NatCen
National Centre for Social Research

Long-term Neurological Conditions Pilot Study
Final Report

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Contents

1 INTRODUCTION

In March 2005, the Department of Health published a National Service Framework (NSF) for Long-term Conditions, with a focus on long-term neurological conditions. The NSF set out 11 quality requirements (QRs) for standards in services for neurological conditions, based on evidence from people providing and using these services, with a commitment to implement these QRs fully by 2015.

The following year, the Healthcare Commission asked the National Centre for Social Research to explore the feasibility of carrying out a survey of people with long-term neurological conditions, with a focus on the areas highlighted in the NSF and its QRs.

The initial phase of research involved a consultation with people working in the field, people with long-term neurological conditions and carers. The objectives of this consultation were to establish whether it would be possible to conduct a survey of people with long-term neurological conditions, to explore priority topics to cover in such a survey, and evaluate potential methods for sampling and carrying out the survey.

The report from the initial phase of the research was produced in August 2007[1]. It showed that there was a lot of support and enthusiasm for a survey, among both professionals and service users. This report focuses on the second phase of the research.

The second phase of research was to develop a questionnaire for a survey of people with long-term neurological conditions, and conduct a pilot survey to assess whether the questionnaire and the sampling methods used would be feasible for a larger scale survey (either a national study or one carried out by individual Trusts).

The pilot work involved developing a questionnaire (designed as a self completion instrument) and deciding the best option for sampling. The plan for the pilot research was submitted to two National Research Ethics Service Ethics Committees for ethical approval, but failed to get approval.

This report outlines the work that was carried out as part of the second phase of the research; the reasons why the ethics committees gave unfavourable opinions; and possible options for taking the research forward.

[1] *Long Term Neurological Conditions Feasibility Study: Consultation Report* NatCen June 2007

2 QUESTIONNAIRE DEVELOPMENT

2.1 Planned methodology

Conducting a survey of people with long-term neurological conditions presents a lot of challenges. As noted in the initial consultation report, some people with neurological conditions may have severe communication difficulties, which makes it extremely difficult for them to participate in a survey via any method.

The consultation study suggested that a face-to-face approach was the favoured methodology because:

- it allows "richer" information (more detailed questions, complex routing);
- people who have trouble writing can take part;
- it could feel more "empowering" to those taking part.

However, this approach was not without its drawbacks:

- It would not be feasible for people with verbal communication difficulties to take part on their own – assistance may be required from a third party.
- People with long-term neurological conditions may not be able to keep appointments if they are having a "bad day", or they may not want to be visited by a "stranger".
- The home situation of some people may make it difficult for them to have a visitor.

There are also pragmatic difficulties with a face-to-face approach. The cost of a face-to-face survey would be large, as a sample of people with long-term neurological conditions would be relatively unclustered (given the small numbers with the conditions of interest). This would mean that the sample would be widely spread across the country, meaning that any interviewer would only have a few respondents that it would be possible for them to visit without involving very long journeys. Thus, a large number of interviewers would need to be involved, each carrying out only a very small number of interviews which would impact on the cost of the survey, making it expensive and very labour intensive.

This would limit the sample size of any national survey, and would make it less straightforward for PCTs and Acute Trusts to conduct their own surveys.

Following further discussions with the Healthcare Commission, it was decided that it would be sensible to pilot a self-completion approach to see if this was a feasible option for a large-scale survey.

Postal surveys, involving mailing out a self-completion questionnaire, have both advantages and disadvantages compared with face to face surveys. Advantages are:

- They are far cheaper to administer than face to face surveys, which allows a larger sample size for a given budget.
- They ensure that people living in all areas of the country are accessible (whereas it may not be cost effective to visit people in very remote areas).

- People can answer the questionnaire when they feel like it and can find the time and space.
- People can also take more time to think about difficult or complex questions which can lead to more thoughtful answers.
- It allows anonymity – there is no social response bias due to giving answers to an interviewer.

However, postal surveys also have disadvantages:

- They typically elicit lower response rates than face-to-face surveys, even if reminders are used. It is also not possible to tell if a low response is due to poor address information or lack of interest in responding.
- The questionnaire needs to be short and simple to complete, so there can not be complex filtering (whereby some questions are only asked of those with specific experiences).
- People with lower literacy levels may not be able to complete the questionnaire, which could cause bias in response.
- Some people with long term neurological conditions may not be physically able to complete a self-completion questionnaire.

The consultation stage suggested that a self-completion option would be suitable, although, as mentioned above, some people may have trouble completing a written form on their own. For this reason, it was decided that a web version and an electronic "Word" version of the questionnaire would also be provided. This would enable people with special computer equipment that helped them communicate to complete the form with minimal help. Having a number of different ways to respond to the survey was something that service users involved in the consultation stage had seen as important.

Piloting a self completion method (with electronic options) would enable us to assess whether it was successful by:

- looking at the response rate;
- comparing those who responded with any information we had on those who did not (ideally looking at response rates by condition);
- seeing how many people completed the form on their own and how many needed help (the questionnaire includes a question on who completed the form).

It was therefore agreed that a questionnaire suitable for self-completion would be developed for the pilot.

2.2 Questionnaire development

A questionnaire was developed in close consultation with the Healthcare Commission, and with colleagues from the Commission for Social Care Inspection (CSCI). Information from the consultation stage on the topics that should be covered in a survey; from the National Service Framework and from other existing surveys was used to help guide the development.

