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RESEARCH ARTICLE

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Protocol for the Rare Dementia Support Impact study: RDS Impact

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Objectives: The Rare Dementia Support (RDS) Impact study will be the first major study of the value of multicomponent support groups for people living with or supporting someone with a rare form of dementia. The multicentre study aims to evaluate the impact of multicomponent support offered and delivered to people living with a rare form of dementia, comprising the following five work packages (WPs): (a) longitudinal cohort interviews, (b) theoretical development, (c) developing measures, (d) novel interventions, and (e) economic analysis.

Methods: This is a mixed-methods design, including a longitudinal cohort study (quantitative and qualitative) and a feasibility randomised control trial (RCT). A cohort of more than 1000 individuals will be invited to participate. The primary and secondary outcomes will be in part determined through a co-design nominal groups technique prestudy involving caregivers to people living with a diagnosis of a rare dementia. Quantitative analyses of differences and predictors will be based on prespecified hypotheses. A variety of quantitative (eg, analysis of variance [ANOVA] and multiple linear regression techniques), qualitative (eg, thematic analysis [TA]), and innovative analytical methods will also be developed and applied by involving the arts as a research method.

Results: The UCL Research Ethics Committee have approved this study. Data collection commenced in January 2020.

Conclusions: The study will capture information through a combination of longitudinal interviews, questionnaires and scales, and novel creative data collection methods. The notion of "impact" in the context of support for rare dementias will involve theoretical development, novel measures and methods of support interventions, and health economic analyses.

KEYWORDS
dementia, dementia support groups, Rare Dementia Support, young-onset dementia

1 | INTRODUCTION

"Rare dementias" are forms of dementia characterised by progressive difficulties with cognitive symptoms other than memory and/or occurring before the age of 65 (www.raredementiasupport.org).[1] Non-Alzheimer or vascular causes may account for up to 25% of all cases of dementia, but prevalence and incidence data are only available for some rare dementias (eg, frontotemporal dementia [FTD]: 15-22/100 000 and 2.7-4.1/100 000, respectively[2]), and the picture is complicated by the clinical and pathological heterogeneity of conditions (eg, approximately 80% of cases of the visual dementia posterior cortical atrophy [PCA] are caused by [atypically distributed] Alzheimer disease[3,4]). The rare dementias are proportionately more common in individuals under 65 years old, but are by no means constrained to this age group alone.[1]

Diagnoses of rare dementias are often delayed (eg, 30% will have received a prior incorrect psychiatric diagnosis[5]), and an early-onset (≤65 years of age) diagnosis raises additional challenges relating to individual and family transitions (eg, work, retirement planning, and care transitions[6,7]). Post diagnosis, many find that existing health, social, and voluntary services do not cater adequately for their individual needs, and more specifically that established support groups are often not particularly relevant to their situation owing to significant differences compared with other group members in age, life situations, and symptoms. Despite high numbers nationally, there is not the density of service needed within most regions to make independent local condition-specific support groups viable.

The term "support group" is used variably, with groups varying in structure, duration, and facilitation (professional, lay, or both). However, a defining feature, as operationalised for systematic reviews,[8] is the opportunity for people with or caring for someone with dementia to communicate and interact socially, irrespective of content (exchanging ideas, emotional support) or form of contact (face-to-face, phone, online).

A recent review of peer support models for dementia concluded that while multicomponent support interventions improve carer well-being in the wider population with dementia, the factors mediating this are unclear.[9] The PM's Challenge Implementation Plan[10] also states that evidence from the National Institute for Health Research (NIHR)'s peer support network (PSN) demonstrator site scheme[11] shows "support for carers can have a positive impact in reducing or delaying people diagnosed with dementia entering residential care" (p. 33). Computer-mediated interventions for informal carers may also have positive effects upon levels of carer burden/stress, depression, and anxiety[12] opening up new technological avenues for connecting with carers in situations where/when attendance at face-to-face meetings may not be practical. Dementia support groups may also yield a social value greater than the cost of investment (£1.17-£5.18 per £1 investment[13]), with benefits reported for people living with dementia (eg, mental stimulation and a reduction in loneliness), their carers (eg, reduction in stress and burden of care), and the volunteers facilitating the sessions (increased knowledge). Other positive outcomes reported for people living with a dementia (PLWD) include a reduction in depression and improved quality of life and self-esteem,[14] and identification with others, commonality of experience, and reciprocity of support.[15]

