, applied in a uniform manner which can be readily replicated (Centre for Reviews and Dissemination, 2008). A search for review papers was conducted as described in the method section which follows. Two systematic reviews were identified that considered cognitive rehabilitation techniques (including EL) in people with AD, both of which included papers published to the end of 2001 (DeVreese et al., 2001; Grandmaison & Simard, 2003). DeVreese et al. (2001) concluded that cognitive rehabilitation techniques for people with AD showed promise, but did not differentiate between different

techniques in terms of efficacy whilst Grandmaison and Simard (2003) concluded that that the EL, spaced retrieval, and vanishing cues either used alone or in combination were effective intervention methods for people with AD.

Four more recent review papers were identified, two of which were selective non-systematic reviews. One purported to review the efficacy of EL in mild AD yet appeared more conceptual in nature and reviewed only four studies before concluding that the evidence for the efficacy of EL was inconclusive (Mimura & Komatsu, 2007). The second appraised the use of EL in neurological rehabilitation settings. However, there was no systematic search for relevant literature (Middleton & Schwartz, 2012). Instead, they selected literature to argue that retrieval practice from long-term memory rather than the avoidance of errors was the most important determinant of learning. Furthermore, they hypothesised that methods of teaching that concentrated on schedules of retrieval rather than extended study of materials would produce more efficacious results, and suggested the exploration of this as a direction for future research. Since neither review systematically searched for relevant literature, it is difficult to ascertain the completeness of the evidence presented (American Academy of Neurology, 2004). Furthermore, neither review commented on the quality of the studies cited so it is not possible to judge the reliability of the findings which may have been influenced by factors such as selection bias, or the use of different methodologies (Centre for Reviews and Dissemination, 2008). Whilst one systematic review of EL in neurological populations was published in 2008 (Ehlhardt et al., 2008) and included four studies contained within the current review, the focus of the previous review was across multiple areas of cognitive rehabilitation and did not specifically consider issues pertinent to people with AD. They recommended using an EL strategy when helping people with acquired memory impairments learn or relearn

information. In contrast, the final review identified was a critical review of EL in rehabilitation for memory impairments published in 2008 which concluded that both EL and EF were effective learning methods for those with mild AD with the caveat that due to the heterogeneity of the population, clinicians should consider the learning goal and the individual's learning preferences when planning interventions (Clare & Jones, 2008). Here, whilst the search for papers was conducted systematically, the search strategy was restricted to group studies which directly compared EL with an alternative learning technique rather than single case experimental designs or studies which did not directly compare EL with another learning approach. Also, the quality of the studies reviewed was not systematically appraised. There was an overlap of two studies between Clare and Jones (2008) and this review.

Therefore, this review was conducted with the aim of systematically reviewing the evidence for the efficacy of EL as a cognitive rehabilitation technique for people with AD. Since, as discussed, the last identified systematic reviews which focussed on the efficacy of EL for people with AD included papers published until the end of 2001; this review considered studies published between 2002 and July 2012.

## **Review Method**

### *Identification of studies*

First, to identify whether any systematic reviews had already been conducted the Cochrane reviews database was searched using the search term "Cognitive Rehabilitation". One relevant review written by Clare and Woods (2008) was identified. This review reported no randomized controlled trials (RCTs) of cognitive rehabilitation in people with either Alzheimer's disease (AD) or vascular dementia



(VcD). Additionally, three relevant databases were identified: PsycInfo, Medline and Web of Science and each database was searched independently for systematic reviews. The reason for conducting separate rather than combined searches was that PsycInfo and Medline have different structures for hierarchical mapping of search terms, whilst Web of Science utilises free text search only. Therefore, when conducting combined searches the ability to map onto related search terms is unavailable hence increasing the risk of omitting target papers.

Searches of the three identified databases were restricted to papers published in the English language, and reviews or systematic reviews with no date limits. In respect of PsycInfo the following expanded search terms: “Cognitive rehabilitation”, “Alzheimer’s disease” and the free text search terms of “errorless learning”, “cognitive rehab\*”, and “Alzheimer\*” were used (See Appendix 1 for detailed search strategy). For Medline the expanded search terms used were: “learning”, “Alzheimer disease”, and “Dementia, vascular”; together with the free text search terms of “errorless learning”, “learn\*” “rehab\*”, and “Alzheimer\*” (See Appendix 2 for detailed search strategy). The search of the Web of Science database was conducted utilising the following free text search terms: “cognitive rehab\$”, “Alzheimer disease” and “rehab\*” (for detailed search strategy see Appendix 3). After de-duplication of results across the three databases, six reviews were identified. Full texts were obtained and scrutinised. Only two reviews systematically evaluated studies which investigated the efficacy of errorless learning in people with AD based on a literature search to the end of 2001 (DeVreese et al., 2001; Grandmaison & Simard, 2003). Therefore the current systematic review considered studies published from 2002 onwards.

Having conducted a systematic search for other systematic reviews and, as part of this process, established suitable date parameters for the current review, a systematic search for papers was undertaken. The searches of the three databases (PsycInfo, Medline and Web of Science) were re-run using the same search parameters as described above but with two differences; the date range was restricted to studies published from 2002 onwards, and the results restricted to non-review studies (for detailed search strategies see Appendices 4-6).

### *Study selection*

Ninety-three studies were initially identified across the three databases. After de-duplication of the search results, the resulting 73 studies were reviewed independently by two researchers to identify those which met the inclusion/exclusion criteria for the current review which were:

#### Inclusion Criteria

- Participants had a diagnosis of AD or mixed dementia (AD plus VcD).
- Studies that were identified during the literature search as appertaining to EL in that, from the abstracts, they reported to have evaluated the efficacy of EL techniques either as the sole intervention method, or augmented with other cognitive rehabilitation techniques providing the elements of the intervention were clearly described; or studies that evaluated the efficacy of EL techniques (with or without augmentation of other cognitive rehabilitation techniques) by direct comparison of outcomes with other clearly defined cognitive rehabilitation methods.
- Studies that investigated learning a specific task or procedure.
- Studies that reported original data whether it be pooled-group data or single case studies.

- Studies written in the English language.
- Studies published from 2002 onwards in peer-reviewed journals.

#### Exclusion Criteria

- Review papers which did not discuss original data.
- Studies which investigate using EL in cognitive training with the aim of increasing overall cognitive functioning in one, or across several, cognitive domains.

The rationale for including studies with people with a diagnosis of mixed dementia of AD plus VcD was that those with this diagnosis would be expected to exhibit the primary episodic memory deficit associated with the early cortical degeneration seen in the initial stages of AD (Braak & Braak, 1995). In comparison, a diagnosis of VcD can be associated with cortical, or sub-cortical and/or frontal changes (Lewy & Chelune, 2007) which may not result in an episodic memory deficit. This would make the applicability of EL debatable and make comparisons across studies difficult. Thus studies which included participants with VcD were excluded. The search criteria were restricted to peer reviewed journals due to the difficulty in obtaining sufficient details on studies presented as conferences abstracts or theses. Difficulties in obtaining accurate, timely translations were the reasons for restricting studies to those published in English.

The process of screening the 73 identified studies for their fit with the inclusion and exclusion criteria is depicted in Figure 1. The author and another researcher independently generated lists of studies that met inclusion criteria. The lists were compared and there was a 100% concordance rate. Fourteen studies were

identified. The references lists of these studies were scrutinized for other studies which might be eligible for inclusion and three were identified. These studies were reviewed by the two researchers independently and there was a concordance rate of 100% that none of the three studies met the inclusion criteria. Therefore no extra studies were included. Additionally, for the journals from which studies had been identified, articles in press were scrutinized. No additional studies were identified (see Appendix 7 for a list of journals scrutinised). Finally, for the papers identified, the names of authors were scrutinised and, in respect of any author who had three or more publications (Linda Clare and Barbara A Wilson), a further search was conducted using Primo Super-search, a facility which searches across databases. The searches were conducted independently on each author's name with the subject topic of "cognitive rehabilitation". No additional papers were identified. Therefore, a total of 14 papers were included in this review.

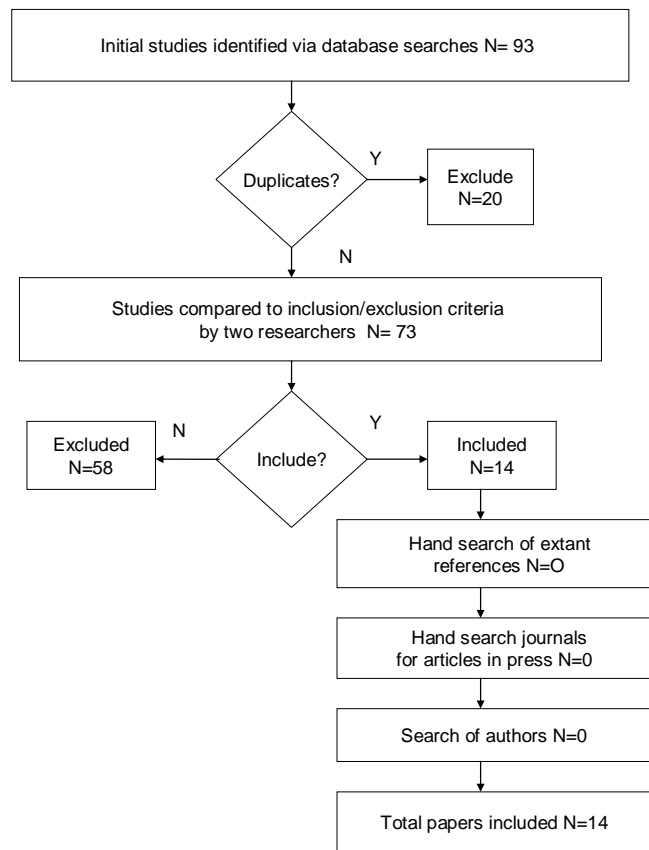


Figure 1: Process for systematically identifying studies for inclusion

### *Data extraction*

To ensure that data from each study were systematically collated and sufficient to undertake the quality assessment and data synthesis, a data extraction form was developed. This was based on the recommendations of the American Neurological Association (American Academy of Neurology, 2004) and the Centre for Reviews and Dissemination (CRD) (Centre for Reviews and Dissemination, 2008) and tailored to fit the requirements of the topic under review. The tailoring process was two-fold: First, to tailor generic headings and make them more specific, e.g. by specifying the particular interventions used in each study and second, to ensure the data extraction form contained the specific information required to complete the

chosen quality assessment criteria, e.g. by recording the setting where the intervention, or interventions, were undertaken. For a copy of the data extraction form see Appendix 8.

### *Quality assessment*

To assist in conducting the quality assessment a search for relevant, valid quality assessment tools was undertaken. During the search process two kinds of studies were identified. These were group studies and studies with a small n, which were either single-case experimental design (SCED) studies or case studies. For this reason two separate quality grids were sought. Starting with group studies, a number of frameworks were reviewed (e.g. American Academy of Neurology, 2004; Caldwell, Henshaw, & Taylor, 2005; Centre for Reviews and Dissemination, 2008) and a systematic review of quality assessment tools used to assess non-randomised controlled trials was identified (Deeks et al., 2003). Deeks et al. (2003) reviewed 213 quality assessment tools against six main criteria with a particular emphasis on issues arising due to lack of randomisation. They identified six quality assessment tools which scored highly across at least five of the six domains they had identified; three of these were reviewed since they were specifically developed to assess the quality of intervention studies (Cowley, 1995; Downs & Black, 1998; Reisch, Tyson, & Mize, 1989). The tool by Downs and Black (1998) was adopted since it had good inter-rater reliability (.75) and test-retest reliability (.99) (For a copy see Appendix 9).

Even when using validated quality assessment tools, adjustments may be required to meet the objectives of specific reviews (American Academy of Neurology, 2004; Centre for Reviews and Dissemination, 2008; Deeks et al., 2003). Therefore, the Downs and Black (1998) checklist was reviewed and adjusted as appropriate, e.g. some of the questions were removed because they were not applicable given the methodologies adopted by the studies under review. For full details of the adjustments made see Appendix 10.

Turning to the SCED and case studies, two quality assessment tools were consulted (Logan, Hickman, Harris, & Heriza, 2008; Tate et al., 2008) The assessment tool of Tate et al. (2008) was chosen since it is brief, had good inter-rater reliability (.88) and content validity based on piloting using 85 published empirical papers.

#### *Piloting of data extraction form and quality assessment tools*

The reasons for piloting the data extraction forms and quality assessment tools were two-fold; first, to ensure that the data extraction forms were sufficient to complete the quality assessments and, second to provide a measure of inter-rater reliability in respect of how the quality of the reviewed papers was assessed. Three papers were chosen at random. The author and another reviewer completed the data extraction forms for the three studies and then completed the relevant quality grid without referring back to the paper. Both reviewers agreed that the data extraction form contained sufficient details to complete the quality grid. The quality ratings ascribed to each item on the quality grid were compared. Inter-rater reliability was calculated using the kappa statistic; chosen because it provides a more robust measure than simple percentage agreements (Uebersax, 1987). The resulting kappa statistic

( $k=0.89$ , 95%CI 0.80-0.97) suggested an “almost perfect” level of inter-rater agreement (Landis & Koch, 1977). Initial disagreements were settled by discussion.

### *Data synthesis*

This systematic review involved the narrative synthesis of the studies under review. This methodology was chosen rather than a meta-analysis because of the methodological diversity of the studies which would render the pooling of the results quantitatively inappropriate (Centre for Reviews and Dissemination, 2008) .

## **Results**

For ease of reference, an overview of the standard techniques used in each cognitive rehabilitation intervention described herein is given in Table 1, using the example of learning face-name associations for illustrative purposes. Except where noted, in the summary and narrative synthesis that follows, these were the methodologies adopted within each of the studies described.



Table 1: Standard techniques for each cognitive rehabilitation intervention

Intervention	Technique
Errorless learning (EL)	A face is presented together with the associated name during learning trials. The participant is asked to repeat or write down the answer to encourage correct encoding. At recall, participants are specifically encouraged not to guess to prevent retrieval errors.
Vanishing cues (VC)	Initially the face is presented with the full name and over repeated presentation trials the letters of the name are gradually reduced e.g. DAVID, DAVI_, DAV_ _ , DA_ _ _ , D_ _ _ _
Vanishing cues with forward chaining (VCfc)	The face is presented with the first letter of the name. If the participant responds incorrectly, a further letter is presented until the correct name response is given e.g. D_ _ _ _ , DA_ _ _ , DAV_ _ , DAVI_ , DAVID.
Spaced rehearsal (SR)	Participants are presented with the face and name. Then, after a very short time frame (e.g. 10 seconds), the participant is shown the face alone and asked to generate the name. If the name is correctly generated, the process is repeated doubling the time-interval at each repetition. If an error is made, it is immediately corrected and the time delay halved for the next rehearsal trial.
Mnemonics (MN)	Participants are presented with the face and name. They are encouraged to develop an association between the face and the name to aid encoding and subsequent recall.
Effortful learning (EF)	Often referred to as trial and error learning. Participants are encouraged to guess the answer before being provided with the correct answer after a predetermined number of guesses. Typically, this method uses non-personally relevant stimuli so that the 'correct' answer can be manipulated to ensure errors are made during learning trials.