This questionnaire went through a number of iterations, with input from the Healthcare Commission and CSCI, until a version existed that was ready to be cognitively tested

with a small number of people with long-term neurological conditions. The questionnaire used for testing is attached at Appendix A.

Respondents for cognitive testing were recruited via a number of member organisations, and the interviews took place in May and June 2008.

In total, 14 interviews took place, with people who had a range of long-term neurological conditions, including:

- Encephalitis (2)
- Cerebral Palsy (2)
- Ataxia (1)
- Transverse myelitis (1)
- Parkinson's disease (3)
- Epilepsy (1)
- Motor Neurone Disease (1)
- Early stroke (2)
- Epilepsy and Charcot-Marie-Tooth disease (1)

In total, 5 women and 9 men were interviewed.

The cognitive interviewing was very useful in highlighting which parts of the questionnaire were and were not working. It also highlighted the difficulty of designing one simple questionnaire to cover such a wide range of experiences. Different neurological conditions have very different starting points, pathways and impacts which can vary from disease to disease and from person to person. Designing a questionnaire that can collect useful information and which makes sense to each individual respondent is a challenge.

However, the questionnaire tested proved a good starting point, and we identified questions that worked, and those that did not. The commentary on the changes we made to the questionnaire following the cognitive testing is attached at Appendix B.

The questionnaire, post-cognitive testing, is attached at Appendix C. This questionnaire is nearly finalised, but would benefit from a further small round of cognitive testing as some of the changes made have not been tested out. This especially applies to the sections on diagnosis, rehabilitation and equipment.

The questionnaire is longer than anticipated, and currently is 15 sides of A4. Ideally, it would be no longer than 12 sides. It is possible that a different layout could reduce the number of sides, but consideration should also be given to removing some of the questions.

Copies of the letter drafted to be sent with the questionnaire and the reminder letter for those who did not respond within a specified time period are also attached in Appendix D.

3 SAMPLING

3.1 The sample frame

Findings from the consultation stage suggested that the best way to sample people with long-term neurological conditions could be via GP registers. However, the initial consultation did not involve GPs or any detailed discussion of this sampling approach.

Therefore, following the consultation stage, the Healthcare Commission set up a meeting with the British Medical Association (BMA), involving the Chair of the General Practitioners Committee (GPC) which is part of the BMA. At this meeting, the issue of whether people with long-term neurological conditions could be identified and sampled via GP records was discussed. The BMA felt that this approach would not be feasible, as GPs may not record all such conditions, and may have coded such conditions in variable ways. A letter from the BMA which outlines their views on the sampling for this study is attached at Appendix E.

The BMA recommended the use of secondary and regional neurological centres:

"We believe that the best way to reach the widest possible group of patients who have neurological conditions would be to use data held by secondary or regional neurological centres. The vast majority of patients with neurological conditions will be known to them, and are likely to be reviewed at least on an annual basis. We believe that using this route it would also be possible to avoid the major confidentiality issues that GPs and practices would have faced. If this method is adopted we believe the survey would have greater validity and might be more successful."

The consultation stage had looked at the use of hospital records, and felt that using this method for sampling would be good for some neurological conditions, but less good for others.

"Many experts felt that hospital records would provide very patchy coverage of people with neurological conditions. Many neurological conditions are chronic, and so there could be limited contact with a hospital. For some neurological conditions, coverage via hospital records could be good – for example, people with MND have specialist clinics and so nearly all people with MND would be found via hospital records. However, other conditions (e.g. MS or epilepsy) may not generally be linked into hospital based services. For some conditions (e.g. Acquired Brain Injury), people will have been in hospital initially – but once discharged, may then never go back into hospital, so it might be difficult to access those who had been living with their condition for some years."

As using hospital records was not ideal, other sampling options were also considered.

Some preliminary work was done looking at the coverage of membership of some voluntary organisations associated with certain neurological conditions. Many existing surveys of people with neurological conditions have used member lists from these organisations as a sampling frame. However, we were not convinced that coverage would be good enough for a national survey that was attempting to cover the whole range of neurological conditions. The table below shows the outcome of some very early work on the numbers of people in contact with a few of the main member organisations, compared with estimates of how many people in the country might have that condition. As this table suggests, there is a lot of variation by condition and in many cases coverage is low. This low coverage, and the self-selected nature of the sample, means that using these membership lists for sampling could lead to extremely unrepresentative samples.

ORGANISATION	NO. OF PEOPLE WITH CONDITON IN CONTACT WITH ORGANISATION	TOTAL WITH CONDITION (Estimate from Neuro Numbers[2])	% covered (based on Neuro numbers)	TOTAL WITH CONDITION (organisation estimate, if different to Neuro numbers)	% covered (based on org estimate)
MND Association	2,877	4,000	72%	5,000	58%
Parkinson's Disease Society	20,000	120,000	17%	Same as NN	17%
Ataxia UK	1,500	Not estimated	n/a	10,000	15%
Scope (cerebral palsy)	3,000	110,000	3%	114,000	3%
Epilepsy Action	11,200	300,000	4%	456,000	2%
Encephalitis Society	820	Not estimated	…	[Min 4000]	[Max 20%]

Using Disability Living Allowance recipients as a potential sampling frame was also considered. Disability Living Allowance (DLA) is a tax-free benefit for children and adults who need help with personal care or have walking difficulties because they are physically or mentally disabled. Given the criteria for applying for DLA, using these records for sampling would exclude people whose day-to-day lives are less severely affected by their conditions.

