Appendix B: Study protocol

Exploring the experiences of those participating in Patient and Public involvement in motor neurone disease.

Protocol

Chief Investigator:

Dr Esther Hobson, Academic Neurology Unit, Sheffield Institute for Translational Neuroscience, 385a Glossop Road, Sheffield S10 2HQ, UK Tel: 0114 222230, Fax: 0114 222290
Email: e.hobson @sheffield.ac.uk

Co-Investigators:

Ms Lucy Musson
Academic Neurology Unit, Sheffield Institute for Translational Neuroscience, 385a Glossop Road, Sheffield S10 2HQ, UK Tel: 0114 222230, Fax: 0114 222290

Sponsor details:

Sheffield Teaching Hospitals NHS
D49, D Floor
Royal Hallamshire Hospital
Glossop Road
Sheffield
S10 2JF

STH project reference number: STH19307

STH Directorate affiliation:

Neuroscience

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Lay summary

Background

In the last few years the involvement of members of the public in medical research has increased. Patient and Public Involvement (PPI) involves consulting members of the public on ways in which research is conducted. This might involve deciding which research should be prioritised, advising on how clinical studies are carried out or sharing the findings of the research with the public. Based in the Sheffield Institute for Translational Neurosciences, the Sheffield Motor Neurone Disease Research Advisory Group (SMND RAG) is the first and only group to specialise in motor neurone disease in the UK. This panel has been working with researchers for over five years. Members include patients, family members, carers and volunteers.

Methods

This research aims to explore the experiences of those involved the Sheffield MND research advisory group, the challenges they face and the things that may help or stop people taking part. A successful project may be able to recommend ways to make this and other PPI groups more successful and enable more people to take part.

This research will interview members of the Sheffield Motor Neurone Disease Research Advisory Group along with researchers, clinicians and scientists who have worked with the group. Interviews will be either face-to-face, telephone, Skype or email interviews depending on the interviewee preference and can be alone or together with e.g. a carer. The researchers will also attend the Research Advisory Group as impartial observers to gain an understanding the group processes.

Results

Interviews will be audio-recorded and transcribed before analysis by the Sheffield MND research team. Results will be discussed in future SMNDRAG meetings prior to publication. The results will be shared with other PPI groups in the UK using posters or at conferences.

Nature of research

This research study is to be conducted by the SITraN clinical research team including a University of Sheffield Clinical Neurology MSc student, under the supervision of the Sheffield MND clinical research team including chief investigator (Esther Hobson).

Abstract

Background
The importance of patient and public involvement in research is now recognised and is a requirement for most medical research. However, panels, such as the Sheffield Motor Neurone Disease Research Advisory Group (SMNDRAG) have only recently been established and it is only recently that members of the public have been involved in aspects of research such as development and oversight of research methods and analysis and dissertation of the results. The most effective way of involving the public to deliver a positive impact has not yet been established.

Methods
Semi-structured qualitative interviews will be conducted with members of the SMNDRAG and clinicians, researchers and scientists who have interacted with the participants. Interviews will be conducted until data saturation is reached, or a maximum of 20 interviews have been conducted. Thematic analysis will identify relevant themes.

Results
The results are expected to provide suggestions which may improve the participating in and impact of the SMNDRAG and will be relevant to other patient and public involvement activities as well as advising how to improve interactions between researchers and the public.

Background

Until recently it was uncommon for members of the public to be involved in the conduct of research. More recently Patient and Public Involvement (PPI) has become a necessary requirement in virtually all clinical research conducted in the NHS. PPI may include individuals or panels of people who have experience living with the disease. With their unique knowledge they can use their expertise to identify priorities for research which may be different from clinician/researcher priorities (1,2). They can also advise (and potentially improve) methods of communication, recruitment and retention of patients within studies. They may also be involved in the study oversight and methodology, facilitate analysis and dissemination, particularly in ways that appeal to a patient or member of the public rather than a researcher (3,4).

PPI may be conducted by panels and individuals may be involved in research management, funding decisions or undertake some of the research themselves. Some PPI panels are disease specific and others generic. The Sheffield Motor Neurone Disease Research Advisory Group (SMNDRAG) was established in 2009. It consists of patients, carers, parents or family members, volunteers or members of charities such as the Motor Neurone Disease Association. It is the only group supporting research in motor neurone disease (MND) and as a result now supports research throughout the UK. The group meet quarterly and some times communicating between meetings. Some participants are unable to attend meetings and communicate by email or Skype. Members of the group have been co-applicants in grants or members of research management groups and have supported co-design projects such as the development of a neck collar: the Sheffield Support Snood (5).