The current project capitalises on the collective experiences of more than 1000 members of Rare Dementia Support (RDS). A vibrant network of six condition-specific support groups, hosted by University

Key Points

- The RDS Impact project will be the first major study of the value of multicomponent support groups for people living with or supporting someone with a rare form of dementia.
- The study will capture information through a combination of longitudinal interviews, questionnaires and scales, and novel creative data collection methods.
- More than 1000 individuals located across the United Kingdom and internationally who are members of Rare Dementia Support will take part in the project.
- The project will explore the impact of multicomponent support groups through five areas of enquiry

TABLE 1 A description of the conditions supported by Rare Dementia Support

Support Group	Description of Condition
Familial Alzheimer disease (FAD)	An inherited form of typical Alzheimer disease, caused by a faulty gene—affecting people as young as 30
Frontotemporal dementia (FTD)	A group of dementias predominantly affecting behaviour and personality.
Familial frontotemporal dementia (fFTD)	A group of dementias predominantly affecting behaviour and personality.
Posterior cortical atrophy (PCA)	A progressive condition predominantly affecting visual and spatial perception.
Primary progressive aphasia (PPA)	A group of dementias predominantly affecting language skills such as comprehension.
Dementia with Lewy bodies (DLB)	A less common form of dementia that is closely related to Parkinson disease, predominantly affecting movement and may include visual hallucinations.

College London (UCL) and attracting 60 new referrals per month, RDS connects individuals affected by FTD (established in 1994), primary progressive aphasia (PPA; est. 2005), PCA (est. 2007), and the directly inherited conditions familial Alzheimer disease (FAD; est. 2010), familial FTD (fFTD; est. 2011) and dementia with Lewy bodies (DLB; est. 2018) (Table 1).

In addition to London-based, condition-specific meetings, there are Carer and Bereaved carer groups (for carer members from across the six conditions) and 26 regional groups from Cornwall to Mersey. RDS also has a widespread international membership, including members in Singapore, China, United States, Australia, and New Zealand. Each group is multicomponent (capitalising on experiential and professional knowledge), open to all (PLWD, current and former carers [at standard meetings and dedicated Bereaved carer meetings]), ongoing (three to four meetings per year), multipurpose (psychoeducation and emotional support), and multiformat (opportunities for PLWD and carers to participate together and separately; mix of whole group talks/Q&As average N = 40-80 members] and small group [N = 8-10] discussions; face-to-face meetings plus phone/email support and regional meetings). Meetings and newsletters are shaped by members and the groups also provide opportunities to inspire, influence, and participate in research. Unlike short-term groups and interventions, the groups ensure continuity over years and even decades. This continuity reflects a culture and community of care, developing and sharing collective knowledge about what works and when, retaining institutional knowledge through many-to-many rather than solely one-to-one relationships in a manner robust to staff and membership changes. RDS provides a stable familiar platform of support that is accessible in different ways over the long courses of people's dementia journeys.

This study will investigate the impact and reach of multicomponent support offered and delivered to people living with a rare form of dementia through five work packages (WPs) (see Boxes 1 and 2). The 5-year study (2019-2023) is led by UCL alongside collaborators from Bangor University, Wales, and Nipissing University, Canada.