A summary of the studies reviewed is given in Table 2. It was decided to split the studies between the two main types of research questions identified, i.e. studies which considered the efficacy of EL when combined with other cognitive rehabilitation methods and studies which compared EL (with or without augmentation) with EF techniques. The results of the quality assessment are shown in Tables 3 and 4 for group and small n studies respectively. Each table is followed by a narrative synthesis of the results.

Table 2: Summary table

Study details	Aims [Design]	Participant details	Task/ Learning sessions [Errors during learning]	Statistics used	Main outcomes
Errorless learning augmented with other cognitive rehabilitation techniques					
Clare et al. (2002)	1. Investigate efficacy of EL using EL+MN+VC+SR v untrained (control) items. 2. Investigate long-term maintenance of treatment gains. [Within]	N=12; mean age 71(SD not given); 75% male; minimal/mild severity.	Re-learning; 6 face-name associations (FNA) of famous faces and friends and family. 6 sessions, 1 FNA per session; asked to practice between sessions. Learning criterion: Correct recall after either 10 minutes or 8 learning trials. [Errors not reported]	Group comparison: Paired t-tests Individual cases: Visual inspection of free recall scores.	Group results: Free and cued recall 1. Sig. increase in correct responses for trained items from baseline to post intervention and at 1, 3, 6 and 12 month follow-up. 2. No sig. increase from baseline for control items at any time point. Individual results: Free recall. Six participants showed clear improvement, 4 some improvement and 2 no improvement from baseline.

Study details	Aims [Design]	Participant details	Task/ Learning sessions [Errors during learning]	Statistics used	Main outcomes
Clare et al. (2003)	1. Investigate efficacy of EL using EL+MN+SR. 2. Investigate long-term maintenance of treatment gains. [SCED]	N=1; age=66; mild severity.	Re-learning; 13 FNA of friends and family. Each FNA: Variable number of learning trials (1-16) to criterion of 6 correct trials. Daily practice between sessions supported by family member. [Errors not reported]	Page test. Comparison: Baseline, intervention, post intervention and follow-up in months 1, 3 and 6.	Free recall: Sig. improvement from baseline at post intervention and all follow-up periods.
Clare et al. (2004)	Compare EL+SR v EL+ VCfc v EL+ VC v EL + MN. [SCED]	N=1; age 73; mild severity.	Learning; FNA for famous People, 4/condition. 16 twice-weekly sessions, 5 learning trials per FNA. [No errors made in any condition]	Page test Comparison: Baseline, Intervention, Post intervention and follow-up months 1, 3 and 6.	All conditions showed sig. increase in learning except EL+VC. Gains maintained at 6 month follow-up.
Thivierge et al. (2008)	1. Investigate efficacy of EL + SR in relearning IADL in mild AD. 2. Tolerability of intervention for patient and carer. [SCED]	N=2; case A: Age 66; mild/moderate severity (MMSE), severe (DRS 2); case B: Age 68; mild severity (MMSE), severe (DRS 2).	Re-learn; 1 IADL. 2 sessions per week; 45-60 minutes over 8 or 9 weeks; practice supported by carer 3 times per week. [Error rates unreported]	Visual Inspection augmented by common language effect size statistic. Comparison: Baseline to intervention, and follow-up after 1 and 5 weeks.	Evidence of significant increase in learning from baseline to intervention. Increase maintained at 5 week follow-up.

Study details	Aims [Design]	Participant details	Task/ Learning sessions [Errors during learning]	Statistics used	Main outcomes
Laffan et al. (2010)	Compare EL v EL+SG v NL (Control). [Within]	N=29; mean age 80.5 (SD 6.3); 40% male; mild/moderate severity.	Re-learn; FNA for famous people, 15 per condition. Ten twice-weekly sessions. Learning criterion not reported. [Error rates EL (mean.01) < EL+SG (mean.81). NL condition: Errors not reported]	ANOVA (ranked data).	Free recall: All training procedures produced better outcomes than NL. EL+SG sig. better than EL alone.
Comparison of errorless learning and effortful learning					
Metzler-Baddeley et al. (2005)	Compare EL v EF for: 1. Re-learning information, and 2. learning novel information. [Case study]	N=4; range 65-72; 75% male; 3 mild, 1 moderate to severe severity.	Re-learn; object names (case A and D), 17 / condition; Re-learn; FNA of famous people, 8/ condition (case B and C). Learn; 6 FNA, all cases. Cases B-D: Eight days consecutive training per condition. Case A: Learning over 4 weeks. [Error rates: Re-learning only case A EF>EL. Case B-D no sig. difference. Novel learning cases C and D EF>EL, other two cases no sig. differences]	McNemar test to compare learning in each condition to baseline. Chi-squared to compare EL v EF.	Combined free and cued recall score. Graded scoring system based on the number of cues required. Then scores converted to proportions. Main effect of learning with sig. learning from baseline in EL and EF conditions for both familiar and novel learning materials. No interaction, i.e. no advantage of EL over EF regardless of whether the learning task involved re-learning or new learning.

Study details	Aims [Design]	Participant details	Task/ Learning sessions [Errors during learning]	Statistics used	Main outcomes
Ruis and Kessels (2005)	Compare EL v EF. [Within]	N=10; range 73-89; 50% male; moderate to severe severity.	Learning, 10 FNA. One session per condition, 2 learning trials per association. [Error rates not reported]	ANOVA.	Free recall: Main effect of learning method with EL>EF after second learning trial. No difference between EL after EF after 10 minute delay.
Haslam et al. (2006): Study 1	Compare EL v EF for learning high-level (general knowledge) v low-level (specific details). [Within]	AD N=3; mean age 83; all female; severity not stated. Controls (HOA) N=8; mean age= 77.5 (SD 8.3); 7 females.	Learning; 10 face-name-occupation associations. Two sessions, 2 weeks apart, 3 learning trials per association. [Error rates not reported]	AD: Chi squared. Controls: ANOVA.	AD: Forced choice recall. Two out of 3 participants performed below chance in all conditions therefore not possible to evaluate outcomes at a group level.
Haslam et al. (2006): Study 2	Compare EL v EF for learning high-level (general knowledge) v low-level (specific details). [Case study]	N=2; mean age 81; 1 male, 1 female; severity not stated.	Learning, 10 face-name-occupation associations. Two sessions, 2 weeks apart, 3 learning trials per association. [Error rates not reported]	Chi-squared: EL v EF. Visual analysis: Levels of knowledge	Forced-choice task: Immediate and delayed recall. No sig. differences in EL v EF. Levels of knowledge difficult to interpret from visual analysis (< chance performance in some conditions).

Study details	Aims [Design]	Participant details	Task/ Learning sessions [Errors during learning]	Statistics used	Main outcomes
Bier et al. (2008)	<p>1. Investigate the efficacy of EL, SR and VC.</p> <p>2. Compare with 2 EF methods: i) Explicit mention of previous learning, and ii) no reference to previous learning.</p> <p>3. Explore the impact of error production on learning</p> <p>4. Explore the contribution of implicit memory to the methods' efficacy.</p> <p>[Within]</p>	<p>AD N=15; mean age 73.3(SD 7.3); 40% male; mild/moderate severity. HOA controls N=15; mean age 72.3(SD 7.9); 40% male.</p>	<p>Learning; FNA, 5/ condition. Ten 45 minutes sessions over 5 weeks. EL and EF: 9 presentations per association; SR 30 minutes or correct recall after 5 minutes; VC 30 minutes or 3 correct trials without cues. [Error rates for EL &lt; than all other conditions. Both SR and VC &lt; than EF conditions]</p>	Wilcoxon test.	<p>Free recall: Immediate AD: Sig. learning from baseline in all conditions but no learning method sig. better than others and low mean learning rates. HOA: Ceiling effects across all learning conditions.</p> <p>Delayed recall (2 weeks): Results from both AD and HOA groups showed floor effects. No condition showing sig. learning from baseline.</p>

Study details	Aims [Design]	Participant details	Task/ Learning sessions [Errors during learning]	Statistics used	Main outcomes
Kessels and Hensken (2009)	Compare EL v EF in people with different severity of AD. [3x2x2 quasi-experimental, mixed methods design]	All N allocated 50% between EL and EF conditions. AD mild/moderate severity N=20; EL mean age 76.5(SD 7.9); 30% male. EF mean age 77.1(9.4); 40% male. AD severe severity N=20; EL Mean age 83.6(SD 8.1); 40% male. EF mean age 83.2 (SD 7.1); 10% male. HOA controls N=20; EL mean age 72.7(SD 11); 70% male. EF=71.9(SD 8.9); 50% male.	Learn; procedural problem-solving task. One session, 1 trial per condition. [Errors not reported]	Mixed factorial ANOVA. Cohen's d (effect sizes).	Free recall: Immediate and delayed (1-3 days). Main effect of learning method with EL > EF. No interaction with severity of dementia. Effect sizes: medium to large. AD mild/moderate severity: d=.52 immediate, d=.61 delayed. AD severe severity: d= .31 immediate, .60 delayed.

Study details	Aims [Design]	Participant details	Task/ Learning sessions [Errors during learning]	Statistics used	Main outcomes
Haslam et al. (2010) Study 2	Compare EL v VC v EL+VC v EF. [Within]	N=22; mean age 75.3(SD7.3); 45% male; mild severity.	Learning; FNA, 10/condition. Two sessions, 90 minutes each, 4 trials per association. [Fewer errors in EL than all other conditions; fewer errors in EL+VC than EF]	Friedman's ANOVA.	Immediate free recall: Main effect of condition, with EF learning < than other conditions. EL+VC better than VC alone.
Mimura et al. (2010)	Compare EL v EL+ VCfc v EF. [Within]	N=18; mean age 77 (4.9); 50% male; mild/moderate severity.	Learning; word pairs. Four sessions one per condition. No learning criterion. [Errors not reported]	ANOVA.	Free and cued recall: Fewer correct responses in EF condition. Response rates in EL and EL+VC did not differ.
Dechamps et al. (2011)	Compare EL v modelling v EF. [Within]	N=14; mean age 86 (SD 5.7); 14% male; 1 mild, 5 moderate, 8 moderate/severe severity.	Learning/Re-learning, 1 IADL per condition. [Noted that all participants made errors during training. Error rates per condition not reported]	ANOVA.	Free recall: Both EL and modelling higher than EF at 1 and 3 week follow up.



Study details	Aims [Design]	Participant details	Task/ Learning session [Errors during learning]	Statistics used	Main outcomes
Noonan et al. (2012)	1. Compare EL v EF. 2. Explore cognitive factors associated with object name learning. [Within]	N=8; age or gender not reported; MMSE Range 9-26 (severe to mild severity).	Relearn; object names, 20 per condition. Ten twice-weekly sessions, each object presented 3 times per session. [Error rates EL (1%) < EF (65%)]	ANOVA; post-hoc t-tests.	Free recall: Sig. learning in both conditions at immediate recall and 1 and 5 week follow-up. No sig. differences between EL and EF at any time point.  Higher semantic memory scores, naming ability and recognition memory scores pre-intervention predicted greater rate of learning.
<p>Key</p> <p>EL=Errorless learning VC=Vanishing cues VCfc=Vanish cues with forward chaining SR=Spaced retrieval MN=Mnemonic EF= Effortful learning SG=Self-generated cues NL=Non-learning Within= Within participants</p> <p>FNA=Face Name Association IADL=Instrumental activities of daily living HOA= Healthy older adults</p>					

## *Studies of EL augmented with other cognitive rehabilitation techniques*

### *Cognitive rehabilitation techniques utilised*

Three studies used the standard techniques described in Table 1. Whilst Thivierge et al. (2008) used the standard SR paradigm to teach an IADL, the EL procedure was based on reduced levels of modelling. Here, four levels of assistance were provided with the assistance levels being decreased across trials. The four levels were: i) the researcher completes the task whilst the participant observes; ii) the researcher named each task step and the participant completed the steps; iii) the participant named the task steps and completed them with help from the researcher, if needed; and iv) the participant completed the task independently. Laffan, Metzler-Baddley, Walker and Jones (2010) compared face-name association learning using the standard EL paradigm with EL with self-generated cues and a non-learning (control) condition. In the EL with self-generated cues condition, rather than being presented with the full name during learning trials, the name always had a fixed number of letters (between 2 and 4 depending on the name length) missing from the end of the name and replaced with dashes. Although the authors referred to this as an EL procedure, it is noteworthy that the participants were asked to guess from the letters presented which would not normally be expected within an EL paradigm. In the control condition, participants were asked to name faces without cues and no feedback was provided.

### *Severity of AD*

All the studies rated the severity of cognitive impairment based on Mini-Mental State Examination (MMSE) scores, with the level of impairment varying across studies from minimal to moderate (Folstein, Folstein, & Hugh, 1975).

Thivierge et al. (2008) in their two participant SCED also reported severity based on the Dementia Rating Scale 2 (DRS) (Jurica, Leitten, & Mattis, 2001). The DRS classified both participants' level of impairment as severe rather than as mild or mild/moderate which was the classification based on their MMSE scores.

### *Learning task, learning sessions and error rates*

Four of the five studies sought to teach participants face-name associations (Clare & Wilson, 2004; Clare, Wilson, Carter & Hodges, 2003; Clare, Wilson, Carter, Roth & Hodges, 2002; Laffan et al., 2010). Of these studies, two used at least some faces which were personally relevant to the participants. Clare et al. (2002) used a mixture of personally relevant faces (friends and family) and famous faces. However, the ratio of personally relevant faces to famous faces was not reported at either a group or individual participant level. Neither was it reported whether the ratio of personally relevant faces was allocated equally to taught or control items. Clare et al. (2003) sought to teach their participant 13 face-name associations of people with whom he attended a social group. Whilst training took place at home, practice was encouraged at the participant's social group meeting thereby increasing the chance of any learning being implemented. The fifth study sought to teach one participant to use voice mail and another to delete messages from his answer machine. Teaching was carried out at home with between session practice facilitated by the person's supporter who had been trained in the intervention techniques (Thivierge et al., 2008).

Whilst all studies reported the number of training sessions, only Clare et al. (2002) reported a specific per-item learning criterion for correct responses during training (6 consecutive trials). Furthermore, three of the studies asked participants to practise between sessions (Clare et al., 2003; Clare et al., 2002; Thivierge et al.,

2008). Finally, only three studies reported the number of errors made during learning trials (Clare & Wilson, 2004; Laffan et al., 2010; Thivierge et al., 2008).

### *Outcomes*

Five of the studies used free recall as the learning outcome measure with only Thivierge et al. (2008) using an alternative outcome measure. They recorded the steps completed on a scale of the degree of assistance required which were then converted to percentages. At the end of the final training session the two participants completed 93.7% and 91.7% of the task respectively. In terms of learning face-name associations, all studies showed some evidence of learning when EL was augmented with other cognitive rehabilitation techniques. Two studies combined EL principles with a mnemonic, vanishing cues and spaced rehearsal (Clare et al., 2003; Clare et al., 2002), one added EL principles to each of the previously mentioned methods separately (Clare & Wilson, 2004) and one compared EL with EL plus self-generated cues (Laffan et al., 2010). Whilst Clare et al. (2002) and Clare et al. (2003) found evidence to support using vanishing cues; these results were not replicated when Clare et al. (2004) separately compared EL-with-vanishing-cues to other combinations of techniques. Follow-up periods were variable with periods of up to 12 months where learning gains were still significantly above baseline (Clare et al. 2002) to no follow-up (Laffan et al., 2010).

