QORU | quality and outcomes of person-centred care policy research unit

Engagement of people with long-term conditions in health and social care research: Barriers and facilitators to capturing the views of seldom-heard populations

Julie Beadle-Brown, Sara Ryan, Karen Windle, Jacquetta Holder, Agnes Turnpenny, Nick Smith, Lisa Richardson and Beckie Whelton

University of Kent

THE LONDON SCHOOL OF ECONOMICS AND POLITICAL SCIENCE ■

UNIVERSITY OF OXFORD

The Policy Research Unit in Quality and Outcomes of person-centred care (QORU) is a collaboration involving researchers in health and social care from the Universities of Kent, Oxford and the London School of Economics (LSE) funded by the Department of Health.

Our aim is to improve the quality of health and social care of people with long-term conditions through generating high-quality evidence about need, quality and outcomes of person-centred care.

Contents

Introduction

The importance of including socially excluded groups in health and social care research has become increasingly recognised, and is underpinned by recent UK government policy (see, for example, Inclusion Health, Cabinet Office 2010). There is an acknowledged need for more sophisticated and flexible responses to improve access and quality of services for socially excluded groups. An integral part of this improvement is the inclusion of the views of socially excluded groups in both consultation and research about health and social care. The definition of seldom heard or socially excluded is not straightforward and, at its broadest, can include the long-term unemployed, those in severe and persistent poverty, people experiencing domestic violence, care leavers, ethnic minority groups, ex-servicemen and women, people living in remote areas, and those who do not meet the necessary eligibility criteria for the provision of statutory provision of care interventions, i.e., self-funders. More commonly, the focus is on those considered to be most vulnerable: homeless people, traveller groups, sex workers, people with intellectual disabilities, refugees, asylum seekers and prisoners or ex-offenders (Social Exclusion Task Force, Cabinet Office 2010). Those with long-term health conditions (an illness or condition which requires treatment, management or support for the rest of someone's life) may be seldom heard because their long-term condition (such as an intellectual disability, dementia, stroke, physical disability, mental health condition, or physical frailty) makes participation more difficult. They may have more than one health issue and/or also be part of one of the socially excluded groups noted above, which further exacerbates their exclusion from research.

A failure to "include all sections of society in research precludes a comprehensive understanding of population (health) issues, including potential differences in the manifestation of such issues within and between population subgroups, which in turn might impact on the development of effective services or interventions. Second, the engagement of socially excluded groups in research is a matter of social justice: Excluding ethnic minorities from health research might perpetuate existing power imbalances and inequalities by impeding action to improve the situation of all members of society. This might, in turn, contribute to marginalization… and widen inequalities" (Rugkåsa and Canvin 2011, p132).

People might be seldom heard in a variety of different ways. Firstly, they may be hard to reach – that is, it might be difficult to find them. For example, people might not be regularly accessing services (primary, secondary or tertiary health services or social services) for a variety of reasons and may not be included in research that recruits via such services. Some groups, such as sex workers or illicit drug users, may not want to be found and may actively resist engagement. Secondly, people who are using services might be hard to engage in research. This may be because of cognitive, intellectual or sensory impairments, language or cultural differences. It is in the field of intellectual impairment that the issue of inclusive research has been most consistently considered in the past.

Difficulties with memory, concepts of time and making comparisons have meant that engagement in research requires adaptations of methods, or the creation of new methods, which can pose challenges. Much of the early research which addressed these issues involved people with intellectual disabilities as participants and was concerned with the evaluation of different residential arrangements, including the move from long-stay hospitals to community-based services (McConkey 1996, Kroese et al. 1998). The growing interest in the conceptualisations and measurements of quality of life (Schalock 1996) also contributed to the involvement of service users as participants in research. A considerable body of literature had emerged by the end of the 1990s that addressed the methodological concerns in eliciting the views of people with intellectual disabilities and enhancing the validity of findings from quantitative and qualitative research (Heal and Sigelman 1996, Ramcharan and Grant 2001).

In qualitative interviews, the key question has been how to enable people with intellectual disabilities to express their views. Early literature addressed questions of interview design, who should be involved and how, where the interview should be held, and what safeguards should be in the data collection (Atkinson 1988). From the 1990s there was also an increased interest in the use of oral and life history research with people with intellectual disabilities (Atkinson 2005, Hamilton and Atkinson 2009).

Despite the rapid development in both researchers' assumptions and methods of including people with intellectual disabilities as participants in research before 2000, there remained some questions. In their review of the representation of the views and experiences of service users with intellectual disabilities, Ramcharan and Grant (2001) argued that one of the main challenges of research was to begin to establish the views and experiences of people with profound intellectual disabilities who lack verbal communication skills. They also warned of the dangers of extrapolating the views of people with intellectual disabilities who can speak for themselves to those who cannot. One approach that has been used in the past where people have communication difficulties has been the use of proxy respondents to comment on the situation for people who are not able to comment themselves. Historically, in large-scale quantitative survey designs, individuals that might now be considered hard-to-engage had been judged 'unsuitable for interview' or 'uninterviewable', and in turn treated as a type of non-response alongside refusals (Moser 1958). The use of proxy respondents has been an attempt to ensure that those people who were not able to be interviewed or complete questionnaires still had their experiences represented in research. There has been a substantial body of research on the validity and reliability of proxy respondents and tools designed to measure quality of life (Dalemans et al. 2009, Nota et al. 2006, Ouellette-Kuntz 1990, Cummins 1991, 1997).