2 | METHODS AND STUDY DESIGN

The study is primarily a longitudinal mixed-methods investigation, with some additional cross-sectional evaluations for theoretical and measure development (see Box 2). Data collected from WP1 in the longitudinal interviews will inform and contribute to analyses in WPs 2 to 5. A feasibility RCT will be carried out in WP4 to develop and test novel online forms of support for people living with, caring for, or working with someone living with a rare dementia.

2.1 | Participants

The main study population for RDS Impact will comprise more than 1000 individuals located across the United Kingdom and internationally who are members of RDS who have opted in to our membership database and, in doing so, have expressed an interest in hearing about research opportunities. These individuals include people living with a diagnosis of a rare form of dementia (eg, fFTD, FTD, PCA, DLB, FAD, PPA, and young onset dementias), people supporting or caring for people living with a rare form of dementia (eg, relatives, friends, and professional carers), and professionals who work with or have a professional interest in people affected by a rare dementia.

2.2 | Predicted sample size

Approximately 92% of RDS members either live with a diagnosis of a rare dementia or alongside someone with a rare form of dementia. All will receive an invitation to take part in WP1. Based on the high level of engagement from members in previous research studies, we anticipate a high response rate. In addition, we note that while individuals may sign up to become a member of RDS, this membership may represent a wider support network of relatives,

Research evidence and theory

Generate a theoretical model of - and the world's first evidence base demonstrating the critical role and significant added value of - multicomponent support groups in providing continuous, sustainable and appropriate support for people affected by rare dementias throughout and beyond the course of their dementia

Education, engagement and communication

- Create and use free, accessible platforms for sharing research outcomes such as user-led social media networks (e.g. condition-specific Facebook group) and official support group sites.
- Collaboratively write, translate and disseminate condition-specific information and resources with/through national dementia organisations.

Service delivery

Initiate, launch, equip and facilitate new theoretically-informed regional, national and international support groups and networks.

BOX 1 RDS Impact study objectives [Colour figure can be viewed at wileyonlinelibrary.com]

friends, and professionals affiliated with the person living with a rare dementia, and we will encourage RDS members to circulate the research invitation to their relatives. To this end, if we approximate that each member represents a dyad (at minimum) and take the predicted response rate into account, we predict our overall sample size to increase accordingly. A large sample size facilitates well-powered subanalyses in a group where research is traditionally hampered by underpowered studies, with consequent potential for type-II error.

Other potential participant groups who will be contacted will comprise individuals who are affiliated with collaborating sites at Bangor and Nipissing Universities by virtue of engaging in regional or international support groups and/or by taking part in previous research undertaken by the institutions.

Individuals who are participating in the study with a diagnosis of dementia will have capacity to consent from the outset.