### *Errorless learning compared with effortful learning*

#### *Cognitive rehabilitation techniques utilised*

Most studies used the standard techniques described in Table 1. Two studies compared EL+VC with EF learning (Haslam et al., 2010; Mimura & Komatsu, 2010) whilst a further study compared EL using verbal instructions to modelling of steps to

be learnt and EF learning in the context of an IADL (Dechamps et al., 2011). Noonan, Pryer, Jones, Burns and Ralph (2012) in their EF condition showed pictures of objects and after one error gave an increasing number of phonemes and letters as cues until the participant was able to name the object or until the whole word was given.

### *Severity of AD*

The MMSE (Folstein, Folstein, & Hugh, 1975) was used to rate the severity of cognitive impairment in all but the two studies conducted by Haslam, Gilroy, Black and Beesley (2006). Here no global rating of impairment was given, although both studies provided adequate neurological profiles. Three studies used participants in the mild or mild/moderate impairment range (Bier et al., 2008; Haslam et al., 2010; Mimura & Komatsu, 2010) and one used participants in the moderate to severe range (Ruis & Kessels, 2005). A further two studies recruited participants with a range of impairments from mild to severe (Metzler-Baddeley & Snowden, 2005; Noonan et al., 2012). Only Kessels and Hensken (2009) directly compared the performance of participants with mild/moderate impairment to participants with a severe global impairment rating.

### *Learning task, learning sessions and error rates*

All but two studies sought to teach participants non-personally relevant information. Five taught face-name associations, one a combination of face-name and object-name associations (Metzler-Baddeley & Snowden, 2005), one taught word-pairs (Mimura & Komatsu, 2010) and a further study used the Action Programme sub-test from the Behavioural Assessment of the Dysexecutive Syndrome (BADS) (Alderman, Burgess, Emslie, Evans, & Wilson, 1996). In comparison, whilst two studies chose tasks which might be considered more personally relevant to the

participants, in neither study were participants involved in decisions concerning the information to be learnt. One study sought to teach object-name associations to a group of people with AD who showed evidence of anomia (Noonan et al., 2012) whilst the other taught IADLs, in the only study where the teaching took place in the setting, i.e. the nursing home, where the skills taught would be utilized. (Dechamps et al., 2011).

Whilst all studies reported the number of training sessions, none reported a specific per item learning criterion for correct responses during training. Furthermore, only three studies reported data to demonstrate that the number of errors made in the EL condition was significantly less than in the EF condition during learning trials (Bier et al., 2008; Haslam et al., 2010; Metzler-Baddeley & Snowden, 2005). Dechamps et al. (2011) reported that errors were made in all three learning conditions but no further details were given.

### *Outcomes*

Five of the studies found an advantage of EL over EF immediately after training (Bier et al., 2008; Dechamps et al., 2011; Haslam et al., 2010; Kessels & Hensken, 2009; Noonan et al., 2012). Whilst Dechamps et al. (2011) found the EL advantage both one and three weeks post training, Bier et al. (2008) found that, two weeks post-training, the EL advantage had dissipated. The majority of studies reviewed used free recall as the learning outcome measure whilst Metzler-Baddeley and Snowden (2005) used a combined free and cued recall score. In contrast, Dechamps et al. (2011) recorded the steps completed on an IADL on a scale, with

total scores converted to percentages to allow across task comparisons. Haslem et al. (2006) reported two studies which both used forced-choice outcome measures. In study 1 the results were lower than would be expected on the basis of chance responding, making them difficult to interpret. Similarly, the results of Ruis et al. (2005) were subject to floor effects suggesting minimal learning in both conditions.

Table 3: Quality grid for group studies

Quality criteria (Adapted from Downs and Black, 1998)	Clare et al (2002)	Laffan et al (2010)	Ruis et al (2005)	Haslam et al (2006) Study 1	Bier et al (2008)	Kessels et al (2009)	Haslam et al (2010) Study 2	Mimura et al (2010)	Dechamps et al (2011)	Noonan et al (2012)
Reporting										
Is the hypothesis/aim/objective of the study clearly described?	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes
Are the main outcomes to be measured clearly described in the Introduction or Methods section?	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes
Are the characteristics of the patients included in the study clearly described?	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes
Are the interventions of interest clearly described?	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes
Are the distributions of principal confounders in each group of subjects clearly described?	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	No
Are the main findings of the study clearly described?	Yes	Yes	Yes	No	Yes	Yes	Yes	Yes	Yes	Yes



Quality criteria (Adapted from Downs and Black, 1998)	Clare et al (2002)	Laffan et al (2010)	Ruis et al (2005)	Haslam et al (2006) Study 1	Bier et al (2008)	Kessels et al (2009)	Haslam et al (2010) Study 2	Mimura et al (2010)	Dechamps et al (2011)	Noonan et al (2012)
Does the study provide estimates of the random variability in the data for the main outcomes?	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes
External validity										
Were the subjects asked to participate in the study representative of the entire population from which they were recruited?	Unclear	Unclear	Unclear	Unclear	Unclear	Yes	Unclear	Unclear	Unclear	Unclear
Were those subjects who were prepared to participate representative of the entire population from which they were recruited?	Unclear	Unclear	Unclear	Unclear	Unclear	Yes	Unclear	Unclear	Unclear	Unclear
Were the staff, places, and facilities where the patients were treated representative of the treatment the majority	Unclear	Yes	Yes	No	Yes	Yes	No	No	Yes	Unclear

of patients receive?										
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Quality criteria (Adapted from Downs and Black, 1998)	Clare et al. (2002)	Laffan et al. (2010)	Ruis and Kessels (2005)	Haslam et al. (2006) Study 1	Bier et al. (2008)	Kessels and Hensken (2009)	Haslam et al. (2010) Study 2	Mimura et al. (2010)	Dechamps et al. (2011)	Noonan et al. (2012)
Internal validity – bias										
Was the methodology suitable to investigate the aims of the study?	Yes	Yes	No	Yes	Yes	Yes	Yes	Yes	No	Yes
Was an attempt made to blind those measuring the main outcomes of the intervention?	No	No	No	No	No	No	No	No	No	No
If any of the results of the study were based on “data dredging”, was this made clear?	Yes	Yes	N/A	Yes	N/A	N/A	Yes	N/A	N/A	Yes
Were the statistical tests used to assess the main outcomes appropriate?	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes
Was compliance with the intervention/s reliable?	Unclear	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes
Were the main outcome measures used accurate (valid and reliable)?	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes



Quality criteria (Adapted from Downs and Black, 1998)	Clare et al. (2002)	Laffan et al. (2010)	Ruis and Kessels (2005)	Haslam et al. (2006) Study 1	Bier et al. (2008)	Kessels and Hensken (2009)	Haslam et al. (2010) Study 2	Mimura et al. (2010)	Dechamps et al. (2011)	Noonan et al. (2012)
Power										
Did the study have sufficient power to detect a clinically important effect where the probability value for a difference being due to chance is less than 5%?	Unclear	Unclear	Unclear	Unclear	Unclear	Unclear	Unclear	Unclear	Unclear	Unclear
Key N/A=Non applicable										

In assessing the papers against the criteria on quality of reporting, most studies were well reported although Noonan et al. (2012) did not report the age range or gender of participants. In terms of external validity only one study, which utilised a convenience sample (Kessels & Hensken, 2009), reported information as to whether the participants recruited were representative of the entire source population. Three studies reported the proportion of participants approached who were prepared to participate (Clare et al., 2002; Kessels & Hensken, 2009; Laffan et al., 2010). Only five studies provided clear evidence that the intervention was completed in places similar to where patients are normally treated.

Two studies used recruitment methods which did not seem appropriate to meet the stated aims of their studies; Ruis and Kessels (2005) because they aimed to compare EL v EF in people with moderate to severe dementia yet the range of MMSE scores (9-22) suggested a wider range of cognitive impairment, and Dechamps et al. (2011) who aimed to compare EL v EF in different stages of dementia yet did not address the question of severity in their reported results or outcomes. No studies attempted to blind assessors to the intervention conditions. The outcome measures and choice of statistics were appropriate in all studies. Furthermore, where additional unplanned analyses were undertaken this was made clear to the reader. With regard to adherence to the intervention, Clare et al. (2002) asked participants to practice the face-name associations between training sessions but no evidence was presented that this practice had been undertaken. Finally, no a priori power calculations were reported for any studies.

Table 4: Quality grid for SCED and case studies

Quality criteria (Adapted from Tate et al., 2008)	Study				
	Clare et al. (2003)	Clare et al. (2004)	Thivierge et al. (2008)	Metzler-Baddeley et al. (2005)	Haslam et al. (2006): Study 2
1. Was the clinical history described adequately?	Yes	Yes	Yes	Yes	Yes
2. Target behaviours The paper identifies a precise, repeatable and operationally defined target for intervention that can be used to measure treatment success.	Yes	Yes	Yes	Yes	Yes
3. Design The study design allows for the examination of cause and effect relationships to demonstrate treatment efficacy.	Yes	Yes	Yes	Yes	Yes
4. Baseline To establish that sufficient sampling of behaviour had occurred during the pre-treatment period to provide an adequate baseline measure.	Yes	Yes	Yes	No	No
5. Sampling behaviour during treatment To establish that sufficient sampling of behaviour during the treatment phase has occurred to differentiate a treatment response from fluctuations in behaviour that may have occurred at baseline.	Yes	Yes	Yes	No	No
6. Raw data record To provide an accurate representation of the variability of the target behaviour.	Yes	Yes	Yes	No	Yes
7. Inter-rater reliability To determine if the target behaviour measure is reliable and collected in a consistent manner	N/A	N/A	No	N/A	N/A
8. Independence of assessors To reduce assessment bias by employing a person who is otherwise uninvolved in the study, to provide an evaluation of the patients.	N/A	N/A	No	N/A	N/A

Quality criteria (Adapted from Tate et al., 2008)	Study				
	Clare et al. (2003)	Clare et al. (2004)	Thivierge et al. (2008)	Metzler-Baddeley et al. (2005)	Haslam et al. (2006): Study 2
<p>9. Statistical analysis To demonstrate the effectiveness of the treatment of interest by statistically comparing the results over the study phases.</p>	Yes	Yes	Yes	Yes	No
<p>10. Replication To demonstrate that the application and results of the therapy are not limited to a specific individual or situation (i.e., that the results are reproduced in other circumstances –replicated across subjects, therapists or settings).</p>	No	No	No	No	No
<p>11. Generalisation To demonstrate the functional utility of the treatment in extending beyond the target behaviours or therapy environment into other areas of the individual's life.</p>	Unclear	No	Unclear	No	No

All the SCED and case studies provided adequate clinical histories, definitions of the learning tasks and used appropriate designs. In terms of baseline testing and sampling of learning during the intervention, Haslam et al. (2006) only used a single learning session. Furthermore, in this study variability was not reported and forced-choice outcomes were used. The performance of two of the three patients was below chance in at least some of the conditions meaning that there were a lot of flaws in this study. Whilst Metzler-Baddeley and Snowden (2005) took a pre-intervention baseline measure in their study, learning was only measured after the six training sessions were completed. Here proportional mean recall scales which took account of baseline responding were reported. None of the studies reported inter-rater reliability or had independent outcome assessors. With the exception of Thiverge et al. (2008) where the outcome measure assessed the ability of participants to complete an IADL which might be open to subjective interpretation, the other studies used concrete measures such as free recall which are less open to interpretation bias.

None of the studies provided good evidence that the results could be replicated across participants, therapists or settings. In terms of generalising the results, only two of the studies used learning tasks of personal relevance to the participants. One, included a self-report by both the participant and his spouse that he was using the learnt face-name associations at his social group meetings (Clare et al., 2003). The second, presented no evidence that this learning was being used routinely by the two participants (Thivierge et al., 2008).



## **Discussion**

The discussion begins by considering the quality of the studies reviewed, followed by a discussion of methodological issues that impact on the conclusions that can be drawn. This is followed by a conclusion regarding the efficacy of EL based on the evidence reviewed and a discussion of implications for clinical practice. Then, suggestions for further research are summarised and limitations of this review discussed.

Starting with group studies, no RCTs were identified during this review. All the studies used a within-participants design with the exception of Kessels et al. (2009) who used a mixed design with the between-participants factor being the allocation of participants to groups on the basis of the severity of their cognitive impairment. Whilst RCTs, which include random allocation of participants to intervention or non-intervention groups, are considered to provide the strongest type of research evidence (American Academy of Neurology, 2004), a within-participants methodology can be useful particularly in disorders such as AD where the manifestation of the difficulties across participants is heterogeneous. Here, within-participants designs help control for intra-participant factors which would be expected to be equivalent pre- and post-intervention. Therefore this methodology increases the internal validity of the results obtained. However, a within-participant methodology can also reduce the generalisability of the results, especially when coupled with the lack of evidence that the participants recruited were representative of the source population and small participant numbers. In group studies, the small sample sizes increase the possibility of making a type 2 error, i.e. concluding that there is no significant difference between the effectiveness of the compared interventions when this is not the case (Keppel, Saufley, & Tokunaga, 1992).

Also, the limited evidence regarding the proportion of participants approached who were prepared to participate raises the question of possible selection bias.

Furthermore, in terms of generalising the results, only five studies provided clear evidence that the intervention was completed in places similar to where patients are normally treated. This is important because if interventions were carried out in a place that was not routinely available, this may make the results difficult to replicate in clinical settings. Also, and particularly relevant to cognitive rehabilitation interventions, is the issue that the results of learning are not expected to be readily transferable across settings (Clare, 2007). This makes it important that learning takes place where the acquired knowledge is expected to be used, yet only three studies (two of them SECDs) did this (Clare et al., 2003; Dechamps et al., 2011; Thivierge et al., 2008). This might be a reflection of the fact that, apart from Clare et al. (2002) which used a mixture of personally relevant and non-personally relevant stimuli, these were the only studies which sought to make the learning relevant to the needs and goals of participants. However, with the exception of Clare et al. (2003) these studies provided no evidence that the learning had actually been used, evidence which would have strengthened the results. Since the aim of cognitive rehabilitation is to address goals that are personally relevant to the recipient of that intervention (Clare, 2007), the lack of ecological validity of the taught tasks is a major limitation of most of the studies under review for a number of reasons. First, low motivation might account for the low learning rates in some studies (e.g. Haslam et al., 2006); second learning is not reinforced between sessions which would help to consolidate the learning; and, third, as stated above it is unclear whether learning, even if effective, would transfer to settings where it would be useful.

Most studies used methodology that was suitable to meet the aims stated. Two studies can be suggested to have utilised unsuitable methodology. First, Ruis and Kessels (2005) who, despite having participants with moderate to severe dementia, allowed only two learning trials per face-name association, leading to floor effects; and second Dechamps et al. (2008). Here, their methodology was suitable to compare the chosen interventions. Yet their aim to compare relearning of IADLs in different stages of dementia was not addressed, thus restricting the conclusions that can be drawn. Furthermore, whilst no studies used blind outcome assessors, this is particularly problematic for Dechamp et al.'s study since they used a graded system for whether learning had been successful. This scale required more subjective judgments to be made than in studies that used free or cued recall of names as outcomes. Although the researchers received training in the scoring criteria, the subjectivity of the outcome measure makes the outcome measure more susceptible to bias. This makes it particularly preferable to use independent assessors in evaluating the intervention implemented.