A practical problem with using DLA records is that to sample from the DLA records would involve relying on the "main disabling condition". Information provided on the application form for DLA is coded into around 100 groups (the main disabling condition) and only one condition per person is coded. The coding of main disabling condition is not seen as being particularly accurate or detailed, and is not backed up by medical records. For these reasons, it was decided not to pursue this option.

Given the lack of possible alternatives, the research team had discussions with two regional neurological centres to assess how practical and useful the sampling approach via such hospitals would be. Representatives at the two hospitals spoken to were optimistic that this approach to sampling would be useful, and that, on a practical basis, such samples could be drawn.

It was agreed that it would be important to sample across a wide time range, so that we would pick up people who had an initial diagnosis some time ago but were no longer in contact with such health services. It was also important to include both outpatient records and inpatient records, as for a number of conditions, patients would not need to stay in the hospital.

Although sampling via hospital records is not perfect, it seemed to offer better coverage than other alternatives. Sampling in this way for the pilot would also allow us to see the range of neurological conditions sampled and whether any appeared to be under-represented (although there is very limited data on the prevalence of various neurological conditions in the population, so it would not be possible to do a scientific analysis of coverage by condition). The pilot would also allow us to assess the practicality of this sampling approach.

[2] *Neuro Numbers: a brief review of the numbers of people in the UK with a neurological condition.* Neurological Alliance April 2003. ISBN 1 901893 32 4

Practically, sampling from these records seemed feasible, as the records were set up electronically in such a way as to make this type of sampling fairly straightforward. Both of the hospitals spoken to could envisage drawing a sample and sending opt out letters to the selected participants, prior to passing contact details for those not opting out to NatCen. (This would, of course require approval from NRES, the local R&D committee and potentially the Patient Information Advisory Group (PIAG)).

It was therefore agreed that the best approach would be to pilot the survey using sample from three hospitals that specialise in neurological conditions.

3.2 The sample protocol

Having agreed on a possible sampling frame, a sampling protocol was drawn up. We hoped to involve three specialist neurological hospitals in the pilot (but had not finalised their agreement to take part).

The sample would be based on patients with an appropriate ICD 10 code (G00 – G99: diseases of the nervous system, but excluding G43 and G44 - migraine and other headache symptoms). Ideally, we would want to include both in-patients and out-patients, and we would like to include records going back for five years. This would allow the sample to include people who had been living with their condition for some time, but only had contact with the hospital at the initial diagnosis. Going back further than five years would be ideal, but there would be logistical problems in that a) some hospitals' records were not computerised more than five years ago and b) address information from more than five years ago would be less reliable as people are more likely to have moved as time goes on.

We planned to only include patients aged 16-65 at the time of their contact with the hospital (as this study was designed to focus on working–age people). Prior to drawing a sample, we would look at the distribution of cases by ICD code to assess whether any disproportionate sampling should be done to allow for a broad range of conditions to be included.

The exclusion of ICD codes G43 and G44 – migraine and other headache symptoms – is based on the findings of the consultation phase. These are the most prevalent neurological conditions and so could potentially make up most of our sample. They cover a huge range of symptoms and levels of severity. In the view of experts at the consultation phase, the majority of headache and migraine sufferers would not be accessing or need to access some of the key services that we are interested in (such as social care, benefits and ongoing neurological care).

At each of three sites, we planned to sample 500 service users. The list of sampled patients would be submitted to the National Strategic Tracing Service to check for deceased service users. A self-completion questionnaire would be sent to all remaining individuals. The questionnaire would be sent by the survey organiser at the Trust. This survey organiser would be a member of the Trust's workforce, or, if a service contract were put in place, NatCen could undertake this work on behalf of the Trust. (A service contract is a binding contract between organisations that makes the employer liable for any breaches of data protection. Legal advice has been taken as to their use and these contracts are seem as preferable to alternatives such as honorary contracts).

This self-completion questionnaire would be sent out with a reply paid envelope, so that it could be filled in and returned. The covering letter and front page of the questionnaire would provide details of a web address where the questionnaire could be filled in on line, and also an e-mail address that could be used to request a Word version of the questionnaire.

A copy of the protocol (as submitted to the NRES Committees) is attached at Appendix F.

4 ETHICAL REVIEW

As we were planning to sample from NHS records, we needed to get ethical approval from NRES (National Research Ethics Service).

We submitted the plans for the pilot study for ethical review by the North West Research Ethics Committee on 20th May 2008.

The study was not approved.

The reasons given by the REC were as follows:

"The REC would expect to see efforts to obtain a survey sample that could be considered to be representative of the groups highlighted in the National Service Framework for Long Term neurological conditions. In its current proposed format the survey sample was not representative of those groups and it was felt that the survey would not include those people that were best placed to comment upon services for patients with neurological conditions.

1 Following on from the above point (A1), it was agreed that further effort should be made, including an increase in the research budget to permit a payment to be provided to GPs in order to secure their assistance in accessing a sample that was a representative group of people with neurological conditions.

2 It was agreed that the current questionnaire was based on a sample of patients without cognitive and communicative impairments and that it would require a complete re-write in order to be suitable for such a group of patients without the need for assistance from their carers.

3 The committee felt that further consideration should be given to conducting face to face interviews rather than using a self completion questionnaire."

Further issues raised by the REC were as follows:

"B The REC agreed that the proposal to exclude individuals who could not adequately understand verbal explanations or written information given in English would lead to the survey potentially missing important issues that ought to be addressed at the pilot stage of the research.