2.3 | Sampling approach

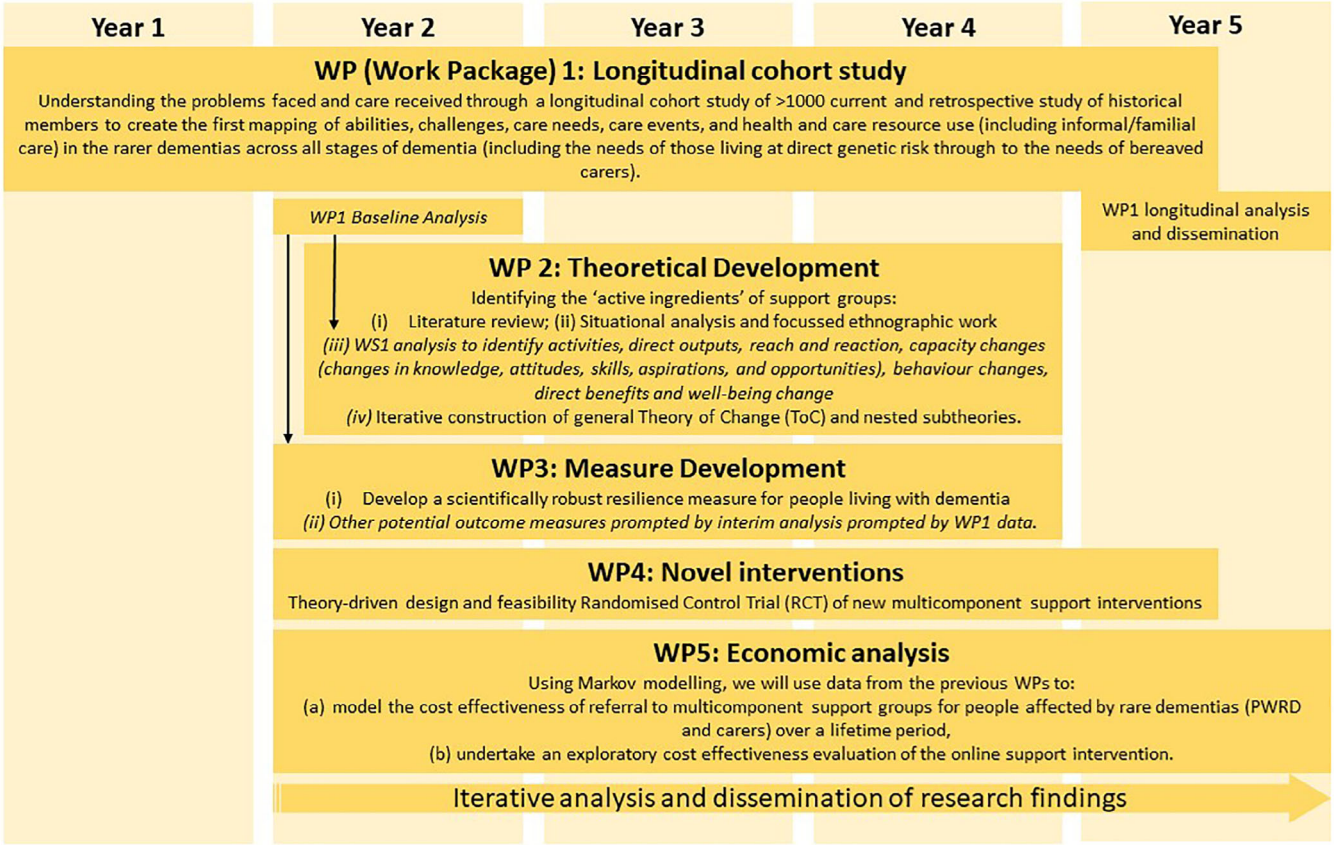
Purposive sampling will take place throughout the sampling and subsampling process for WPs 2 to 4 in order to achieve as broad a sample as possible (eg, incorporating different backgrounds and different diagnoses of rare dementias). Participants' preferences will be taken into account in accordance with the participants' convenience and will be considered in line with the time commitment required to avoid overburdening.

The majority of participants in this study will be recruited for biannual interviews (as part of WP1) but may specify a wish to take part in the other work packages over the course of the study (see Box 2).