None of the studies reported a priori power calculation which, given the small n used across studies, is problematic. Insufficient sample sizes increase the possibility of making a type 2 error i.e. concluding that there is no significant difference between the efficacy of interventions when that is not the case (Keppel, Saufley, & Tokunaga, 1992). Related to this is the question of effect size. As part of a power calculation decisions are made about effect size that would be required to illustrate the clinical utility of intervention. That is whilst a p-value can provide comfort about the reliability of an observed difference, it does not reflect the magnitude of the observed difference (Cohen, 1992), i.e. a difference between EL and EF learning might be statistically significant but the magnitude of the difference might be small and therefore not particularly useful in terms of clinical practice. Whilst all studies reported p-values, only two

reported related effect sizes making it difficult to comment on whether the effect is clinically meaningful. Also, in terms of clinical utility few studies included any long-term follow-up. One study which included follow-up at 6 and 12 months was conducted by Clare et al. (2002) who demonstrated that, whilst as would be anticipated with a degenerative disease process, not all learning had been retained, recall even at month 12 was significantly above baseline.

Turning to SCED and case studies, the most noteworthy quality issue was the lack of evidence of replication and whether the results could be generalised. This is a common difficulty with this type of design and is reflected in this type of evidence being graded the lowest in terms of overall quality (American Academy of Neurology, 2004). Despite this SCED and case studies can be useful particularly when testing novel interventions and in rare conditions, or those for which recruiting participants is particularly problematic (Matson, Turygin, Beighley, & Matson, 2012). This is pertinent to people with AD since, whilst AD is the most commonly diagnosed dementia, a diagnosis of dementia and age have been identified as barriers to participating in research (Woodall, Morgan, Sloan, & Howard, 2010).

All studies which augmented EL principles with other methodologies provided evidence of efficacy. However, it is not possible to conclude that EL was responsible for the learning, or whether it is more efficacious due to the combination of multiple learning methods. This issue is compounded by the failure of most studies (regardless of methodology) to report the number of errors made during learning (Clare & Jones, 2008) meaning there is no assurance that the methods employed were “errorless”. Indeed authors have classified some methodologies as EL techniques whilst others have classified the same type of intervention as EF. For example vanishing cues (VC) is often considered an EL technique analogous to the fading cueing

paradigm used in the original animal learning literature (Terrace, 1963). However in their review, Middleton and Schwartz (2012) reclassified studies using VC as errorful, regardless of how the original papers classified this technique, on the grounds that it could lead to errors; thus demonstrating that reporting errors is crucial. A further complication occurs in the case of SCED studies. In this type of study it is important to establish a stable baseline from which to measure subsequent learning (Tate et al., 2008). However, no studies reported whether, during the collection of these baseline measures, EL principles were adopted i.e. whether participants were encouraged not to guess. Given that the premise of errorless learning is error elimination or minimisation (in order to reduce competing memory traces), this methodology appears to present a possible confound within the data. Collection of the baseline measures, which use the same materials as used in the intervention which follows, may produce errors which could interfere with subsequent learning and recall during the intervention phases of the study. These errors could be accounted for if errors made were routinely reported.

The number of training sessions varied from one to 16 sessions and one learning trial to up to 16 learning trials. Only one study, Haslam et al. (2010), piloted the number of learning sessions required and also the number of items that could reasonably be learnt to avoid floor effects. Furthermore, few studies considered setting a learning criterion. If it could be demonstrated that participants were able to meet the set criterion during learning this would suggest that the number of training sessions was sufficient. Also, some studies taught all the learning materials in one session whereas others taught one item per session. This meant that when learning was evaluated at the end of the intervention the period between learning and retest was not consistent. Although it can be argued that this was equidistant between participants in

each study, it does not speak to the number of learning trials required, nor the period over which information can be retained.

The MMSE was used to rate the severity of dementia. However, the MMSE is prone to distortion and is insufficiently sensitive in those with either high or low levels of education. It is therefore more suited to monitoring change over time than to staging the severity of the disease (Crum, Anthony, Bassett, & Folstein, 1993). Furthermore, even in studies which were more comprehensive in terms of neuropsychological profile, severity was rarely accounted for in the results obtained. Whilst the purpose of cognitive rehabilitation is not to improve global cognitive functioning, rates of learning would still be anticipated to be related to severity of impairment.

Based on the literature reviewed, there is evidence that EL (when augmented with other strategies) can be efficacious in promoting learning in people with mild dementia. However, it is not possible from these studies to draw strong conclusions about the effectiveness of EL per se. Only one study directly compared EL with either EL plus spaced retrieval or with a control condition (where no learning strategies were used) (Laffan et al., 2010). Here, EL was found to be more effective than the control condition, but not as efficacious as when it was augmented with a spaced retrieval technique. In respect of studies which compared EL and EF learning, six studies included participants classified as having AD of either mild or mild/moderate severity. Of these six studies, none showed an advantage of trial-and-error (EF) learning over EL. Furthermore, four studies demonstrated better learning when EL was used (Bier et al., 2008; Haslam et al., 2010; Kessels and Hensken, 2009; Mimura et al., 2010). However, in the case of Bier et al., (2008) the EL advantage had dissipated two weeks later. Furthermore, three studies also compared EL with other non-trial-and-error effortful learning methods. Two reported no significant differences in the effectiveness of EL over either spaced retrieval or vanishing cues

(Bier et al., 2008; Mimura et al., 2010), whilst Haslam et al., (2010) found that combining EL with vanishing cues was better than using vanishing cues alone. In summary, there is evidence for the efficacy of EL over trial-and-error learning in people with mild/moderate dementia. However, the author concurs with the conclusions of Clare and Jones (2008) that there is no clear evidence for using EL over other more effortful learning methods (such as spaced retrieval or vanishing cues) in people with mild dementia. In terms of more moderate/severe dementia, three studies including participants with more severe levels of impairment. Here, there is limited evidence from two studies of a preference for EL over EF in respect of procedural tasks (Dechamps et al., 2011; Kessels & Hensken, 2009).

Despite the lack of clear evidence in support of EL over other more effortful learning strategies, in terms of clinical practice there are still situations where this approach may be useful. Whilst acknowledging that in 'real world' situations using the principles of EL can present practical challenges and be time-consuming, EL can be useful where the outcome is important to the person with AD, for example, where they want to relearn limited amounts of personally relevant information, and/or where forgetting this information is causing the person distress and negatively impacting on their quality of life, e.g. the names of close family members. Also, EL may be useful to augment other strategies, for example, suppose the task is to learn how to use a telephone answering machine which consists of a number of steps. Rather than using EL to teach the steps so that they can be remembered by the person with AD; it might be beneficial to use written and/or pictorial cues placed by the answering machine. These cues could be supplemented by using EL to teach the person with AD to look for the cues when using the answering machine. This has the advantage of reducing the memory load since the person with AD only has to learn one step, i.e. look for cues rather than a series of steps. To further increase

the efficacy of EL interventions, wherever possible, help from a supporter to encourage practice between intervention sessions should be used; with the supporter receiving training in the intervention techniques. This could be achieved by them joining the last part of each session and receiving a written, user-friendly guide explaining the techniques.

To improve the quality of the evidence base, future research to address a number of methodological issues would be useful. First, the lack of ecologically valid tasks related to the goals of the participant is problematic and could be addressed by collaborative goal setting with the participant and/or their supporters. Second, studies should include follow-up periods to assess the utility of EL. For learning to be clinically useful, it must have a degree of longevity, although given that AD is a degenerative disorder (McKhann et al., 2011) the period of follow-up required would be relatively short. Studies should also monitor cognitive and functional ability over the follow-up period. By doing so, a generalised decline in abilities can be accounted for when considering the benefit of the intervention over time. Third, recording of errors at baseline and during training should be undertaken and reported to clarify whether the interventions employed are indeed either errorless or produce minimal errors. Fourth, the rationale for the learning schedule, i.e. number of sessions, trials per item should be clearly explained and piloted to reduce floor effects and make replication easier. Setting a learning criterion may help with this. Fifth, the outcomes should include a measure which assesses whether the learning is actually being used in the participant's day-to-day life. This might be based on self-report by the participants, reports of their supporters or carers, or direct observation. Finally, large scale studies with larger sample sizes would provide stronger evidence, providing the methodology used was appropriately designed and implemented, with appropriate controls to reduce sources of bias.



This review has a number of limitations. First, the search strategy only captured studies which had been published in peer reviewed journals. Whilst this was planned, it does mean that the number of relevant studies identified may have been understated since studies can be reported in a number of ways, e.g. through conference abstracts or theses (Centre for Reviews and Dissemination, 2008). Excluding these studies may lead to an overestimate of the efficacy of interventions due to a publication bias, where studies are more likely to be published if they report findings that allow the null hypothesis to be rejected (Dubben & Beck-Bornholdt, 2005). A similar issue applies to the decision to include only papers published in the English language. Again, aside from reducing the quantity of evidence available, studies conducted in languages other than English are more likely to be published in English language journals if they report significant results therefore potentially overestimating efficacy (Moher, Pham, Lawson, & Klassen, 2003).

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EMPIRICAL PAPER

**LIVING WITH A DIAGNOSIS OF BEHAVIOURAL-VARIANT  
FRONTOTEMPORAL DEMENTIA: THE PERSON'S EXPERIENCE**

Word Count: 8286

## Abstract

Research investigating behavioural-variant frontotemporal dementia (bvFTD) has concentrated on identifying and quantifying people's difficulties; yet few studies have considered how people with bvFTD make sense of their difficulties. This study sought to elicit the subjective experiences of five people living with this diagnosis. Participants were interviewed and interpretive phenomenological analysis was used to analyse the data. Two super-ordinate themes emerged: firstly, '*Bewilderment*' and secondly '*Relationships with others*'. In the case of '*Bewilderment*', this reflected the feelings of the participants from the start of their dementia journey and was divided into two main themes (1) '*Awareness of change: What's the problem?:* Awareness of changes in behaviour or lifestyle, and (2) '*Threats to self: This is not me:* Changes in behaviour or lifestyle which negatively impact on their sense of self. The second super-ordinate theme, '*Relationships with others*', reflected difficulties with social relationships and comprised two main themes (1) '*Family and friends: Things haven't changed... but do I say anything wrong?:*' Paradox between feeling their relationships were unchanged but an awareness that something was not as it was previously, and (2) '*Coping with threats to self: Blame others or just avoid them:* Ways participants sought to cope. The themes were discussed in relation to literature evaluating the difficulties associated with bvFTD together with implications for clinical practice.

Key words: dementia, relationships, coping, identity, interpretative phenomenological analysis (IPA), qualitative analysis.

## Introduction

Frontal-variant frontotemporal dementia (fvFTD) is a progressive neurological disorder which is associated with insidious changes in personality and behaviour (Hodges et al., 1999). At the time this study was designed, fvFTD was the commonly used term to distinguish behavioural presentations of frontotemporal dementia from the temporal lobe variant which is associated primarily with language difficulties (Mendez, Lauterbach, & Sampson, 2008). However, more recently, revised consensus criteria have been published where the term fvFTD has been revised to behavioural-variant frontotemporal dementia (bvFTD). Therefore, this term will be used through the remainder of this paper. The revised consensus criteria require that, for a diagnosis of possible or probable bvFTD to be made, there must be evidence of changes in behaviour and/or cognition which are progressive; coupled with three difficulties from: Behavioural disinhibition (e.g. socially inappropriate behaviour), apathy, lack of empathy, stereotypical or compulsive behaviour, hyperorality, and finally a cognitive profile following neuropsychological testing of deficits in executive function combined with relatively preserved memory and visuospatial abilities (Rascovsky et al., 2011).

Research with people with bvFTD has largely concentrated on identifying and quantifying areas of difficulty, for example, evaluating performance across multiple cognitive domains (e.g. Hodges et al., 1999; Rahman, Sahakian, Hodges, Rogers, & Robbins, 1999). Here, it has been shown that bvFTD is difficult to detect on traditional neuropsychological tests due to the lack of a social component to these tests. A second strand of research has concentrated on the differential diagnosis between people with bvFTD and Alzheimer's disease (for a review see Hutchinson &

Mathias, 2007) and has highlighted the relatively preserved memory of people with bvFTD compared with those with earlier stage Alzheimer's disease. Finally, a third area of research has focussed on social cognition. Here deficits have been found in Theory of Mind (TOM) (Fernandez-Duque, Baird, & Black, 2007; Gregory et al., 2002; Snowden et al., 2003), moral reasoning (Mendez, Anderson, & Shapira, 2005; Lough et al., 2006) and the recognition of emotions (particularly negative emotions such as anger) (Fernandez-Duque, & Black, 2005; Keane et al., 1999; Lough et al., 2006). This body of research has demonstrated that people with bvFTD show impairments in moral reasoning, and difficulties in both identifying the intentions of others and understanding their view point; coupled with deficits in recognising the emotional states of others.

In contrast, there is a paucity of research investigating how people with bvFTD understand and make sense of the changes within themselves, and of the effect the diagnosis has had on their lives; although the view of family members has been explored (Oyebode, Bradley & Allen, 2012). One reason for this is the assumption that people with bvFTD lack insight into their difficulties and so would have little to offer by way of reflective accounts (e.g. Rankin, Baldwin, Pace-Savitsky, Kramer, & Miller, 2005). Historically, a similar assumption was made about people with Alzheimer's disease. However, this has been found not to be the case. In mild Alzheimer's disease semi-structured interviews have been successfully employed to gain people's views on: Adjustment and coping (Clare, 2002; Pearce, Clare, & Pistrang, 2002), awareness of their difficulties and beliefs about the cause (e.g. Clare, 2003; Clare, Goater, & Woods, 2006; Devlin, MacAskill, & Steed, 2007), and the impact of the diagnosis of Alzheimer's disease on their sense of identity (Beard, 2004; Sabat & Harre, 1992; Caddell & Clare, 2011;

Menne, Kinney, & Morhardt, 2002; Phinney & Chesla, 2003). Also, similar methodologies have been used to investigate the views of people with moderate to severe Alzheimer's disease residing in residential care (e.g., Clare, Rowlands, Bruce, Surr, & Downs, 2008).