C The committee requested clarification regarding the time points at which the first and second reminder letters would be sent out to non-responders to the invitation to take part in the survey. In addition, copies of the reminder letter should be submitted to the REC for formal review.

D In relation to the questionnaire, the following points were raised

1 Question 15 - there appeared to be a typographical error as it stated "1 months" and it was assumed that this should read "12 months"

2 Question 29 - it was agreed that this question potentially raised expectations on the part of participants, i.e. if they answered 'no' then they might expect action to be taken on the issue.

3 The final question was numbered "50" but should be "53"

E Further to the discussion at the meeting, and as stated above, the initial invitation letter must provide clarity/definition of what was meant by the term 'neurological condition' in relation to the proposed study. It would be helpful to provide examples of the type of conditions included in the definition.

F It was noted that the application stated that the initial invitation letter to potential participants would include a free phone telephone number for patients to phone for further information about the survey and/or opt out of participation. However, whilst the letter did include a free phone telephone number it did not state that this could be used to opt out of participation. Nor did the section headed "Do I have to take part?" refer to the free phone number as a method of opting out."

Following discussion with the Healthcare Commission, it was decided to resubmit the application to another REC.

Although we agreed with some of the points made, we felt that the basic protocol (postal self-completion survey, sampled via hospital records) was still the best option available to us. Therefore, we made changes that dealt with some of the more minor issues, but did not change the basic methodology or sampling strategy. Instead, we spent more time in the second REC application form trying to justify our decisions, and enclosed documents such as the letter from the BMA which had caused us not to pursue the GP option further.

The study was resubmitted to Brighton East Research Ethics Committee on 16th October 2008. Again, the study was rejected. The reasons given were as follows:

"1 Members were concerned that the results of the study based on the methodology used may not be a representative sample of this group of service users. They strongly felt that the current methodology would not answer the ultimate research question that will enable you to assess whether the objectives set in the National Service Framework for long term conditions were being met. Hence members did not feel that it would be ethical to approve the study as currently designed.

2 It was felt that many of these service users especially from specialist centres would be dependant on their carers, who may possibly complete the postal questionnaire on behalf of the service user. It was pointed out that the views of the carers would be different from that of the service user. In addition, there would be no way of knowing whether the questionnaires returned were the views of the service user or their carers, which will once again would introduce bias and defeat the purpose of the study.

3 As you are recruiting this group of individuals through specialist neurological centres, members were concerned that the results of the study would be biased as the questionnaire after the pilot will be designed, based on the views of individuals from specialist centres only. They felt that the views of service users from specialist centres may be different from the views of those from non-specialist centres. Hence, members felt that it was important you include trusts that do not have specialist neurological centres as well, so that the views of a wider range of service users can be obtained.

4 Members agreed that the questionnaire was not intrusive but were concerned that the questionnaire on the whole was too long and not well phrased especially for those service users with more severe neurological problems. They were particularly concerned that individuals may start off completing the questionnaire but may not be able to complete it and this in turn had the potential to cause distress for this already vulnerable

group of participants (especially as there was no fall back mechanism in place to support these individuals should this happen). They strongly felt that as currently designed the possibility of distress for this vulnerable group outweighed the benefits of the study.

5 They were not sure that a questionnaire-based study was the best way to obtain the views of this group of individuals. They instead proposed a large scale, short, simple questionnaire in combination with in-depth qualitative research methods to obtain the views of this group of service users. Members also suggested that it may be a good idea to check the GP research database which would give you a guide to the number of people living in a particular area with neurological problems."

5 OPTIONS FOR TAKING THE RESEARCH FORWARD

The paper below was drawn up by NatCen and the Healthcare Commission in January 2009 and outlines the options for taking this research forward.

Options for the survey of people with long-term neurological conditions

Where we are currently

We have designed a self-completion questionnaire (around 15 sides of A4), which has had some small-scale cognitive testing. The questionnaire is nearly finalised (ideally we need to carry out a few more cognitive interviews in order to test changes that we made following the first round of cognitive testing).

In terms of sampling, having discussed options with the BMA and with some hospital trusts, we agreed that the best approach would be to draw a sample of people who had attended a specialist hospital about a neurological condition either as an inpatient or an outpatient, in the last 5 years.

The next stage planned was to pilot the questionnaire and the proposed sampling methodology to assess how well they worked. For piloting, we hoped to use three hospitals – two regional neurological specialist trusts and one acute trust with a specialist neurological unit - to assess how practical it was to draw the sort of sample we needed, and to assess, from the returns received, how effective this sampling appeared to be and how well the questionnaire worked.

Our proposed approach has been rejected by two Research Ethics Committees, who do not feel that the sampling method will provide a fully representative sample and that the self-completion methodology will either not be accessible enough for people with severe disabilities caused by their condition or could cause distress due to the limited support available using this methodology.

What can we do next?

This paper sets out a range of options about how we might proceed with the project from this point. As the purpose of this paper is to support discussion, no recommendations are made here; rather, we are keen to fully consider all of the available options.

1. Give up

As this is a feasibility study, we could decide on the basis of the feedback from the ethics committees, that, as it is not possible to conduct perfect research among people with long term neurological conditions, it is best not to conduct research at all. This would mean abandoning the project altogether.