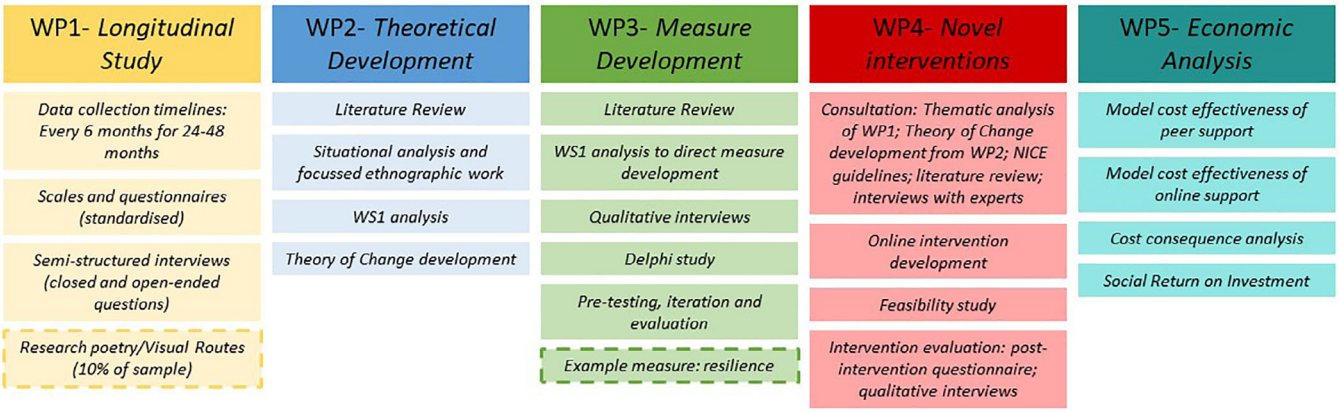
2.4 | Methods

2.4.1 | Work package 1—longitudinal cohort study

This WP comprises a combination of cross-sectional and longitudinal semistructured telephone/virtual (and, where appropriate, face-to-face) interviews to establish the experiences and access to support in relation to the diagnosis of a rare dementia (Box 3). Researchers will gather quantitative and qualitative health and membership demographic data. A range of standardised measures will be used to characterise interview themes (eg, health and care service use, resilience, personal difficulties, and health/functional status). The interview constructs and measures will be derived based on the findings of a preliminary consensus exercise involving RDS members as co-researchers using the nominal groups technique.[16] The interviews will also gather data to inform WP3 (measure development) and understanding current member use of online support to inform WP4 (eg, motivation to use the internet, access, and/or barriers etc.)



BOX 2 RDS Impact: timeline and methodological outline for each work package [Colour figure can be viewed at wileyonlinelibrary.com]



BOX 3 Breakdown of work package methodologies (see Section 2) [Colour figure can be viewed at wileyonlinelibrary.com]

We will evaluate how membership and degree of involvement are associated with primary (eg, QoL, connectedness, coping, knowledge of condition, knowledge and use of appropriate services, stigma; to be informed by WSs1-3) and secondary (eg, resilience, stigma, mental health) outcomes over the course of the project.

Quantitative data will be summarised using statistics appropriate to data characteristics, and precision of estimates will be expressed using 95% confidence intervals. Analysis of variance (ANOVA) and multiple linear regression techniques will be used as appropriate to specific questions. Qualitative data will be interrogated using thematic analysis[17,18] of data from the open-ended questions to provide a richer picture of experiences.

Findings from the quantitative and qualitative data collection will be grouped in relation to timeframe to create disease staging documents. These timeframes will be compared and contrasted with novel data-driven computational event-based models of these sequences of events.

Innovative analytical methods will also be developed and applied by involving the arts as a research method in dementias,[19] examining lived experience in a subset of approximately 10% of participants including research poetry[20] and developing visual responses (Visual Routes) to personal experience over time.[21,22] The research poem[23] builds on traditional forms of thematic analysis[18] and will be a powerful tool to convey affective, social, and experiential aspects of group membership and caring over time. Our plan is to involve a subset of members as co-researchers to interview a designated number of other group members; their interviews will be used to jointly create research poems based on verbatim accounts and identified TA themes. The poems will reflect experiential accounts of members, rather than a more literary use of the poem,[20] research poems will contribute to WS2 and WS4 and will be used in interactive public engagement activities.

2.4.2 | Work package 2—theoretical development

Following a literature review, researchers will subsample members and facilitators (and others, eg, commissioners, charity leads, referring professionals) of 10 different support group meetings in order to develop a theoretical understanding of the processes, contexts and people involved in these groups using situational analysis (SA).[24] SA has not, to our knowledge, previously been used in dementia care research. SA has its roots in grounded theory but goes beyond examining social processes in order to develop "situational maps," which centre on elucidating the key elements, materialities, discourses, structures, and conditions that characterize the situation of inquiry (an RDS group), rather than focusing only on individual participants through interviews.