Some studies have considered the experiences of people with dementia as a diagnostic group rather than at the level of particular sub-types of the condition. For example, Harman and Clare (2006) interviewed nine people with early-stage dementia (including two with a diagnosis of bvFTD) about their experience of living with dementia. They reported two themes of recognising that their difficulties would get worse and trying to maintain a sense of identity. However, studies which interview people with diagnoses that fall under the umbrella term of 'dementia' make it difficult to extract issues of particular relevance to people with rarer types of dementia such as bvFTD. Also, they are predicated on the assumption that all dementia-type disorders are similar, i.e., there is homogeneity in relation to the phenomenon being explored (Smith, Flowers, & Larkin 2009). However, whilst Alzheimer's disease and bvFTD fall within the cluster of dementia disorders, there are important differences in presentations between the two conditions. Alzheimer's disease is characterised by an initial amnesic syndrome with neuropathology focused in the medial temporal lobe structures that are important for encoding new memories (Braak & Braak, 1995). In contrast, bvFTD is associated with early pathology in the frontal cortex and is associated with difficulties in social relationships and disinhibited behaviour (e.g., Neary et al., 1998). Therefore, it would be anticipated that people's subjective experience of living with these two conditions would differ. Finally, whilst bvFTD is a relatively rare disorder, accounting for approximately 2% of all dementia diagnoses made, it is more commonly diagnosed in people between the ages of 45 and 65. Here the prevalence rate rises

from 2% to 12% (Alzheimer's Society, 2012). Given that the age of onset of bvFTD tends to be younger than for the overall cohort of people with dementia, this would be a further reason to hypothesise that those with bvFTD would have different subjective experiences of living with this diagnosis from people with dementia generally.

Understanding the perspective of the person with bvFTD is important both in terms of promoting engagement with services, and also to assist in designing and evaluating interventions that are sensitive to, and respectful of, the perspective of the person with bvFTD. Therefore, this study sought to elicit the views of people with bvFTD on their experience of living with this diagnosis.

## **Method**

### *Participants*

The participants comprised five patients with a diagnosis of probable bvFTD (3 males and 2 females). The diagnosis of bvFTD was made in accordance with the relevant consensus criteria (Rascovsky et al., 2011). Although two of the participants were diagnosed using the previous consensus criteria (Neary et al., 1998), it was confirmed with the medical professional who made the diagnosis that they also met the revised criteria for probable bvFTD. A diagnosis of bvFTD was the main inclusion criterion. The second criterion was that participants should be aware of their diagnosis. This criterion was in place for two reasons, first it is best practice in dementia care that people be informed of their diagnosis unless they express a wish not to be told (National Institute for Health and Clinical Excellence, 2011) and second, it would be difficult to discuss people's experience of living with bvFTD if they were unaware of their diagnosis. People were



excluded if they were unable to give valid consent or, if their verbal English language skills were insufficient to enable them to take part in an interview, e.g. where English was not their first language. This criterion was in place due to the difficulties associated with ensuring the fidelity of people’s verbal accounts obtained via interpreters.

The participant demographics are shown in Table 1. All lived with spouses or partners. Three participants also had an adult-child living at home. Four were seen in their own homes whilst one was interviewed in hospital.

The study was approved by the local NHS ethics committee (appendix 12) and the recruiting Trust’s R& D department (appendix 13). Sponsorship for the study was provided by the University of Birmingham (appendix 14). All participants gave written informed consent. No one was paid for their participation.

Table 1: Participant demographics

Pseudonym	Gender	Age	Time since diagnosis	No. of interviews
Patrick	Male	62	1-2 years	1
Tom	Male	60	< 1 year	3
David	Male	62	1-2 years	3
Christine	Female	58	<1 year	1
Jayne*	Female	46	< 1 year	1

\* Requested that her partner was present during the interview

## *Design*

This qualitative study interviewed people with bvFTD using semi-structured interviews. Whilst a schedule of topic areas and related prompts was devised to help guide the interviews (appendix 15), this was used flexibly to allow each participant to tell their own story in their own way in line with the principles of interpretive phenomenological analysis (IPA) (Smith, Flowers, & Larkin, 2009). All participants were asked to choose the topic that they wished to start with.

The interviews were transcribed verbatim and the principles of IPA were used to analyse the resulting transcripts. IPA was used since the aim of this study was to explore the participants' subjective experiences and the meanings they made of their experiences. Throughout, the focus is on 'personal meaning-making' rather than a comparison of that meaning to 'objective facts'. IPA is a double hermeneutic process with the participant making meaning of their experiences and then the researcher endeavouring to make sense of these experiences through their transcripts (Smith et al., 2009). Thus the researcher's interpretations are subjective since there are multiple possible interpretations and different researchers may place different meanings or emphases on different parts of the transcripts during the interpretative process. IPA is an idiographic approach with participants' transcripts being initially analysed as single entities with interpretations being integrated across participants at the end of this process.

To maximise the quality of the data collected and avoid overburdening participants, all were offered the opportunity to take part in either one interview or several shorter interviews (Paterson & Scott-Findlay, 2002; Snyder, 2003). Participants did not have to decide immediately whether they preferred one or several interview sessions. Rather, they were asked to express an initial preference, but the timings of the meeting(s) was flexible to allow for them to change their

mind during the interview session, e.g. if they started to feel tired or, alternatively, if they decided they were able to manage a longer session. Second, since people with bvFTD often have communication problems which manifest themselves as poverty of speech and a tendency to introversion which makes social interactions difficult (e.g. Rankin et al., 2005; Levenson & Miller 2007) to help participants to express themselves a number of methods were employed. During the interviews, participants were offered the opportunity to discuss photographs and or objects that were important to them (Lloyd, Gatherer, & Kalsy, 2006; Robinson, 2000). When phrasing questions sometimes concrete prompts were used, such as referring to specific family members by name to ensure understanding, or asking about photographs or objects participants brought to the interviews. Also, closed questions were used to clarify answers. To reduce the potential bias that might be introduced, the same issues were revisited in slightly different ways and the answers triangulated to ensure consistency.

### *Procedure*

Potential participants were identified by clinical staff (independent of the research study) from a working age dementia service in a large urban centre in the UK. Whilst the NHS ethics and Research and Development permissions also allowed recruitment from the older adults memory service, only one potential participant was identified from this service. This person chose not to participate. Given that bvFTD is more common in people of working age, it was not expected that many potential participants would be identified through the older adult service. However, permission was obtained for recruitment from this source to make the potential participant pool as broad as possible.

Staff briefly informed potential participants about the study and asked if they might be interested in participating. Those who expressed an interest were given an information sheet and consent form to take away (appendices 16 and 17). Permission was gained to give the potential participant's contact details to the researcher who contacted them a minimum of 48 hours after the initial approach and arranged an appointment to meet with the participant. At this meeting the research was explained further and any questions answered. All were informed that choosing to either participate, or not, would have no effect on their ongoing treatment and that they could withdraw from the study at any time until the final write-up without affecting their current or any future treatment.

If participants chose to take part, consent was then taken. Under the Mental Capacity Act (2005), capacity to consent is assumed unless there is evidence that the individual is unable to give valid consent. The researcher sensitively assessed the participants' understanding of the research, and its potential benefits and risks; the participants' understanding that they were free to decline to take part and that they were free to withdraw at any time until the final write-up. Since, for some people, their level of understanding might vary throughout the day all meetings were arranged for a time of day when the participant felt 'brightest'. All gave valid written consent.

## **Analytic Process**

IPA involves a series of steps, the first four of which are completed on a case-by-case basis whilst the final stage involves bringing the analysis together across cases (Smith et al., 2009). First, each individual transcript was read multiple times (in conjunction with notes regarding the photographs and personal objects one participant brought to the interview) and the researcher recorded initial thoughts on the descriptive comments, linguistic content (use of pronouns, repeated phrases etc), and conceptual comments. Second, the researcher identified emergent themes within each text which sought to encapsulate the initial notes made, which were grounded in the text. Third, the researcher created a structure out of these by looking for connections, whether commonalities or polarities. This process was completed for all transcripts. Lastly, the themes were compared across participants and a final theme structure was produced.

Since, as discussed earlier, the interpretations of the researcher are subjective, two methods were employed to increase the credibility of the analysis. Firstly, two supervisors, who have extensive experience of working with people with dementia, oversaw the analysis from initial coding through to the development of final themes. Secondly, regular meetings were attended with a peer group undertaking qualitative research in different fields from those explored in this study. Here, portions of transcripts were shared and initial coding and emergent themes discussed. The balance between supervisors with extensive experience in the field of dementia and a peer group whose knowledge of bvFTD was more limited helped to increase the range of ideas and interpretations considered and reduce researcher bias.

## Results

In this section the themes will be described together with relevant quotes and commentary on how the quotes illustrate the themes. The theme structure is displayed in Table 2. Four main themes emerged from the analytic process. These were ‘Awareness of change: What’s the problem?’, ‘Threats to self: This is not me’, ‘Family and friends: Things haven’t changed... but do I say anything wrong?’ and ‘Coping with threats to self: Blame others or just avoid them’. These were further consolidated into the two overarching super-ordinate themes of ‘Bewilderment’ and ‘Relationships with others’.

Table 2: Theme Structure

<b>Super-ordinate themes</b>	<b>Main Themes</b>
Bewilderment	Awareness of change: What’s the problem?  Threats to self: This is not me
Relationships with others	Family and friends: Things haven’t changed... but do I say anything wrong?  Coping with threats to self: Blame others or just avoid them

### **Super-ordinate theme: Bewilderment**

This super-ordinate theme reflects the feelings of the participants from the start of their dementia journey through their first contact with health professionals to diagnosis and beyond and how they have tried to make sense of changes in themselves and/or their lives.

*Awareness of change: What's the problem?*

All the participants, in varying degrees, were able to report some changes that either they had noticed, or which had been reported to them by members of their families. However, two participants thought that these changes did not warrant medical intervention; whilst a third participant thought any changes in behaviour noticed by his wife were the result of a fall. Tom, for example, specifically questioned the need for medical help. He talked about difficulties he experienced with expressive language and at the start of the first interview he kept fidgeting with papers on the coffee table and made statements such as:

I was trying to get it out without making ... getting too many words in (laughs). Em. When I, sorry, when I try to get some words individually, I'm not as good at (laughs) what I do, what I do, when I do it. If you understand that? (laughs)

Tom

Yet, despite this marked expressive language difficulty, Tom seemed bemused as to why he had needed to see his GP or be referred to specialist services. He talked about how he “wasn't sure of the need” and how he “just sort of got into it” and “literally, I don't know how they knew”. He used the word “strange” and “weird” on multiple occasions when describing how he felt about his involvement with mental health services, for example, in reference to “going to see people two or three times and don't know why”. He described health professionals visiting him at home as “weird people just turning up” and “like it is some sort of mistake that's how I sort of make sense of it”.

Tom also identified dramatic reductions in previously enjoyed activities, for example, leisure pursuits that previously had occupied large amounts of his free time. He chose to show the interviewer pictures of himself taking part in a number of outdoor activities such as gliding,

white-water rafting and skiing which he described as “absolutely brilliant”, “just the sort of good day of fun” and “thrilling”. He also showed pictures of holidays aboard where he enthusiastically described activities such as going for walks and horse-riding saying: “You know what it’s like. Living in a town for long periods of time” and “it can be like becoming a robot”. He also showed a medal he had won for running and talked about the importance of obtaining good performance times and going running with a local club several times a week. Yet, whilst describing these changes he did not seem overtly bothered by the enormity of changes in his behaviour. He acknowledged that he “don’t get to do that stuff now” explaining:

I’m, I mean I’d like to do loads of stuff but sometimes you can’t be, be, cannot get up off the seat like, get, get the motivation to get out there and do it, like.

It’s just one of those things. It’s changed hasn’t it. Things are not really the same. You just don’t get to do those things these days. You think to yourself I want to.

Tom

Similarly, David talked at length about his passion for sports cars saying “I love high-speed cars”. He talked about the number of sports cars he had owned (e.g. make, type, colour) and the numerous pleasure trips he had taken in them. For example:

I used to drive it all over the place, because I used to go down to Ramsgate, Margate, all along the coast and I used to just drive round and then ... I used to go down on a Friday, Friday night, and then come back on Sunday. Yeah, it was great.

David

Yet, these detailed descriptions of his love of cars and driving were incongruent with the brevity of his explanation for not driving anymore: “I haven’t got a car now, because once I left work I got rid of my car.” In a similar vein, David talked about his love of golf and went through a list of golf courses which he had played at. Again, his explanation of why he no longer played “because I don’t have anyone to play with” was brief compared with his narrative about playing



golf. He also talked about his former busy job and gave lots of concrete details about the nature of his work, the responsibility it entailed and the amount of travel involved. However, when asked what he now does during the day he said “I don’t do hardly anything in the daytime because I don’t go to work”. He explained that he often takes the bus to a local shopping centre and wanders around because “when I’m at home I’m on my own all the time, because my wife, she’s at work, and my daughter’s at work”. Despite David being able to give detailed descriptions of changes in his behaviour and of giving up activities which sounded as if they had been very central to his life and a major source of excitement; when asked “Do you think you’ve changed at all?” and “Do you think some of your hobbies have changed?” David responded “No”. His simple monosyllabic response to being asked about whether he had changed, seemed totally at odds with the factual description he had given.

Despite showing no concern over the behavioural changes he had talked about, David described how his wife took him to the doctor, who “Just wanted to check my brain” thus illustrating the paradox between David’s personal awareness of changes, and his awareness, at some level, of his wife’s concerns and yet the lack of personal concern or emotional response in his narrative. In a similar vein David, who was in hospital when interviewed, gave the reason for being there as “I have frontal lobe dementia but it has not affected me.” A statement he repeated several times during the interviews. Also, David explained that the people in hospital with him “had problems with their brains” but then continued to say the doctor had said he “must have that [frontal-temporal dementia] but I haven’t because they do things wrong, but I don’t do things wrong”. This did not seem to reflect a lack of understanding of his diagnosis at a cognitive level since David showed a rudimentary understanding of this. (Interviewer: What do you think

frontal-temporal dementia is? David: Well, that affects like how you think.) Rather, it suggests a difficulty ascribing the diagnosis to himself.

Similarly, Patrick described his first contact with the Health Service following a fall and demonstrated awareness that his wife was concerned about him. However, whilst Patrick complied with this request he showed no personal concern:

I had a fall. We were living in [name house] at that stage, our house before this house, and we had a leak in the conservatory, and so I went up on to the conservatory roof to see if I could cure the problem. And it was damp so I thought, right, I shall dry that off, I'll then put white spirit on it, and I'll then seal it. That's all I remember doing. The next thing I can recall was I was on the patio below; I had fallen off, hit my head on the wall and knocked myself out. I fractured my left arm. And [my wife] was on the telephone, when I came to, and she's speaking to paramedics, and they came to collect me in an ambulance. I was taken down on a board, down the steps, and taken to hospital. I had my arm dressed, they put a cast on it, and a fortnight later [my wife] said, 'You've been affected. You need to go and see a doctor.' Okay. So I went to see my GP and she referred me to a psychiatrist. And the psychiatrist said, 'Right, I'm just going to do a scan,' which she did, and this was in the millimetres, or centimetres rather, and she said, she believed I had frontal lobe dementia.

Like David, Patrick did not feel that he had changed, other than a reduction in his short-term memory. He also showed an understanding of his diagnosis saying: "I've got a medical diagnosis now which is I've got frontal lobe dementia, and I recognise that people who have frontal lobe dementia must behave in a different way, I suppose". This is understandable given that neither of them perceived that they had any difficulties requiring medical help. If you perceive no difficulties why would you expect to receive a diagnosis and how would you make sense of it? Patrick repeatedly stated that he "didn't feel any different". For example:

To be perfectly honest, I don't feel any different now than I did before I was diagnosed. I don't do anything different. The only thing that appears to have been a factor is my short-term memory. My long-term memory is fine; I can still remember what I did 20 years ago.