2. Wait for further evidence

The NHS Service Delivery and Organisation Research and Development Programme (SDO) are currently funding a project, being led by Ray Fitzpatrick at the University of Oxford, which is looking at people with three specific progressive neurological conditions (Parkinson's disease, multiple sclerosis and motor neurone disease). The evidence from this will provide a benchmark against which to assess progress in the implementation of the NSF for long term conditions. However, a significant limitation of this survey is that it will not use a probability sample but rather will recruit participants from support groups for

people with the relevant conditions – this means that the sample is likely to exhibit bias and cannot be considered representative of all people with long-term neurological conditions or, indeed, of those people with the three specific conditions being investigated.

This research is due to report in early 2009. It may be that the findings from this provide insight into the most useful areas for research, and help us to redevelop this project if necessary. As such, it might be worth waiting for that report and discussing whether further research is necessary, and then making a decision whether – and how – to proceed.

3. The tool kit approach

As we have developed a questionnaire, and can provide ideas for sampling, we could publish a research toolkit for Trusts who want to conduct this sort of research themselves. This would allow the research to be taken forward by interested Trusts in a reasonably standardised way (but would mean that there would only be research at a local level, and there would not be national data).

A secondary benefit of this would be to support a programme of work following on from the Department of Health's recent review of customer experience information. However, we would need to be open about the fact that the toolkit methodology had been refused ethical approval, which might make it less attractive to Trusts. Furthermore, the main priority for the work arising from the DH review is to develop surveys that have already been undertaken nationally so that trusts can repeat them locally for quality improvement purposes. Since this survey would not have had a national roll out, its usefulness in this context might be limited; and it is debatable whether the role of the regulator is to provide such resources when they are practically unconnected with the existing national programme.

4. Continue as planned, but without ethical approval

It is unclear whether surveys such as this one require NRES approval, as they can be seen as 'service evaluations': such work does not strictly require NRES approval, although applications can be made on a voluntary basis. Given the need to access patient records, however, it would probably be necessary to get section 251 approval via PIAG[3] if the survey was not NRES approved. It is, however, open to debate whether this work constitutes 'service evaluation' or 'research', since the primary objective is not to support performance assessment, as in other surveys, but to gather new knowledge.

The Healthcare Commission have been in discussion with PIAG informally, and are currently considering the option of making an umbrella application to cover the use of patient information in all of their surveys. It is not yet clear if this is necessary, but if such an application were to be successfully made it would strongly support the use of patient identifiable information for survey purposes without prior consent of patients. Since it could reasonably be argued that the greatest ethical issue in the surveys is this use of patient information, it could also be argued that NRES approval would be unnecessary for the surveys if section 251 approval was in place, as there would already exist a clear high-level agreement that the use of data was appropriate and necessary.

Therefore, it would theoretically be possible to apply for PIAG approval, and if this is achieved, to proceed as originally planned.

[3] Section 251 of the NHS Act (2006) (previously enacted under section 60 of the Health and Social Care Act (2001)) permits the common law duty of confidentiality to be set aside in specific circumstances for medical purposes. PIAG (the Patient Information Advisory Committee) is the body set up to advise the Secretary of State on this issue.

However, at a local level, it is unclear whether Trust R&D committees would allow us to sample from their databases when the methodology has been rejected by two NRES RECs.

5. Appeal against the ethics decision

We could appeal against the latest decision of the NRES committee. This would mean the most recent application being sent on to a third committee, who would review it alongside the rejection letter from the MREC who most recently rejected it. However, having been rejected twice, it is unlikely that a third committee would be willing to overturn the decision.

6. Submit a new but substantially unchanged application to a different REC.

We could take the view that the rejection of the last application stemmed not from serious ethical concerns with the survey design but rather from a failure of the committee to adequately understand the methodology. If so, we could make a new application with few or no practical changes to the methodology but with more comprehensive explanatory notes provided to explain the methods and the reasons for them. However, since the project has already been rejected twice it seems unlikely that a third committee would be willing to make a different decision.

7. Pilot the survey using a different sampling method

There are sampling methods we could use which would not require NRES or PIAG approval. If we were to sample via patient groups (e.g. the MS society), we would not need these, and could probably proceed in the near future. We would still need some ethics approval, but the internal NatCen ethics committee could scrutinise the application.

This would allow us to draw a sample and pilot the questionnaire. However, sampling via patient groups and organisations is not representative, and we would not advocate its use for a larger-scale or national survey. The problem is that patient groups are self-selecting – people have to have chosen to join the group, and so we would not be able to get the views of people who were not part of a patient organisation.

While this approach would allow us to see how well the questionnaire works in the field, it would not allow us to test a sampling method that we could use in any large-scale survey. If we wanted to proceed to undertake a large-scale survey, we would then face the problem of embarking on this with an untried and untested sampling methodology, which could cause problems (both in terms of practicalities and in terms of the sample generated).

8. Change the methodology in line with the NRES comments and apply again

The comments provided by both Committees are not clear as to how this project should proceed. While the sampling is seen as being unrepresentative, the only alternative options suggested are via GPs (first committee), or (possibly) via a greater range of hospital trusts (second committee). The BMA's GPC assessed the GP route and felt that it would be even less representative and more problematic than the hospital route, which was why we decided to seek ethical approval a second time for our original suggestion. The second committee did not suggest any alternatives, but made a comment about also using non-specialist centres (which would be feasible). However, it was not clear if this change would be sufficient to ensure that approval would be granted, as the implication was that the sampling approach as a whole was unrepresentative.