Analysis of the data will inform and subsequently allow the research team to establish a broad Theory of Change (ToC) of multicomponent support groups for people with rare dementia and nested subtheories of change tailored to particular populations (eg, genetically at risk individuals) and formats (eg, online support). Broadly speaking, a ToC describes the causal assumptions specific to an intervention's sequence of events or steps leading to impact (eg, how did the support group contribute to positive changes in well-being). A ToC is also valuable for the ongoing management and evaluation of support groups and assessing their scalability.[25]

2.4.3 | Work package 3—measure development

Few outcome measures are designed specifically for people living with rarer forms of dementia. Our lack of understanding of the lived experience of people with these conditions also means that this in-depth study of their experiences may bring to light topics, issues, or concepts not previously considered in studies of people with dementia more generally. Despite global interest in resilience and health,[26] there are no resilience outcome measures for people living with dementia. Consequently, they cannot be considered as one of the set of "core outcomes" proposed for dementia research.[27-29] This is an important area for further investigation if we are to understand how the resilience of people living with dementia can be enhanced by health, psychological, and social care services or interventions. We describe below the framework for developing a resilience measure, involving a subsample of up to 490 participants. For any further new measures prompted by participant responses from WP1, a similar framework would be adopted.

We will develop and test a resilience outcome measure for people living with dementia, including those with a rare dementia (and proxy response measure) that is appropriate for evaluating the impact of health, psychological, and social care services and interventions.

Following a literature review to identify the components of resilience as described by people living with dementia, researchers will conduct qualitative interviews to explore what matters the most in terms of the challenges experienced, strategies and resources for dealing with challenges, and the endpoints that they would find most meaningful (including data from WP1). A subsequent two-round Delphi survey of stakeholder groups will be undertaken to gain consensus on the core components identified in previous steps of development. Measurement items will be developed from the agreed conceptual framework and appropriate response categories and question stems identified. Items will be pretested with a small group of people living with dementia, with subsequent revisions made in response to this process. The penultimate stage will involve field testing and a preliminary evaluation of psychometric properties to identify and eliminate items with poor psychometric performance, eg, through exploratory factor analysis. Finally, a psychometric evaluation of the novel measure will take place.

Thematic analysis[17,18] will explore the qualitative interviews to inform the development of the theoretical framework. We will then identify and eliminate items with poor psychometric performance by conducting an exploratory factor analysis and investigate the acceptability, reliability, validity, and responsiveness of the reduced item questionnaire. We will undertake convergent and discriminant validity analysis and ensure internal consistency before evaluating the final iteration of the developed measure.

2.4.4 | Work package 4—novel interventions

This work package will subsample participants, who will be involved in developing a blended manualised intervention, with content informed by a combination of (a) thematic analysis of WP1 data; (b) our ToC arising from WP2; (c) 2018 NICE guidelines on interventions for carers of people with dementia; (d) recent systematic reviews elucidating the active components of online carer interventions,[30] (e) evidence as to the support needs of carers of people with a rare dementia; and (f) consultation with experts on the delivery and design of online interventions and online support research. Thematic analysis of responses from WP1 online intervention questions and focus groups will be undertaken throughout the iterative intervention development process.

We will enhance the accessibility of our intervention to those with sensory impairment and disability by following web content accessibility guidance[31] and will build into the intervention an initial consultation with a professional to support those with low computer literacy.

Seventy-five participants will be invited to take part in a feasibility study. Following consultation of demographic and premeasure information (from WP1), participants will be randomized 2:1 to intervention or treatment as usual (TAU) by an independent clinical trials unit (North Wales Organisation for Randomised Trials in Health and Social Care (NWORTH), Bangor) using Russell et al's dynamic randomisation procedure.[32] Researchers collecting the outcome measures and analysing the data will be blind to allocation.