I felt confused and annoyed because I believe that it was the fall and I hit my head on a brick wall to cause the problem. [The doctor] said, 'That was probably just coincidental,' and I probably had frontal lobe dementia before that, that picked it up.

Both Patrick and David used the word "just" when describing the medical assessments undertaken. The use of the word "just" in both cases might reflect their beliefs that there was not a problem, or given their wives were concerned enough to suggest medical intervention, it might reflect the health professionals' wish to downplay the investigations being undertaken in an effort to address the wives' concerns without upsetting the participants. Even if the latter were the case, this downplaying of the medical investigations may have served to reinforce David and Patrick's sense of not having a problem.

In contrast to the three male participants, both Christine and Jayne reported changes that they felt required medical intervention. However, both reported somatic health complaints rather than features typically associated with bvFTD. This shows an ability to recognise some health related changes yet also illustrates why they might be bewildered either by the symptoms or by the medical response to them. Christine said that both she and her husband were concerned about headaches she was experiencing and problems with her memory. These difficulties led her to visit her GP:

Christine: It's because I have these terrible headaches, here [points to right temple], and then it came on to across my forehead [draws figure across forehead from right to left], and that just kind of ... I can't really express it but the headache was just driving me barmy, because I can't understand why it doesn't stop doing it. I couldn't remember a lot of things and that wasn't very good.

Christine hoped that she would be prescribed medication. Since Christine had recently received extensive treatment for cancer this might have reflected her prior experience of contact with health services.

Christine: Well, I went to the doctor's, and the doctor then said, 'You probably need just to take some tablets' to like reduce the pain in my head. And then I went to [specialist's name] because by then it was getting worse, if you know what I mean!

Interviewer: Okay, so taking tablets to reduce the pain wasn't working?

Christine: Wasn't working. So I went to see [name of specialist] and he didn't put me on anything, he didn't put me on any tablets - I don't think he did, I don't think he did, I don't think he ever did.

Jayne, who had worked in a delicatessen during the day and instructed adult fitness classes in the evening, reported that it was her partner and work colleagues who first noticed that she had become slower at doing things. She agreed with them noticing that: "Yeah, I became a lot slower and things... I was doing generally you know like walking a lot slower. Um.. That was it really". She described "having guessed something was wrong" and how she "wanted an answer".

Jayne's responses also seemed paradoxical. Her concern about walking more slowly was inconsistent with her lack of concern about changes in her lifestyle. She talked about previously instructing four evening fitness classes and participating in additional ones each week. In contrast, she no longer does this and her only exercise now was walking the dog around the estate on which they lived. Despite her stating that she "missed it" and that she was "missing the physical activity", there was no sense of concern about the magnitude of the change. Jayne explained "As long as I'm keeping active I don't mind really."

In summary, the theme of ‘Awareness of change: What’s the problem?’ captures participants’ subjective experiences of change and whether they viewed these changes as problematic.

*Threats to self: This is not me*

This theme encompasses participants’ narratives about events which have threatened to negatively impact on their sense of self. This occurs at various levels. At a diagnostic level, some participants struggled with the label of bvFTD whilst for others the threat came from their reduced abilities, or from lifestyle restrictions imposed upon them due to their illness.

In respect of the diagnostic label of bvFTD, for example, Jayne became tearful when discussing her diagnosis. Her wish had been to find out what was wrong yet the diagnosis was overwhelming for her. Jayne’s daughter who lived at home worked locally in a home for older people with dementia and Jayne had visited the home and heard stories about her daughter’s work. Her bewilderment seemed to arise from being unable to process the discrepancy between her previously physically fit 46 year old self who had noticed walking more slowly and her mental model that ‘dementia equals old people.’ She described feeling “Well I’m okay in myself, I’m not odd or nothing. I’m just a bit slower really” and:

Jayne: They could not give a diagnosis at first. And then as the time went on dementia started to be mentioned.

Interviewer: Okay. How did you feel when they first mentioned that?

Jayne: Horrified.

Interviewer: Okay. Can I ask what images came into mind? What did you think?

Jayne: Christ (wells up with tears). That’s for old people.

In contrast, Christine's language when describing her diagnosis was blasé, for example, "I do get this frontal lobe thing" which suggests that she may have seen the condition as transient, like a headache, which would fit with her wish for medication. Christine had recently been offered some medication as part of a medical trial. Her explanation suggests that the receipt of medication helped to protect against the threat to her sense of self which came from the loss of skills and independence rather than from the diagnostic label of bvFTD.

Interviewer: So how did you feel when [the specialist] offered you medication?

Christine: Much happier because it's just like, I mean I can't, and now it's because I've kind of lost, I mean because I was really, when I was at work and things like that, I used to do everything for everybody, I worked for loads of people, everybody. And um, I can still remember going back to those days when I was at the [names past employer], I could do everything, but that suddenly just all disappeared for me.

Christine also talked about her love of driving and how she could not drive anymore: "Well, I can't do anything now because I can't ... I had a car, I can't drive the car now because of this frontal lobe now, I just can't do it" and:

Christine: I couldn't drive because they said, 'There's nothing we can do about your driving.'

Interviewer: And how did you feel when you were told, they advised you not to drive?

Christine: It's because of my head and I couldn't do it, I went there and they just said, 'No, you can't drive.'

Interviewer: So can I ask, did you agree with them?

Christine: Yeah

Her narrative concentrated on the effect that not being able to drive had had on her life. For example: “Well it has really because I can’t do anything, I can’t go out. Well, [partner’s name] takes me out, he always tries to accompany me, but ...” and

I used to go shopping, then we used to go with, we used to all, we used to (talking about two of her sisters) always take it in turns, I used to go up in my car, and then, like do things like that. And then we used to all like take it in, like my sister wanted to come here, she’d come here, then we’d go over to my other sister, we’d go to places like that, which was really nice.

For Patrick and Tom the threats to self occurred in situations where they perceived that control had been taken away from them in relation to aspects of their lives which they particularly valued. The threat to self for them seemed to be related to their roles which afforded them status and a sense of worth, for example:

I’ve lost my job because I was told I couldn’t do my job any longer because of my medical condition, so rather than going to 65, which I wanted to, I had to retire at 62, or 61, which I was annoyed at.

Patrick

Both Patrick and Tom reported missing work saying: “I did love my work, I must confess; I did love my work. I missed it.” (Patrick) and:

Interviewer: Do you want to tell me about the work you did?

Tom: I used to be, or when I say I used to be it sounds like your life’s gone (laughs).

Tom

When Tom was asked “So, what did you feel about finishing work?” he became very tearful and did not reply for several minutes at the end of which he stated “It’s difficult”. He expressed the importance of doing qualifications to “get on in work” and explained that he had

completed an Open University degree in his spare time to help secure promotion. He showed the interviewer a medal he had received for work-related achievements together with a picture of him being presented with this medal by the then Prime Minister. Likewise, Patrick emphasised his multiple degrees and his professional role which he had held for forty three years “man and boy”. He also described how he did the work himself and “never had an assistant”.

Both Tom and Patrick’s feeling of loss of control extended to other areas of their lives. In terms of day-to-day activities, Tom referred to deferring to his wife and feeling he was not given choices about activities:

It’s like [partner’s name] is doing something and you know, this yourself. You just have to be quiet and do the same in a way you wouldn’t be in days gone past. When you’d say I don’t fancy that [laughs]

For Patrick losing his driving licence was difficult but not on a practical level. He explained that getting around by bus was easy for him and explained the local bus routes in some detail. However, at an emotional level it was difficult for him to feel that he had to rely on others especially given that he did not perceive his driving ability to be compromised. He said his wife does not mind driving but he feels it would be “an impertinence for me to expect her to do it”. He also explained: “My driving licence, I’m affected by that, I loved driving and I miss not being able to drive. Nothing I can do about it”. Patrick was aware that his family did not like his driving with his wife telling him that “The children don’t like your driving. They won’t come if you’re driving, they wouldn’t come with you.” He minimised this by explaining that his wife “was a perfectionist”.



In a similar vein, Patrick was annoyed that he had been unable to renew his shotgun licence:

The other thing is, I've got a shotgun certificate and my shotgun certificate was about to expire in about a month's time, and so I applied for another certificate to be sent to me. The chap came out from the [name of force] police in [name of local station] to see me, to look at the guns in the cabinet here, and he put a report in. I had a letter come last week to say that because of my medical condition, they're not renewing my shotgun certificate; that will now go when it expires. The good thing is that the shotgun is still in my possession because my son has a shotgun certificate and I've transferred it to him. That's another thing that annoys me.

Again on a practical basis this was not problematic but affected Patrick due to the perceived loss of control it implied:

Patrick: It doesn't particularly bother me now, the shotgun is up in the cabinet upstairs in my study, and it stays there. I've never ... I've never used it, I haven't used it for about four years, I suppose. You get a shotgun to go rabbit shooting perhaps sometimes, shotguns into the wood. . . .

Interviewer: . . . So when you described earlier it was annoying for you when they wrote to you saying you can't have your shotgun licence, in terms of what you do day to day, it wasn't the sort of thing that you were doing regularly?

Patrick: No. It hasn't affected me at all, it's just I feel a little bit aggrieved that I no longer have a shotgun certificate. If I want to go out and do some shooting with my son, for company, I couldn't now go with my shotgun certificate because I don't have a certificate now so I couldn't use it, so I'll just go with him and just watch what he does. That's rather annoying, but that's academic, I haven't shot for about four or five years anyway.

In summary, the theme of 'Threats to self: This is not me' represents changes that negatively impact on participants' sense of self whether due to the label of dementia, a reduction in abilities, or due to lifestyle restrictions imposed upon the participants due to their illness.

### **Super-ordinate theme: Relationship with others**

This super-ordinate theme reflects the narrative about relationships running through all the participants' stories whether it be at the level of not understanding the effect of their behaviour on family and friends or at the level of difficulties in interacting with people within their wider system.

#### *Family and friends: Things haven't changed... but do I say anything wrong?*

This theme looks at relationships with those with whom it would be expected that participants would have had at least a reasonable social relationship. The sub-theme of 'Family and friends: Things haven't changed... but do I say anything wrong?' really illustrates the paradox between not understanding that their relationships have changed and the vague feeling that something about their interactions with others is not quite as it used to be.

All participants said there were no changes in their relationships with close family and friends. However, this statement was incongruent with three participants' descriptions of feeling that the way people behaved towards them had changed. For example, Tom's relationships with friends had been important to him. He showed the interviewer pictures of himself with groups of friends who he described as "nice friends sort of lovely" and talked about "getting a good bunch of friends together" and "everyone mucking in". However, he described how he now picks up "vibes" from people and is aware that people treat him differently which he described as a "bit, um stressful" and that this is "when you get down". He keeps these feelings to himself and copes by "trying to get out of the situation". He said he did not see friends much anymore but was unable to explain why this might be, or consider whether changes that he had described in

himself might be related to this, leaving him with a sense of perplexity and mild anxiety about why his friendships had faded.

Similarly, Jayne when asked whether her relationship with her two daughters (one of whom lives with her) had changed said:

Jayne: “You know I don’t think so. I’m quieter with them’

Interviewer: Okay. In what way quieter? Can you give me an example?

Jayne: I don’t speak very much to them’.

Interviewer: Is that a change?

Jayne: I think so. Yeah. I like peace and quiet.

Jayne recognised a change in her own behaviour, but showed neither recognition nor concern for how that might feel from her daughters’ perspective. This narrative continues when she describes the relationship with her partner, which she also feels has not changed. Jayne described that “she loves it” when talking about having him home all day (he had recently given up work to care for her). However, she continues by describing him as “really helpful” and saying “He does the cooking and the erm washing and he’s basically taken over everything”, reflecting a practical connection rather than an emotional one. She described how “[My partner] would go, out of his way to make a conversation with me and I am just like, ‘yeah’, ‘no’ (laughs).” Recall that Jayne had requested her partner be present during the interview hence these comments, which imply that she cannot be bothered to make an effort to converse, were made in front of him, which violates social norms. Jayne also talked about how she used to go out a lot with her partner and friends but now “I just can’t be arsed really”, and she minimised the importance of these friends: “They were never like really close, close friends anyway”.

Only one participant made reference to the effect his condition had on his family. However, the emphasis was on their concerns for him, rather than his concern as to how they may have been affected:

Interviewer: I'm wondering, how do you think it's affected your family, all these changes?

Patrick: Well, my family are concerned that I have this dementia, they sympathise with me, but there's nothing they can do for me. It's getting people to accept it, basically. I don't think they think I've changed in any way. I don't believe I've changed, and .....

Patrick

Paradoxically, both Jayne and Christine raised concerns that they might say the wrong thing, which suggests some level of social awareness.

Christine: Um, I mean sometimes I can talk to [partner's name] quite openly, because he's quite good with me, um but um I never knew what to say, never, I never know [emphasised this] what to say and what to do or anything, or ...

Interviewer: So I'm wondering, when you say you don't know what to say, is it that you're not sure what [partner's name] is asking, or whether you worry about saying the wrong thing?

Christine: Yeah, I think that's it, yeah

Interviewer: You worry about saying the wrong thing?

Christine: Mm.

Interviewer: Do you think that happens a lot?

Christine: Um, Yeah, I would say so.

Christine

I listen carefully in case I say the wrong thing or something. It's a thought in the back of my mind that if I was to say the wrong thing I would upset somebody and the conversation would go all flat and quiet wouldn't it.

Jayne

In summary, the theme of 'Relationships with others: Things haven't changed... but do I say anything wrong?' illustrates the participants' difficulties in maintaining social relationships and appreciating the effect their behaviour has on others.

*Coping with threats to self: Blame others or just avoid them*

This theme brings together different ways participants sought to cope with threats to their identity, reflecting two dominant coping styles; first externalising the problem by blaming others, and second avoiding situations where difficulties have been encountered.

For example, Tom externalised the reasons for his job loss. From his perspective it resulted from the unreasonable actions of others. He described having no warning that his performance was unsatisfactory, feeling that he did not know what was expected of him and that his workload was unreasonable: “The first part that I knew about it was when I was called in. And as a result I just had to go with it” and “They’re kidding themselves if they believe you can get just one person to do all that stuff in one job.”

Tom described feeling that once he had been promoted people felt differently about him and perhaps were disrespectful of his position: “People don’t think good things about you. Your name might be there but it does not mean to say they think you are at the top.” Tom also talked about feeling “manipulated”, that people “don’t see who they are talking to” and of not knowing “who’s been a shit to him”. He felt that he had done his best and people from work had put him in this position and had not helped him.

Similarly, Patrick used externalisation to explain difficulties he had experienced securing alternative employment:

I’ve written to the [professional] Society to ask if I can go into private practice by myself, and because the way the [profession] is going at the moment, and [my field] particularly, the [professional] Society has reduced the number of [people] who can do [this work] now, they have to have a franchise, and the [professional] Society has reduced the amount

of money coming into the [system] by 20%. So I've spoken to my friends who are [name's job], who work in private practice, 'Can you give me a job?' And so because the income's gone down by 20%, they can't afford to employ me. I only want to work two days a week, but they can't afford to employ me. So I've written to the [professional] Society to ask if I can go into private practice by myself, and they'll reply to that and they'll decide whether I can or not. They'll probably say no because you have to go into private practice by yourself for three years, generally, worked with others not by yourself, so I, suspect they'll say 'No you can't.'