With PPI being a relatively new phenomenon there are many areas of uncertainty. There is some evidence for the developmental role of public involvement, such as enhancing awareness, understanding and competencies among lay participants. The evidence of it's impact on the research conducted remains scarce (6). Those participating in PPI may face physical, psychological or financial challenges. These may apply to all conditions but the SMNDRAG will face challenges unique to MND and there is requirement to support PPI members to enable them to participate effectively.

Aims and objectives

The aim of this study is to explore the experiences of those participating in, organising and working with the Sheffield MND Research and Advisory Group.

The objectives of the study are to:

- Conduct a literature review of patient and public involvement
- Conduct and analyse semi-structured qualitative interviews of participants, staff and researchers working with the Sheffield MND Research Advisory group. These interviews will explore:
    - The experiences of attending the group
    - The motivation for joining and participating in the group.
    - The barriers and enablers to participation, both physical and psychological
    - The role of the group as a whole to the individual participants
    - The experiences of clinicians and scientists who interact with the group
    - The perceived impact the group has on research in MND both within SITraN and beyond

The results of this study may recommend ways to improve participation and impact on research. The results will be relevant to other future MND panels as well as other PPI groups facing similar challenges.

Plan of investigation

This is a qualitative study conducting semi-structured interviews with SMNDRAG participants and research staff.

Prior to the interviews the following will occur in order to prepare interview topic guides and guide the subsequent interview analysis:

1. A literature review of the subject, examining the INVOLVE (7), academic and grey literature of PPI in other diseases
2. Observation of the PPI group in progress by LM, who will take reflective field notes and feed back to the CI
3. Brief feedback from the PPI group about the good and bad aspects of the group (already completed in preparation for the study).

Inclusion criteria:
1) Members of the Sheffield MND Research Advisory Group past or present
OR
2) Staff who have interacted with the group for research purposes (usually employees of either Sheffield Teaching Hospitals NHS Trust or the University of Sheffield).

Exclusion criteria:
Those who are unable to give informed consent or undergo an interview/questionnaire due to severe ill health, language or cognition difficulties.

Recruitment
Participants will be approached in person, by letter or email (according to their usual method of communication). They will be provided with a participant information leaflet and given at least 24 hours to consider the study. They will be invited to discuss the study further with the study team.

Consent
Written consent is required but in those patients who are unable to provide written consent, verbal consent or using a communication aid can be taken witnessed by a family member or friend.

Sample size
The SMNDRAG consists of a total of 20 members. When consulted on this study all those who had attended the group indicated they would be interested in participating. Ideally interviews would continue until data saturation is reached (as judged by the research team) but it is estimated that a representative sample of 10 interviews involving patients, family members, volunteers plus 3-4 researchers would provide a reasonable dataset. This is a student project with limitations on time and therefore a maximum of 20 interviews will be conducted. Patients will be able to be interviewed with a carer, as if often preferred in this population.

Withdrawal
Participation will not impact on care patients or families receive and confidentiality will be discussed as part of the consent process. Participants will be given two weeks following the interview to withdraw from the study to allow them to reflect on what they discussed. If they withdraw all data collected with be destroyed. After this data will be retained for analysis.

This is made clear in the participant information leaflet and consent form. Participants will also be offered to redact aspects of their transcript either after the interview or before dissemination of results.

Interviews

Semi-structured interviews will be conducted at the Royal Hallamshire Hospital, SITraN, an alternative private area or at the participants home (whichever is most convenient to the patient). The interviews will be conducted by Lucy Musson (LM), an MSc in Clinical Neurology and supervised by Dr Esther Hobson (EH).

LM will receive training on interviewing, listen to examples of qualitative interviews and the audio recording of each of the first two interviews and subsequent transcripts will be reviewed EH to ensure the interviews are being appropriately conducted, the data gained rich and any participant concerns addressed.

Interviews will be audio-recorded and transcribed. Interviews duration will depend on the participant, their disease and may range from very short (10-15 minutes if the patient is very frail but wishes to be involved) to approximately one hour if the participant wishes to expand on their answers. The Sheffield MND team has experience in conducting qualitative interviews in MND and is able to adapt interviews with MND in mind. We will also offer to conduct telephone, Skype or email interviews and allow participants to expand on their answers by providing written answers after the interview.

The interviews will be conducted using a topic guide based on a literature review and input from the SMNDRAG and results from early interviews will inform later interviews.