In terms of method, the first committee felt that a face to face approach would be preferable, while the second suggested a very short postal questionnaire alongside in-depth qualitative interviews. Both committees felt that it was problematic that in many cases we would have to rely on carers to provide the information we needed if the person with the condition was not be able to take part in the research due to cognitive or communication impairments.

Much research relies upon self completion questionnaires, and in a number of surveys these will, in some cases, need to be completed by carers. Saying that this approach is not acceptable is inconsistent in the face of so many projects where this methodology has been approved.

To attempt in-depth qualitative research among people with long term neurological conditions would be even less representative and even more problematic (in terms of inclusion of those with cognitive and communication difficulties), and so does not seem worth pursuing. To move to a fully face to face approach would be prohibitively expensive (as the sample will be fairly unclustered[4]), and would not solve the problem of how to interview people with severe cognitive or communication impairments.

Therefore, trying to change the methodology to fit in with NRES suggestions is likely to lead to an even less representative survey.

9. Adapt our approach, and start again

There may be more minor changes we can make to our methodology that address some of the issues raised by the Ethics Committees without fundamentally changing our approach. For example, we could consider including some face to face interviews alongside the self completion methods, in order to assess how the different methods work in practical terms; and/or we could include a wider range of Trusts in the pilot sampling.

If we take a step back, consider where we are, what other research is available and what feedback we have had, we could then start afresh with our pilot. We could discuss with Trusts whether having PIAG approval is sufficient for them to allow us to sample from their patient records, or whether NRES approval is also required.

If NRES approval is still required, we would need to re-apply, but we could identify changes that we had made to the design that addressed some (if not all) of the previous issues raised. If NRES approval is not needed, we could seek ethical approval from an alternative (for instance, the National Centre's own Research Ethics Committee) and proceed on that basis.

PIAG approval could be sought separately, especially if we felt that we did not need NRES approval to move forward.

[4] For instance, we would expect wide geographical distribution; people are unlikely to live in the same areas simply because they have a neurological condition.

APPENDIX A QUESTIONNAIRE USED FOR COGNITIVE TESTING

APPENDIX B COMMENTARY ON COGNITIVE TESTING

COMMENTARY ON COGNITIVE TESTING OF QUESTIONNAIRE V1 FOR P2736: SURVEY OF PEOPLE WITH LONG-TERM NEUROLOGICAL CONDITIONS

FRONT PAGE

Some respondents gave inaccurate answers because of problems reasoning or remembering. Hence on the front page, the section about getting help completing the questionnaire has been slightly re-worded and made more prominent to encourage respondents to do this where it is needed.

DIAGNOSIS SECTION

Diagnosis was an extremely varied phenomenon. Some had their conditions from birth, while others developed them later on either suddenly or gradually. If there was a medical crisis respondents may have been unconscious or highly confused initially. The process of diagnosis can be fairly immediate or can frequently be very drawn out and a very iterative process because of the uncertainty around how people's conditions will progress/ the specific condition or variant they have/ the fact that for many conditions no two individuals will develop the same symptoms, the varying period of times over which conditions can be accurately diagnosed etc.

People found the information questions difficult to answer because they did not know what time period/ stage in their diagnosis was being referred to and also because the diagnosis/ provision of information usually involved more than one professional, one of whom may have been bad and another good. They also significantly often pro-actively gained information from specialist organisations and felt torn as to whether to answer with healthcare professionals or the specialist organisation in mind. They were additionally unclear about exactly what considerate meant in Q2. Respondents also struggled with the term 'given information' as sometimes they had to pro-actively seek it.

Respondents did not clearly understand what distinction there was between information given around things like 'affect your everyday life', 'manage your condition' 'change over time' 'care or treatment' etc. and there were specific problems around some of these (e.g. doctors often did not know how the condition might change over time because the condition was too unpredictable).

In a later section on social care and other support we ask about advice given on welfare benefits. In fact, most respondents said they had got this from a specialist organisation rather than a social care or healthcare professional.

For these reasons:

- We changed the title of the section from Diagnosis to Information about your condition'.
- We have added 'I have had a neurological condition from birth' to Q1.

- We have focused remaining Qs on information gained anytime since first finding out they had a neurological condition and not just around 'the period of diagnosis'.
- We deleted Q2 as it did not seem to be consistently understood or eliciting any useful information.
- We separated out questions about healthcare professionals and refer to being 'given' information or told things by healthcare professionals.
- We ask whether they received information enough information from a healthcare professional and about whether it was helpful/ timely.
- We specifically ask if any healthcare professional has told them about the relevant specialist organisation for their condition.
- We moved questions about information on welfare benefits to this section and are more specific about asking where these came from.
- The questions about information from specialist organisations are more targeted and the questionnaire refers to 'getting' information from them, which includes pro-actively seeking information.
- We moved the question about welfare benefits to this section as we
- We ask whether the respondent has received information from a specialist organisation so will have an estimate of what percentage of people did get information from a specialist organisation and how useful/ timely this was (enabling some comparison with information received from healthcare professionals).

YOUR CARE PLAN/ PLANNING YOUR CARE

These were generally considered to be related sections and so we merged them with Qs 9, 10, 11 and 12 moving into the Planning Your Care section. Qs 13 and 14 were actually more about professionals and these were moved to later sections.