Patrick gave a detailed descriptive account of failing a driving assessment:

The assessor didn't like that I drove in the nearside line of the carriageway, and I then went to the nearside to turn left. Nowadays they drive and go in the centre of the island. I was taught to drive to the left, a different way. When I was taught to drive, of course, to a crossroads, crossing this way, I went round the back of cars, and now they go in front of the cars; a different way of doing things now. And I have a hearing difficulty, my left ear is particularly difficult, and so he said, 'I want you to carry on in a straight line, I'll tell you to turn right or left.' He told me to turn right, I didn't hear him, and so I carried on. He didn't like that. I explained my difficulties afterwards but it didn't affect it. And then on an island, we came to an island, and there was a car coming to my right, so I was just slowing down, I wasn't going to stop, I was slowing down, but he did an emergency brake. I knew what I was going to do, he obviously didn't, and that annoyed me.

He externalised this failure on two levels. Firstly, he repeatedly stated that "this was how I was taught to drive 45 years ago". Secondly, Patrick's use of language when describing his interactions with the assessors suggested conflict with repeated use of "I" and "them" and expressions such as that was "what they came up with" which was augmented by his stories of repeatedly writing to the DVLA to question the decision to revoke his driving licence and to query the competence of the assessors asking "Are they medically qualified?"

In terms of using the coping style of avoidance, to avoid saying the wrong thing, both Jayne and Christine talked about changes in the way they behaved. Jayne talked about how she "prefers to listen" and "not to join in so much". Whilst, Christine said she let her partner do the

talking because he is better at it. She said that at times she would like to ask questions but does not do so, for example, when describing visiting a health professional:

Christine: Well, I think [partner's name] is much better off than me, he knows what to say, and when he says to me, he says ... I don't because I know that he's so good at talking, but I'm not and I have to like kind of think about it, and then I'll just think, oh I don't know what to say now! (laughs)

In summary, the theme of 'Coping with threats to identity: Blame others or just avoid them' reflects the coping strategies participants use to manage the threats to their identity.

## **Discussion**

This study explored the subjective experiences of people living with a diagnosis of bvFTD. The super-ordinate theme of 'Bewilderment' reflects how whilst all participants showed some awareness, albeit in varying degrees, of changes in their behaviour or lifestyles, they all experienced difficulties in making sense of changes and relating them to their diagnosis and their view of themselves. This is illustrated by the two main themes of 'Awareness of change: What's the problem?' and 'Threats to self: This is not me'. In respect of 'Awareness of change: What's the problem?' it was noticeable that whilst all participants were able to report some changes, albeit not necessarily ones they felt required medical intervention, none of the participants directly reported being aware of personality changes, despite changes in personality being one of the hallmark features associated with bvFTD (Hodges et al., 1999; Rascovsky et al., 2011). This suggests a lack of self-awareness. Indeed, Rankin et al. (2005) demonstrated that self-ratings of people with bvFTD of their personality traits showed less congruency with those of informants than was the case for those with mild Alzheimer's disease or healthy older adults; with people with bvFTD tending to over-estimate positive personality traits such as gregariousness and

extroversion whilst underestimating negative traits such as cold-heartedness and introversion. However, people with bvFTD's description of their personality traits were congruent with their informants' retrospective evaluation of their personality prior to the onset of bvFTD suggesting an impairment in self-awareness. Abu-Akel (2003) suggested that self-awareness involves a three stage process. Stage one entails information being received in parietal structures; stage two involves evaluating incoming information for personal and emotional meaning, processes which he suggests are mediated by the limbic and paralimbic systems; whilst stage three involves executive function such as error-monitoring and identifying discrepancies which rely on the integrity of dorsal medial and dorsolateral pre-frontal brain regions. Abu-Akel (2003) purported that impairment in the paralimbic and dorsolateral pre-frontal regions, which are commonly seen in those with bvFTD, means that the process of self-awareness is disrupted at stages two and three of the process. This fits with the ability of participants to recall and, at some level, report some changes and explain their diagnosis due to the relative sparing of damage to parietal structures early in the disease process. It may also explain why they find it difficult to ascribe that diagnosis to themselves; hence leading to a sense of bewilderment.

Difficulties with self-awareness also link with the second main theme of 'Threats to self: This is not me' which represents changes that negatively impact on participants' sense of self. If people with bvFTD have difficulty monitoring feedback it is understandable that this would impact on their sense of self. What is noticeable is the contrast between the participants' reactions to changes over which they felt a degree of control, e.g. decreased leisure activities; and changes that were imposed upon them, which were outside of their control e.g. losing their jobs, whether they be due to the diagnostic label or due to lifestyle restrictions imposed by others. The results of this study illustrate an important difference between the experiences of people with bvFTD



and those of people with Alzheimer's disease. It has been shown that people with Alzheimer's disease tend to have awareness of their memory difficulties but often attribute them to ageing rather than an illness (e.g. Clare, 2003; Clare et al., 2006; Devlin et al., 2007). In comparison, people with bvFTD are doubly disadvantaged. Firstly, limited awareness of their difficulties with respect to markers for bvFTD makes attributions concerning the possible cause difficult. Secondly, lay-beliefs linking memory difficulties and ageing are common-place (Devlin et al., 2007) and seem to provide people with Alzheimer's disease with a plausible explanation for their difficulties. These lay-beliefs may serve a protective function against threats to their self-identity early in their dementia journey and avoid them becoming overwhelmed (e.g. Harman and Clare, 2006). However, no such lay-beliefs exist to help people with bvFTD for whom age of onset tends to occur prior to retirement age and for whom memory is relatively spared.

Whilst a number of models have been proposed to explain threats to self-identity in people with dementia, these models have focussed on people with Alzheimer's disease (for a review see Cattell & Clare, 2010). As discussed in the introduction, BvFTD is associated with a different profile of cognitive deficits to Alzheimer's disease. This cognitive profile is coupled with changes in personality and/or behaviour early in the disease's progression. One theory of self-identity which can reconcile the difference sources of threats to identity experienced by people with either Alzheimer's disease or bvFTD is self-discrepancy theory (Higgins, 1987; 1989). This theory explicitly identifies patterns of incongruence between different aspects of the self and predicts distress-related emotions based on these patterns. The three different aspects of the self proposed were: the *actual* self which represents how people believe themselves to be; the *ideal* self which represents how a person feels they would like to be; and the *ought* self which represents how a person feels they should be. Incongruence between the actual self and the ideal

self is purported to lead to dejection related emotions such as disappointment and sadness. This fits with the views of “being useless, depression, feelings of loss and feelings of wishing one would rather be dead” expressed by people with AD (Clare et al., 2006, p.764). However, since people with bvFTD often have difficulty identifying changes in themselves or, even if they do, tend not to view them as problematic, they are less likely to identify a discrepancy between their actual self and ideal self. However, self discrepancy theory also considers the three purported aspects of the self from the viewpoint of a significant other. Higgins (1987; 1989) purports that discrepancies between the actual self (from the viewpoint of the individual) and the ought self (from the perspective of a significant other) will lead to feelings of fear, being threatened and resentment. Furthermore this theory predicts that the larger the magnitude of the discrepancy, the greater the magnitude of the associated feelings. Rankin et al. (2005) in their study of self-awareness in people with bvFTD demonstrated an incongruence between how people view themselves and how they are viewed by others which may explain the threats to self experienced by some participants in this study.

The second super-ordinate sub-theme of ‘Relationship with others’ illustrates the difficulties participants have relating to others, whether within the context of family and friends or people within their wider network. The first main theme of ‘Family and friends: Things haven’t changed... but do I say anything wrong?’ illustrates the paradox between participants’ narratives that their relationships had not changed; yet their awareness that people’s behaviour towards them had changed which resulted in some participants feeling that they might be doing something wrong. In terms of emotional responses, participants displayed some emotions when describing events that directly appertained to them, but not in relation to their interactions with others. These subjective experiences are consistent with the findings that people with bvFTD,

rather than showing a global impairment in the expression of emotion are able to express simple emotions such as happiness, sadness and fear (Levenson & Miller, 2007; Werner et al., 2007) but are impaired on what Sturm, Rosen, Allison, Miller and Levenson (2006) and Sturm, Ascher, Miller and Levenson (2008) referred to as “self-conscious emotions” such as embarrassment, fear and shame. They hypothesised that these emotions are more cognitively complex and require an understanding of social interactions. The experiences are also congruent with the identified deficits people with bvFTD display on Theory of Mind tasks (Fernandez-Duque, Baird, & Black, 2007; Gregory et al., 2002; Snowden et al., 2003) which neuro-imaging studies have shown to be mediated by a neural circuit including the frontal lobes. In a similar vein, people with bvFTD have been found to have an impaired ability to recognise emotions, particularly negative emotions in others (Rankin et al, 2006; Werner et al., 2007) which might be reflecting in some participants’ experiences of feeling that they might be doing something wrong. Finally, neuro-imaging studies have shown that integrity of the frontal lobes is important for empathy; which has also been found to be impaired in people with bvFTD (e.g. Vollm, et al., 2006). This suggests that people with bvFTD would have difficulty with inter-personal relationships, which is reflected in the participants’ experiences, where it was seen that some participants had difficulty in seeing things from others’ perspectives.

This links with the second main theme of ‘Coping with threats to self: Blame others or just avoid them’. Blaming others is conceptualised as a fight response and a response to feelings of injustice, and in these participants may have arisen from difficulties in being aware of changes. Here, to protect the sense of self, others are blamed for the situation that participants find themselves in. This is an understandable response given that participants’ who reported a tendency to cope in this way did not agree that there were valid reasons for restrictions on their

preferred lifestyle. In contrast, avoidance is conceptualised as a flight response where participants cope by avoiding situations thus protecting themselves from further assaults on their self-identity. This way of coping tended to be adopted by participants who had an inkling that they may be acting in ways that were socially inappropriate.

These two coping strategies have strengths and limitations in respect of effectiveness. The fight response of blaming others, gives participants a sense of power through taking action. Whilst understandable on a cognitive level, i.e. if you are unaware of a change then it follows that the problem must be due to something external, the failure to recognise change coupled with a reduced capacity to recognise the reciprocity needed to build and maintain relationships is likely to render this strategy unsuccessful. Rather than achieving the aim of regaining control, the more likely outcome is that this will antagonise and may result in increasingly more punitive responses from others; thus reinforcing and intensifying the feelings of injustice when the participants' efforts are thwarted. Indeed, this explanation fits with informant reports that people with bvFTD have a tendency towards negative personality traits such as coldness (e.g. Rankin et al., 2005). The second coping strategy of avoidance or fleeing, either by talking less or letting others talk for them, may help reduce instances of faux pas. However, this social withdrawal may be misconstrued as rudeness or disinterest (Rankin et al., 2005) leading to a negative reaction from others which the person with bvFTD will struggle to understand hence further exacerbating their already problematic social relationships and increasing levels of family stress (e.g. de Vugta et al., 2006; Merrilees et al., 2012).

In terms of implications, this study raises an interesting question about how mental capacity is assessed (Mental Capacity Act, 2005). Under the Act, capacity to make a decision is

assumed unless there is evidence to the contrary. If people with bvFTD, at least in the early stages of the disease, show a cognitive understanding of the decision to be made it is possible that their struggle to process information at an emotional level may be overlooked. This suggests that there may be situations where people are deemed to have capacity even though emotionally they may have limited ability to understand the possible impact of their decision. Therefore, this should be considered when assessing capacity and provision made for supporting people in the decision-making process.

In terms of interventions, it is argued that people with bvFTD require specialist interventions tailored to meet their particular needs. Currently the guidelines on best practice in dementia care (National Institute for Health and Clinical Excellence, 2011) encompass all forms of dementia and recommend group-based cognitive stimulation therapy for treating the cognitive symptoms of people with mild to moderate dementia. For people with bvFTD, for whom, at least earlier in their dementia journey, cognitive symptoms are not so troublesome; this type of therapy may not hold the same benefits as for those people with a diagnosis of Alzheimer's disease or vascular dementia. Interventions are required which seek to reduce the impact of their difficulties with social interactions. For example, future research could look at the use of family based interventions (which include the person with bvFTD) with a psycho-educational component. These have been demonstrated to have efficacy in other groups for whom social relationships are difficult, e.g. people with schizophrenia (Pitschel'Wcdz, Leucht, Bduml, Kissling, & Engel, 2001).

One limitation of this study is the potential increase of bias arising from adjustments made from the standard way interviews are routinely conducted in qualitative research. These

adjustments were made in order to minimise the impact of the communication difficulties such as poverty of speech routinely observed in this client group. So the risk of bias, when designing the study, was balanced against the need to give participants appropriate support to maximise their opportunity for their voices to be heard. A further difficulty with the process of triangulating elements of the interviews, where issues had been revisited to reduce this potential bias, was that the quotes obtained were more brief and fragmented than those which might be obtained from individuals without communication difficulties, hence making synthesis of their personal accounts challenging. Also, it is acknowledged that the interpretation of the participants' experiences is reliant on the reflectivity of the researcher; despite the safeguards employed to reduce biases arising from this. The researcher has a passion for working with people with dementia and championing their viewpoint which may have impacted on the interpretations made. A further limitation of this study is that the results cannot be generalised, due to the methodology chosen which considers the experiences of small numbers of participants. The decision to use this methodology, whilst appropriate, was driven primarily by the objective of seeking to understand the experiences of those with bvFTD. However, it also reflects a degree of pragmatism. BvFTD is a relatively rare sub-type of dementia and engaging people with this diagnosis in research is difficult. Nevertheless, given that this area is under-researched it is hoped that this study will lead to more research in this area.

In conclusion, this study has demonstrated that engaging people with bvFTD in conversations about their subjective experiences is possible. The results show how hard it is for those who have a limited awareness of changes that impact on their interpersonal relationships

and illustrates ways that people with bvFTD try to make meaning of their situation and the coping mechanisms they employ.