Fields notes will also be taken by the interviewers and Lucy Musson will also attend the SMNDRAG as an impartial observer.

Results

Data analysis

Results will be analysed using thematic analysis (8). This will involve the following steps

1. Familiarisation with the data using line-by-line coding (EH and LM)
2. Independent coding for initial themes (EH and LM separately)
3. Searching for themes (EH and LM together)
4. Reviewing themes (EH and LM together +/- separately if further coding required)
5. Triangulation of data from the staff and PPI member interviews (EH and LM) (9)
6. Defining themes (LM and EH)
7. Writing up (LM +/- EH)

Results will be discussed in research meetings with the wider MND team and the SMNDRAG and participant and staff interviews triangulated to explore complex themes such as research impact and participant-staff relationships.

Dissemination and potential impact

The results will be presented to the SMNDRAG to enable the group to input into the analysis and to disseminate the research to other patients/families. An academic paper will be submitted for publication and a "blog" article included on the SITraN website.

The results will be provided to the SMNDRAG who may wish to implement any suggested changes to the group. The experiences and potential changes may be relevant to other PPI groups, both with SITraN and other groups.

Limitations and ethical considerations

This study is examining a limited group of participants but at present, this is the only MND research advisory group to examine. However the results will be compared to other studies of PPI groups e.g. stroke (10). The interviewers have no influence on any of the potential participants and whilst Dr Esther Hobson has been involved with the SMND RAG in the past, Lucy Musson has not.

The interviews may cover sensitive or potentially upsetting topics. The Sheffield MND clinical team have extensive experience in qualitative research in patients, carers and members of staff. It has developed successful methods of ensuring participation is possible for patients with significant disabilities and can support participants who may be discussing potentially distressing topics. The research team and the SMNDRAG have experience in supporting participants in these circumstances. The "ground rules" for qualitative interviews will be explained prior to commencing interviews and interviews terminated should participants become distressed, tired or at their request.

Although quotes used in dissemination will be anonymised given the limited number of participants it may be possible for participants' responses and quotes to be identifiable. Full transcripts will only be available to the interviewer and Dr Esther Hobson who will use their discretion when reporting the results if the responses are identifiable or upsetting and will exclude responses if any risk of harm to the participant is possible. They will also take into account any potential impact the responses may have on participants' research and employment if they are staff participants. If required they will allow the participant to decide whether to include a particular response. This will be explained in the participant information sheet and consent form.

Ethical and sponsor approval is required for this study.

The study aims, protocol, patient information sheet and consent form have been reviewed by the SMNDRAG and clinical research team to ensure the study is acceptable to potential participants. The group members indicated that it was a worthwhile study.

Data entry, security and confidentiality
Data (including audio-recordings) will be collected and retained in accordance with the Data Protection Act 1998 and Caldicott Principles. Data will be anonymised prior to entry onto a database and stored on the secure Sheffield University intranet which is password protected. Study documents will be retained in a secure location during and after the study has finished according to SITraN SOPs. Personal data (consent forms, recruitment log) will be held for up to 12 months and research data up to 10 years.

Adverse events
No adverse events are expected in this study. As such all adverse events (including serious adverse events) will be recorded and reported to the CI and MND research team and sponsor.

Access to source data
Monitoring and audit by the relevant health authorities will be permitted by the sponsor. These include the Research Ethics Committee and local R&D departments. The sponsor will be allowed to monitor and audit the study at each site and be allowed access to source data and documents for these purposes. Intellectual property generated by University of Sheffield researchers is managed by the University of Sheffield Research Office.

Project management
Weekly oversight through the Sheffield MND clinical research team.

Costs
Publication costs will be met by student fees. The student will transcribe initial interviews but if required, student fees can cover a small number of transcriptions.

Service users
The SMND RAG have reviewed the study principles, participant information and given guidance on topic guides.

Target dates:
The project will commence in April 2016 following ethics and sponsor approval.
Initial report to the research team will be submitted in August 2016 following which the results will be prepared for academic publication.

Staff underpinning the study: The study will be conducted by Ms Lucy Musson, MSc student with day-to-day supervision from Dr Hobson. She has received Good Clinical Practice training and holds an honorary contract with Sheffield Teaching Hospitals NHS Trust.

Other investigators expertise: Dr Esther Hobson is an NIHR Doctoral Fellow and specialty registrar in neurology. She also has experience caring for patients with MND and other chronic diseases and conducting qualitative research in MND. She holds an honorary

contract with Sheffield Teaching Hospitals NHS Trust and works in the Sheffield MND care and research centre.

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