Primary quantitative outcomes will include the acceptability (percentage of participants completing intervention and percentage of sessions attended) and feasibility (recruitment of an adequate sample over the timeframe and retention rate in the study). Secondary outcome measures will include the outcome measures we would use in a full trial and will be informed by WS2 (theoretical development) and WS3 (measure selection and development). Constructs measured may include depression, carer burden, quality of carer/person with rare dementia (PWRD) relationship, quality of life, positive aspects of caring, and challenging behaviour of the PWRD and resilience. A primary measure of effect (eg, carer well-being; EQ-5D[33]) will be used for exploratory cost effectiveness analysis in WP5. Scales (eg, the RUD[34]) will be used to capture service use across different agencies including the NHS, local authority, and third sector.

A "stop-go" measure for proceeding to a full trial will relate to our success criterion: If 70% or more of recruited participants meet criterion, proceed to roll out of trial; if it is 60% to 70%, consider a modified design to increase adherence; if it is less than 60%, do not progress to a full trial using this method. Our second criterion for success is to recruit our target sample size within the planned study timeframe. We will report proportion of missing data on measures and use this as an index of measure acceptability. Preliminarily analysis of quantitative outcome measures will be undertaken using linear mixed models to establish feasibility and estimate likely effect sizes. No hypothesis testing will be undertaken, and all estimates will be presented with their associated 95% confidence intervals. Data analysis will be supported by NWORTH.

All participants will complete a postintervention questionnaire including open questions on barriers and facilitators. We will purposively sample 10 to 15 participants (we will include those who self-identify as "non- expert" computer users as well as those with English as a second language in order to examine acceptability of translation tools) to take part in qualitative interviews. In line with process evaluation guidance,[35] implementation and potential mechanisms of impact will be a focus as will intervention design and content.

2.4.5 | Work package 5—economic analysis

Using Markov modelling, we will use data from the previous WPs to model the cost effectiveness of referral to multicomponent support groups for people affected by rare dementias (PWRD and carers) over a lifetime period and the exploratory cost effectiveness of the online support intervention.

A systematic review and—where possible—meta-analysis will be undertaken to establish the effectiveness of multicomponent support groups. The systematic review protocol will be registered with

PROSPERO. This will complement effectiveness data generated by WP3 and WP4. Searches will be conducted in biomedical databases such as MEDLINE, Cochrane Controlled Trials Database, EMBASE, Clinicaltrials.gov, and the FDA and EMA websites to identify eligible studies.

We will identify a primary measure of effect (eg, DEMQOL [Dementia Quality of Life][36]) in WP3 to enable exploratory cost effectiveness analysis based on parameter data on service use and cost, utilities, and other outcomes from WS1 and WS4. Data for parameters of the economic model will be extracted using a standardised template and an assessment of bias made using the Cochrane risk of bias tool.

A cost-effectiveness analysis will be undertaken through a process of cost effectiveness literature review, effectiveness data from the study (WP3 and WP4), costing of multicomponent support group interventions and the online intervention, modelling of cost-effectiveness over the lifetime, assessment of uncertainty, and generation of an estimate of cost-effectiveness. This will be exploratory in the case of WP4 as the data are from a feasibility trial.

As the intervention develops, we plan to investigate using social return on investment (SROI), which is widely used in public health to evaluate services and interventions. SROI allows us to take account of a wide range of stakeholders and offers the opportunity to consider the outcomes for a much broader set of stakeholders than more traditional methods used in health economics. We will follow the Cabinet Office guide for SROI as recommended by the SROI Network and the New Economics Foundation (NEF).

Hard outcomes are reported widely using traditional methods of evaluation and are easier to report as they use numerical data to demonstrate differences. Soft outcomes are more difficult to report, as they often depend on subjective measures such as changes in confidence or behaviour. SROI offers the opportunity to report hard and soft outcomes in tandem, resulting in an evaluation that reveals the difference an intervention can make not just in figures but in terms of the difference the intervention has made to the person, community, and wider stakeholders.