HEALTHCARE PROFESSIONALS

We moved Q13 about having a named health professional(s) you could approach for advice down into this section. Respondents found it hard to answer Qs 18 and 22 about getting answers you could understand. Sometimes the professional did not know the answer, either because they lacked training/ expert knowledge or because the information was unknowable at this stage given the respondent's condition. The questions also conflated this issue of getting information that was accurate/ useful with whether things were explained in plain English. In the end it did not seem that these Qs were able to elicit any particularly useful information – given the difficulties around distinguishing between these different aspects - so was removed. Other Qs in this section were fine.

SOCIAL CARE AND OTHER SUPPORT

We removed reference to healthcare professional in Q23, added an example of what we meant by emotional or psychological support and moved the Q on welfare benefits to the first section on Information. Otherwise this section was fine.

REHABILITATION

Respondents were not clear what was meant so we have tried to provide a succinct definition including some examples. Given the huge variety of rehabilitation undertaken (including variation of location, frequency, residential or day, duration etc. and that it could be ongoing) respondents found it hard to answer questions about location and timing. Given that rehabilitation was often ongoing they also found it hard to answer Q29 on sufficiency although could answer Q32 about whether it had made a positive difference. This section was therefore reduced considerably to ask whether or not they had received any (and if not whether they'd wanted it) and whether it had made a difference.

EQUIPMENT AND ADAPTATIONS

Despite the definitions given at the beginning, respondents were not always clear what was meant by adaptations or aids. There also appeared to be some 'blurriness' around exactly which changes we were asking about at different points in the section.

Consequently, we have streamlined the section. We refer to changes to your home rather than adaptations and make it clear we mean structural changes and the addition of aids such as grab rails. We have introduced clearer definitions and examples throughout and have ensured questions are clear about which changes we are referring to.

APPENDIX C QUESTIONNAIRE AMENDED FOLLOWING COGNITIVE TESTING

APPENDIX D PROPOSED COVERING LETTER AND REMINDER LETTER

To be printed on NHS Trust headed paper? Text in square brackets to be edited.

[Date]

Dear [Name]

National Survey of People with Long-Term Neurological Conditions (pilot survey)

We are inviting you to take part in a pilot survey of people with long-term neurological conditions. These include conditions like epilepsy, multiple sclerosis, myasthenia gravis, Parkinson's disease and Alzheimer's disease that affect the brain and nervous system in some way.

What is the purpose of the survey?

Your views are very important in helping us to find out how well the health and social care provided for your condition meets your needs, and how it can be improved. This is your chance to have a say in how services relating to neurological conditions are provided in the future.

Why have I been chosen?

You are being invited to take part in this pilot survey because you have been in contact with a hospital regarding your condition. Your name was randomly selected from a list of people who had used hospital services for a neurological condition.

Do I have to take part?

No. Taking part in this survey is **voluntary**. If you choose not to take part it will not affect the care you receive from the NHS or Social Services in any way. If you do not want to take part in the survey, or to answer some of the questions, you do not need to give us a reason.

If you do not wish to take part, please could you return the blank questionnaire in the FREEPOST envelope. You can also call us to say that you do not want to take part on the helpline number given at the end of this leaflet. If we do not hear from you we may send you a reminder.

If you would like to take part but there are some questions that you do not want to answer please complete the questionnaire leaving these questions blank.

Who should complete the questionnaire?

The questions should be completed by the person named on the envelope, if possible. If that person needs help to complete the questionnaire, the answers should be given from his or her point of view, not the point of view of the person who is helping.

What would I have to do?

If you decide to take part, please complete the questionnaire and return it in the FREEPOST envelope. No stamp is needed. The questionnaire should take around 20 minutes to complete.

If it is easier for you to complete the questionnaire electronically, then there is a web version at www.XXX.org.uk or you can request a Word version by e-mailing LTNC@natcen.ac.uk.

Who is organising the survey?

The survey is being carried out by researchers from the National Centre for Social Research on behalf of the Healthcare Commission.

Will my response be kept confidential?

Yes. You have been given a unique number so your name and address are not on the questionnaire, and your name and address will never be linked to your responses. Your responses will only be used to provide information about the quality of the services provided and help to improve these services.

Your personal data are held in accordance with the Data Protection Act 1998 and the NHS Confidentiality Code of Practice.

Contact for further information

If you would like more information about the survey, or have questions on how to complete the questionnaire, you can call [our FREEPHONE help line /us] on [phone number] [at no cost to yourself] and we will do our best to help. The line is open between [opening time] and [closing time], [days].

Thank you

Yours faithfully,

[Letter from Trust Chief Exec, Neurologist (depends on Trust preference)]

To be printed on NHS Trust headed paper? Text in square brackets to be edited.

[Date]

Dear [Name]

National Survey of People with Long-Term Neurological Conditions (pilot survey)

We recently contacted you about a survey of people with long-term neurological conditions. We are contacting you again because we have not received a questionnaire from you.

If you have recently returned the questionnaire to us you do not need to do anything in response to this letter.

We are writing to provide some more information about the study, including answers to some of the questions we have been asked by those invited to take part.

The survey is about the experience of people with long term neurological conditions and is designed to assess health and social care provided. We would like to collect the views of as many people as possible to get a clear and accurate picture of how well health and social care meets the needs of people with long term neurological conditions.

You response is valuable to us because we can only include people invited to take part in the survey and we cannot replace you with another person. We would like the information we collect to represent as many people as possible.

We have included responses to frequently asked questions on the back of this letter. If you would like further information, have a question about the survey that is not answered here, or would prefer us not to contact you again, please contact us on FREEPHONE [insert telephone number].