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Search for review papers in PsycInfo

Appendix 1

Search for review papers in Medline

Search for review papers in Web of Knowledge

Appendix 3

Search for papers in PsycInfo



Search for papers in Medline

Search for papers in Web of Knowledge

Journals searched for in-press articles

Aging & Mental Health  
American Journal of Alzheimer's Disease and Other Dementias  
Journal of Clinical and Experimental Neuropsychology  
Neuropsychiatric Disease and Treatment  
Neuropsychological Rehabilitation: An International Journal  
Neuropsychology  
Psychogeriatrics

## Data Extraction Form

First author	Journal	Year
STUDY TYPE		
AIMS (1)		
HYPOTHESES (1)		
Inclusion/Exclusion criteria		

Participant characteristics				
	Intervention			Controls
	EL	Other	Other	
If other please specify: EF, SP, VC				
Number of participants (3)				
Gender (numbers / %, etc) (3,5)				
Diagnosis (if applicable) (3)				
Severity of AD inc how measured e.g. MMSE score (3,5)				
If single case, overview of case history (1)				

<b>Method</b>	
Description of recruitment source (3,11,21)	
Describe time-frame for recruitment for all groups (22)	
Description of how people were selected to be approached, e.g consecutive patients, random sampling (11)	
% of people approached who consented to participate (12)	
Describe how participants were allocated to different groups (23)	
Describe assessment measures used to control for possible extraneous variables between groups (5)	
Describe evidence that sample is representative of population (12)	
Specify no of participants followed up (N,%) (9,26)	
Were participants blind to the intervention they received? If yes, describe how this was achieved (14,24)	
Were staff recruiting participants blind to the group allocation process? If yes, describe how this was achieved (24)	
Were outcome assessors blind to the intervention participants had received? If yes, describe how this was achieved (15)	
Power calculation (27)	
Within/between subjects etc	

Task	
No of sessions/length of sessions	
Setting, e.g home or clinic. State whether representative of setting where learning will be most applied (13)	
Practice between sessions	
Details of carer involvement in intervention	
Description of errorless learning (EL) method (4)	
Description of other cognitive rehab methods used. Specify whether used as augmentation or as comparison (4)	
Description of intervention procedure. Specify whether standardised and whether there is evidence that this procedure was not followed (4, 19)	
Description of 'to be learnt' materials. Specify whether related to participant goals	
Specify outcome measures used and whether valid and reliable. Where in the paper were these described? (2,20)	

<b>Analysis</b>	
Specify statistics used; including adjustments for confounding variables (if applicable) (18,25)	
Summarise main findings (6)	
Specify descriptives reported e.g means, medians (6)	

Specify variations in data reported e.g. SD, inter-quartile range (as appropriate). (7)	
Describe any unplanned analysis and whether clearly identified in paper (16)	
Specify follow up period and whether consistent across participants (17)	

<b>Conclusions</b>
<b>Limitations</b>
<b>Additional Notes</b>

Quality criteria for group studies before review







## Amendments made to quality criteria for group studies

- i. Question 8 concerning adverse outcomes was removed since it appeared more appropriate for intervention involving medication or invasive procedures.
- ii. Question 9 Have the characteristics of patients lost to follow-up been described?;  
low n, few long term follow-up
- iii. Question 10 actual probability rather than  $>.05$ ; as not being a great threat to validity
- iv. Question 14: Was an attempt made to blind study subjects to the intervention they have received? Since only one study was between-subjects, this question was deemed not applicable.
- v. Question 17: In trials and cohort studies, do the analyses adjust for different lengths of follow-up of patients, or in case-control studies, is the time period between the intervention and outcome the same for cases and controls? No studies had different follow-up periods
- vi. Question 21: Were the patients in different intervention groups (trials and cohort studies) or were the cases and controls (case-control studies) recruited from the same population? Since only one study was between-subjects, this question was deemed not applicable.
- vii. Question 22: Were study subjects in different intervention groups (trials and cohort studies) or were the cases and controls (case-control studies) recruited over the same period of time? Since only one study was between-subjects, this question was deemed not applicable.

- viii. Question 23: Were study subjects randomised to intervention groups? Since only one study was between-subjects, this question was deemed not applicable.
- ix. Was there adequate adjustment for confounding variables in the analyses from which the main findings were drawn? Since only one study was between-subjects, this question was deemed not applicable
- x. Question 26: Were losses of patients to follow-up taken into account? low n, few long term follow-up
- xi. Question 27. The criterion in relation to power was simplified to: Was a power calculation undertaken and was the sample actually recruited in line with this calculation?
- xii. Three options were considered for each question: yes, no or unclear rather than the dichotomous yes/no criteria adopted by Downs and Black (1989)
- xiii. An additional question was added under internal validity. Is the methodology used suitable to meet the aims of the study, i.e. does the study investigate what the authors set out to investigate?

## **LIVING WITH A DIAGNOSIS OF BEHAVIOURAL-VARIANT FRONTOTEMPORAL DEMENTIA: THE PERSON'S EXPERIENCE**

This paper describes a qualitative study conducted by Julie Griffin as part of a thesis submitted to the School of Psychology, University of Birmingham for the Doctorate in Clinical Psychology.

### **Background and Aims of the Study**

Behavioural-variant frontotemporal dementia (bvFTD) is a relatively rare, progressive, degenerative condition resulting from damage to the brain (Neary et al., 1998). Unlike more common forms of dementia such as Alzheimer's disease (AD) which is associated with progressive memory loss, bvFTD (at least in the early stages) leads to personality and behavioural changes with memory being relatively preserved (Hodges et al., 1999).

Understanding people's views of their difficulties is important since it helps ensure that Health Service provision is sensitive to people's needs and preferences. Whilst the views of those with mild AD have been sought on a variety of issues relating to their difficulties, such as coping (e.g. Clare, 2002), this research has not been extended to those with bvFTD; perhaps because it has been assumed that people with bvFTD lack insight into their difficulties making obtaining their views problematical (e.g. Rankin, Baldwin, Pace-Savitsky, Kramer, & Miller, 2005). Therefore, this study sought to elicit the views of people with bvFTD on their experience of living with their diagnosis.

## **Method**

Five people diagnosed with bvFTD were interviewed about their experiences. The interviews were semi-structured, which allowed participants the freedom to tell their own story in their own way. After transcription, the interviews were analysed using Interpretative Phenomenological Analysis (IPA) (Smith, Flowers, & Larkin, 2009). IPA was used since this technique focuses on the meaning people make of their subjective experiences rather than 'objective facts'. The researcher's role was to make sense of these subjective experiences by developing a hierarchy of themes.

## **Results**

Two overarching themes were identified. The first theme of 'Bewilderment' reflected the participants' feelings from the start of their dementia journey. It was divided into two main themes. Firstly, 'Awareness of change: What's the problem?' which captured the participants' awareness of change and whether they viewed these changes as problematic. The second main theme, 'Threats to self: This is not me' encapsulated changes due to events, such as receiving a diagnosis, or in lifestyle, e.g. the loss of their job that were incompatible with how participants viewed themselves.

The second overarching theme of 'Relationships with others' reflected the difficulties participants have with maintaining relationships. The first main theme of 'Family and friends: Things haven't changed.... but do I say anything wrong?' reflected the paradox between not recognising that their relationships have changed and the vague feeling that something about their interactions with others was not quite as it were. The second main theme of 'Coping with threats

to self: Blame others or just avoid them'' brought together the two main ways participants sought to cope; either by blaming others, or by avoiding social situations.

## **Conclusions**

Whilst the people with bvFTD in this study were able to report some changes in their behaviour or lifestyle that either they or family members had noticed, not everyone felt these changes warranted medical intervention. Furthermore, no one directly reported being aware of personality changes which is a hallmark feature of bvFTD; suggesting a lack of self-awareness. This finding is in line with other studies which have demonstrated that people with bvFTD self-reported personality traits are significantly more positive than those given by family members (Rankin et al., 2005). As they are unable to recognise personality changes that will have played a part in their diagnosis and hence the reasons for the loss of things, such as jobs, which are important in terms of their feelings of self-worth, it is understand that they might feel bewildered.

Difficulties with self-awareness also link with the second overarching theme of 'Relationships with others' whether that be family and friends, or people within their wider network. Here, participants have difficulty reconciling their beliefs that nothing had changes with feelings that something had changed but not really understanding why. Furthermore, their attempts to cope either by blaming others, or avoiding social situations whilst understandable tends to compound their problems with their actions being misconstrued as perhaps aggression or rudeness.

The subjective experiences of the people within this study are in line with the difficulties associated with bvFTD. These include a reduction in self-awareness, empathy and the lack of ability to see the viewpoint of others due to progressive damage in an area of the brain called the

frontal lobes (Abu-Akel, 2003; Fernandez-Duque, Baird, & Black, 2007). Damage to this brain area is associated with difficulties in inter-personal relationships, which is reflected in the participants' experiences. Also, in contrast to people with Alzheimer's disease, people with bvFTD are often unaware of their difficulties in this area which makes understanding the responses they receive from others particularly hard for them.

People with bvFTD require specialist help to meet their particular needs which, at least early in their dementia journey, are distinct from those of people with more common forms of dementia where the focus is on minimising the impact of memory difficulties. For example, family based interventions (which include the person with bvFTD) which have helped other groups for whom social relationships are difficult, e.g. people with schizophrenia (Pitschel-Wecz, Leucht, Bduml, Kissling, & Engel, 2001).

In conclusion, this study has demonstrated that people with bvFTD can talk about their views and experiences, and the results have shown how difficult it is for them to make sense of things which threaten their sense of self and their difficulties maintaining social relationships due to their difficulty understanding that their personality and hence emotional responses have changed.

## **References**

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*Interview Schedule: Living with a diagnosis of frontal variant fronto-temporal dementia (fvFTD): The persons' experience*

*(Version 1 dated 14/10/10)*

Notes for the researcher:

- This document is a guide. It is intended to be used flexibly to allow each participant to tell their own story in their own way.
- It lists subject areas to discuss and a possible order [with prompts in brackets].
- In order not to overburden participants it is envisaged that a series of short interviews will be undertaken to cover the suggested topic areas.
- It also includes a list of information at the end that if not covered elsewhere may be useful to obtain.

*Life history*

- 1) Can you tell me a bit about your life? [What were you like as a child? What sorts of things did you like to do? And what about after you left school? What job did you go into? What hobbies and interests did you have?]
- 2) How would you describe yourself as a person? [What sort of person?-reserved, 'life and soul of the party', happy, moody, worrier?]

*Day- to- day life*

- 1) Can you tell me about your daily life now? [What sorts of things do you like to do?]
- 2) Do you think things have changed? [What has changed- interests, habits, likes and dislikes, relationships with friends and family?]
- 3) Do other people think you have changed?

*Effects on life*

- 1) Do you see your role differently now? [What is different? How do you feel about these changes?]
- 2) How do you cope? [come to terms with the situation?]
- 3) How do you see your life in the future?

*Health*

- 1) What led up to you seeking help from the Working Age Dementia Service? [What were your concerns? Family concerns? ]

2) What support do you have now? [What support did you get/not get, what would have been useful?]

### **Diagnosis**

1) Have you received a diagnosis? [When did you get it? Who gave it? Is it useful?]

2) What do you understand about the diagnosis? [What does it mean to you? What words come to mind? What images? Do you have a nickname for it? How do you feel about it?]

### **Other areas**

1) Is there anything else you think is important in understanding how you feel that we have not talked about?

*Check that the following has been covered during the interview:*

- Age of patient
- Education & employment / occupational history
- Family – children – ages, living at home? Regularity of contact?
- Spouse – together, separated, divorced, widowed?



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**Participant Information Sheet (version 2, dated 18.2.11)**

*Title: Living with a diagnosis of frontal variant fronto-temporal dementia (fvFTD): The person's experience*

*Researcher: Dr Julie Griffin*

You are being invited to take part in a research study. The information given below explains why this research is being done and what it will involve. Please take time to read the following information carefully and discuss it with others if you wish. Please ask if anything is not clear or if you would like more information. Take time to decide whether or not you wish to take part.

Thank you for reading this

**What is the purpose of the study?**

This study is interested in your experiences of living with a diagnosis of frontal variant fronto-temporal dementia. The study is being completed as part of Dr Julie Griffin's thesis for her Doctorate in Clinical Psychology.

**Why have I been chosen?**

You have been invited to take part as you have been diagnosed with this difficulty.

**Do I have to take part?**

It is up to you to decide whether or not to take part. If you decide to take part an initial meeting will be arranged with Dr Julie Griffin (the researcher) who will answer any questions you have and ask you to sign a consent form.

If you decide to take part you are still free to withdraw at any time until the interviews are analysed without giving a reason. A decision to withdraw, or a decision not to take part, will not affect the standard of care you receive currently or at any point in the future.

### **What will happen if I take part?**

Dr Julie Griffin (the researcher) will usually arrange to visit you at home to interview you about your experience of living with a diagnosis of frontal variant fronto-temporal dementia. However, if you prefer, you can meet with her at the Department of Psychology, Birmingham University. In this case transport will be arranged for you (and a member of your family if you wish) or your travel costs will be reimbursed.

### **What will I have to do?**

The interview will take approximately 1 ½ to 2 hours depending on how much you wish to talk about your experiences. You can decide whether to take part in one interview session or several shorter interview sessions. You will not have to decide immediately whether you would prefer one or several interview sessions. Rather, you can express an initial preference but will be free to change your mind during the interview, e.g. if you start to feel tired or, if alternately, you decide that you are able to manage a longer session.

With your permission the interview(s) will be recorded in order to help me to remember what you have said.

### **What are the possible disadvantages and risks of taking part?**

This study involves discussing your personal experiences of living with a diagnosis of dementia which potentially might be upsetting for you. If you find talking about your experiences upsetting you may stop the interview at any time. You can discuss any upsetting issues in confidence with the researcher. Alternatively, the researcher will provide you with contact details of someone independent of the research study who you can talk to.

Please note that the results of the study will include direct quotes from participants. However, as part of the analysis all identifying information will be removed and participants will be given fictitious names, therefore no individual will be identifiable. If you would like to, you can review the direct quotes I would like to use before they are included in the final report.

**What are the possible benefits of taking part?**

There are no direct benefits of from taking part in this study, although it is hoped that the study may help us to better understand peoples' experiences of living with a diagnosis of frontal variant fronto-temporal dementia.

**What happens when the research study stops?**

The results of the research study will be included as part of Dr Julie Griffin's thesis for her Doctorate in Clinical Psychology and will also be published in psychology journals. A copy of the thesis will be available at the Main library in the University of Birmingham.

If you would like a summary of the study's results, a copy will be posted to you.

**What if there is a problem?**

If you any concerns during the study you can discuss them in confidence with the researcher. Alternatively, the researcher will provide you with contact details of someone independent of the research study who you can talk to.

You can also talk to a member of the NHS team responsible for your care.

**Will my taking part in the research be kept confidential?**

Whilst with your permission your GP will be informed you are taking part in a research study, the results of the research will be kept confidential. However, parts of the data may be made available to the NHS team responsible for your care but only if any issues of risk to your safety are disclosed during the interview.

**Contact for further information**

If you require further information or you have any concerns please do not hesitate either of the people named below:

[Redacted]	[Redacted]	[Redacted]
[Redacted]	[Redacted]	[Redacted]

This patient information sheet is for you to keep.

**CONSENT FORM** (version 2, dated 18.2.11)

*Title of Project: Living with a diagnosis of frontal variant fronto-temporal dementia (fvFTD): The person's experience*

Researcher: Julie Griffin

Please initial box

1. I confirm that I have understood the information sheet dated 18.2.11 (version 2) for the above study. I have had the chance to consider the information, and any questions I asked have been answered.
2. I understand that my participation is voluntary and that I am free to withdraw at any time during the research interview, without giving any reason, without my medical/social care or legal rights being affected.
3. I understand that the research interview will be audio-recorded.
4. I understand that the data collected during this study will be looked at by the researcher and two colleagues at the University of Birmingham to ensure that the analysis is a fair and reasonable representation of the data. Parts of the data may also be made available to the NHS team responsible for my care but only if any issues of risk to my safety should be disclosed during the interview.
5. I understand that direct quotes from my interview may be published, but that my name will not be attributed to any such quotes and that I will not be identifiable.
6. I give permission for my GP to be informed that I am taking part in this study.
7. I agree to take part in the above study.

.....  
Name of participant                      Date                      Signature

.....  
Name of researcher                      Date                      Signature

[Redacted]

[Redacted]

[Redacted]

[REDACTED]