The additional challenges of undertaking economic evaluation of dementia have been noted in the literature.[37] We will use experience from our previous work in the field of dementia economics, this literature, and data from other WSs to inform the economic modelling component of this study.

3 | DISCUSSION

3.1 | Ethical considerations

The study has been approved by the University College London Research Ethics Committee. Informed consent will be obtained and data collected in a variety of ways according to the participants' preferences, either via (a) face-to-face written/recorded responses, (b) virtually via videoconferencing and teleconferencing software, or (c) questionnaire/scale data and consent forms completed online. Appropriate written or oral translation of consent forms and research materials will be provided where the first language of the participants is not English. In accordance with the Mental Capacity Act (2005), all participants will be considered by the researcher as to whether they are able to understand the research information that is presented to them and retain this information in order to weigh up whether they would like to take part. Participants with a diagnosis of a dementia taking part in the RDS Impact study will have capacity to consent.

The interview data collected in person or via tele/videoconferencing will be recorded and automatically transcribed via the GoToMeeting platform, a GDPR-compliant online meeting, desktop sharing, and videoconferencing software package that enables participants to meet with researchers via the Internet in real time. Questionnaire and scale data will be collected online via a GDPR-compliant web-based survey tool, or in hard copy format. All data will be uploaded to the UCL Data Safe Haven, which has been certified to the ISO27001 information security standard. The repository uses a "walled garden" approach, where the data are stored, processed, and managed within the security of the system, avoiding the complexity of assured endpoint encryption. A file transfer mechanism enables information to be transferred into the walled garden simply and securely. Where possible, data will be analysed within the repository. Where specialist software is required and not supported by UCL Data Safe Haven, a pseudonymised version of the data will be downloaded to institutional servers and analysed locally, before being reuploaded to the Data Safe Haven. Hard copy data collected outside of UCL will be transported on a quarterly basis to the Dementia Research Centre, UCL, and stored securely.

The data we are collecting for the RDS Impact study centres on asking individuals about their lived experience of a dementia. The research team have considered the risks involved with asking questions from which emotional responses could arise and have set out robust distress and safeguarding protocols to manage the risks involved. Participant will additionally be made aware of their right to withdraw from the study at any time without their clinical, legal, and/or support needs being compromised.

3.2 | Patient and public involvement (PPI)

RDS members contributed to sketching out the motivations and theoretical background for this study. Co-applicant Roberta McKee-Jackson, who has been a member of the PCA Support Group since 2007 and Bereaved Carers Support Group since 2017, has emphasised support groups' provision not only of "support, advice, guidance, and encouragement" but notably also "respect," achieved through the "opportunities to share in a safe environment, develop new friendships and a support network of peers, and to access specialist consultants, nurses and clinical staff." McKee-Jackson will co-lead PPI during the research programme with Crutch, who previously led on PPI for the Queen Square Dementia Biomedical Research Unit.

We will continue to encourage RDS members who are PLWD and carers to be involved through (a) contributing and directing content of meetings, websites, newsletters; (b) co-writing advice sheets, eg, FTD Carer Stories, The Stages of PCA; (c) contributions to the RDS Advisory Committee and RDS Governance Subcommittees which currently include FTD and PCA members; and (d) co-designing the study alongside the research team to reach consensus about the measures used in the main interviews.

4 | CONCLUSION

The study aims to investigate the impact and reach of multicomponent support offered and delivered to people living with a rare form of dementia. The study will capture information through a combination of longitudinal interviews, questionnaires and scales, and novel creative data collection methods. The notion of "impact" in the context of support for rare dementias will involve theoretical development, novel measures and methods of support interventions, and health economic analyses.

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CONFLICT OF INTEREST

None declared.

DATA AVAILABILITY STATEMENT

Data sharing not applicable - no new data generated

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