Thank you.

Yours faithfully,

[Letter from Trust Chief Exec, Neurologist (depends on result of PIAG discussions)]

National Survey of People with Long-Term Neurological Conditions (pilot survey)

What is the survey about?

The survey is designed to assess how well health and social care meets the needs of people with neurological conditions. We would like to collect information from people with a range of different conditions to get a clear picture of health care needs.

How was I chosen?

Your name was chosen from a list of people who have used hospital services for a neurological condition. Only people who have been invited to take part in this survey can take part.

We rely on the goodwill and voluntary co-operation of people invited to take part to get an accurate picture of health care for people with neurological conditions.

What happens to the information I give?

The information that you give will be kept confidential and held in accordance with the Data Protection Act 1998 and the NHS confidentiality Code of Practice.

The answers that you give will be added to questionnaire responses of other people invited to take part. When we report on the survey findings we will talk about the information from the group as a whole, e.g. we may say that 50% (half) of people found out that they had a neurological condition in the last year.

Do I have to take part?

You do not have to take part.

If you would prefer to opt out, please return the blank questionnaire to us in the FREEPOST envelope or call us on our FREEPHONE number [insert telephone number]. We may send another reminder if you do not hear from you.

If you would like to take part but there are some questions you would not like to answer please leave these questions blank.

Can I give the information another way?

The questionnaire is carefully designed to collect the same information from everybody who takes part. Everyone invited to take part was sent a paper version of the questionnaire.

If you prefer to use a computer to complete this questionnaire, a web based version is available at our website (insert web address) or you can request a Word version by contacting us at LTNC@natcen.ac.uk.

You can also ask a friend, family member or carer to help you if you have any difficulties completing this questionnaire.

Contact for further information

If you have any questions about the survey or how to complete the questionnaire, you can call our FREEPHONE number [insert phone number].

APPENDIX E LETTER FROM BMA ON SAMPLING ISSUES

APPENDIX F SUGGESTED PROTOCOL

Protocol for pilot survey of people with Long Term Neurological Conditions

In March 2005, the Department of Health published a National Service Framework (NSF) for Long-term Conditions, with a focus on long-term neurological conditions. The NSF set out 11 quality requirements (QRs) for standards in services for neurological conditions, based on evidence from people providing and using these services, with a commitment to implement these QRs fully by 2015.

The following year, the Healthcare Commission asked the National Centre for Social Research to explore the feasibility of carrying out a survey of people with long-term neurological conditions, with a focus on the areas highlighted in the NSF and its QRs.

The initial phase of research involved a consultation with people working in the field, people with long-term neurological conditions and carers. The objectives of the consultation were to establish whether it would be possible to conduct a survey of people with long-term neurological conditions, to explore priority topics to cover in such a survey, and evaluate potential methods for sampling and carrying out the survey.

The report from the initial phase of the research was produced in August 2007. It showed that there was a lot of support & enthusiasm for a survey, among both professionals and service users.

The next stage of the research is to **pilot a survey on a small scale**, to assess how practical and useful it would be to conduct the survey on a national scale and / or to provide advice to PCTs and Acute Trusts on how they can conduct their own surveys.

The initial report identified a number of sources from which a sample could potentially be drawn. These included patient records from specialist hospitals or GPs, as well as other sources such as DWP benefits records. Having considered the options and discussed them further with the BMA and with a small number of Trusts, we feel that the optimal approach in this instance would be to sample via hospital records. More so than any other method, this is both practical (as we can sample from electronic records), and provides reasonable (albeit not perfect) coverage of the population of interest (i.e. people with long term neurological conditions).

To test out this approach, we will ask three hospitals that specialise in neurological conditions to draw us a sample.

The sample would be based on patients with an appropriate ICD 10 code (G00 – G99, but excluding G43 and G44). Ideally, we would want to include both in-patients and out-patients, and we would like to include records going back for 5 years. We would only include patients aged 16-65 at the time of their contact with the hospital. We have discussed this sampling with a small number of hospitals, and all agreed that, with proper ethical and data protection scrutiny, it would be feasible to draw such a sample. Prior to drawing a sample, we would look at the distribution of cases by ICD code to assess whether any disproportionate sampling should be done to allow for a broad range of conditions to be included.

We would ask Trusts / hospitals to check the sample for deceased service users via the National Strategic Tracing Service (which is standard practice for other patient surveys).

The size of the sample will be around 500 per hospital (to be randomly selected from all eligible patients).

For the patients sampled, we would minimally need to have access to the following information:

- Name (including title)
- Address (including postcode)
- NHS number (to ensure that each person is only included in the sample once)

We would also ideally like to have the following information. However, this information could be provided in aggregate for the sample to allow us to compare the achieved sample with the issued sample to assess non–response bias.

- Sex
- DoB or age
- ICD 10 code
- Date of most recent contact with hospital

Those sampled would be sent a self completion questionnaire by post. A covering letter will explain the voluntary nature of the survey, and provide a freephone number that people can use to opt out. The letter will explain that if the named respondent is not able to fill out the form on their own, then they can seek help with completing the form. We propose sending up to two reminders to those who do not respond. These mail outs could be undertaken by either NatCen or the Trust, depending on discussions with each Trust. (If NatCen undertakes the mailing, we would set up a service contract with the Trust).

The questionnaire will be between 12-16 sides of A4. As well as the postal version, the questionnaire will also be available on the web, and as an electronic form (in Word).