Another way that has been used to ensure that the views and experience of people with severe and profound intellectual disabilities as well as more recently those with dementia are heard has been the use of observation. In particular, non-participatory research has been used since the early 1980s, to look at the day-to-day lived experience of people with

4

intellectual disabilities living in supported accommodation. This type of approach carries its own issues in terms of gaining consent as well as some biases associated with observation, but it has been found to provide a picture of the lives of people and the quality of the support they receive (see Mansell 2011 for a recent review).

This rapid review aimed to explore the recent literature about the barriers and facilitators to including seldom heard groups as participants in research. The review did not include the issue of emancipatory or participatory research, except where this was explored as a specific method to increase the involvement of seldom-heard groups as participants in research. Three core questions which have relevance to policy and future research practice guided this review:

- What do we know about whose views and experiences that are excluded from research, and how often does this happen in health and social care research?
- Why are some people's views and experiences not heard?
- What methods are there for facilitating people's views to be heard, and are these facilitators population-specific or can they be applied to other groups and guide good research practice more generally?

Methods

The search activities were undertaken by a team of eight researchers between May and August 2011. An initial scoping exercise was carried out to explore the existence of papers or reports that evaluated or assessed mechanisms for effective inclusion of 'seldom-heard' groups. As the review was undertaken to inform the research programme of the policy research unit in Quality and Outcomes of person-centred care, which is concerned with long-term conditions (LTCs), there was a particular interest in research with and about people with LTCs. A range of key words were identified that included:

- Specific long-term conditions or diagnoses (e.g., dementia, chronic obstructive pulmonary disease, schizophrenia, intellectual disability);
- Marginalised populations (e.g. asylum seekers, black and minority ethnic groups, homeless, traveller groups, etc.);
- The type and extent of involvement and engagement across the research pathway (e.g., access, consultation, mistrust, attrition etc.); and
- A broad range of research methodology (e.g., action research, focus groups, systematic reviews etc.).

Research relevant to both health and social care was included. In addition to research relating to long-term conditions, three initial inclusion criteria were put in place: the literature review or empirical research had to be English language, peer-reviewed and published after January 2000. A listing of 37 databases, along with web-sites of government agencies, academic and third-sector networks, was developed to support this initial search.

Following this early exercise, parameters were refined, due to the sheer volume of studies returned. Only studies that commented on engaging people from seldom-heard groups as research participants were included. Research which focused on participatory research was not included in this review unless using service users as researchers was employed specifically as a way of increasing recruitment and engagement in the research. In order to make the review manageable and to ensure that the results reflect the relevant policy and practice context, only research conducted in the UK and published in peer-reviewed journals from 2001 onwards were included. The final inclusion and exclusion criteria are summarised in Table 1. Eighteen databases were identified for the final search, and slight variations in the key search terms were applied to different databases, ensuring appropriate identification and retrieval of abstracts. In addition, key references in the extracted articles were followed up and, where they met the inclusion criteria, were included in the review. A full list of the search terms and databases used can be found in the Appendix.

Table 1: Systematic rapid review: inclusion and exclusion criteria

Area	Inclusion	Exclusion
Country	UK	Non-UK
Date of publication	January 2001 – current date	Before 2001
Language	English	Non-English
Design	Any study design, including reviews, primary research, discussion and opinion articles	None
Publication type	Peer-reviewed articles	Non-peer-reviewed articles, grey literature, books, book chapters
Focus of the paper	Research participation and engagement in the field of health and social care (which may or may not be the focus of the primary research where relevant)	Other primary findings relating to these populations
Participants	Adults with a long-term health condition/s plus people with an intellectual or developmental disability, cognitive or sensory impairments, and those from minority groups such as BME groups, travellers groups, etc. (See full list in the Appendix).	Children and adolescents under 18, those with cancer or receiving palliative or end of life care; conditions which were viewed as possible risk factors for long-term conditions, such as drug and alcohol misuse or dependency, hypertension, obesity and pain, smoking etc., prisoners or ex-offenders

In total 2,031 abstracts were identified. Each citation title was then checked for relevance against our review criteria by a second team member. Non-relevant papers were excluded (n=1494) and a total of 537 papers were retrieved. Abstracts were reviewed by one reviewer to assess inclusion or exclusion in the review. Where there were queries or concerns as to inclusion, the abstracts were read by a second team member and consensus reached through discussion. In some cases this process was repeated with full papers at a second stage of review. A total of 107 papers were included for full extraction. Following assessment of quality (see below) a further 24 papers were excluded, leaving 83 studies. (see Figure 1).

Figure 1: Identification of eligible papers for the systematic rapid review.
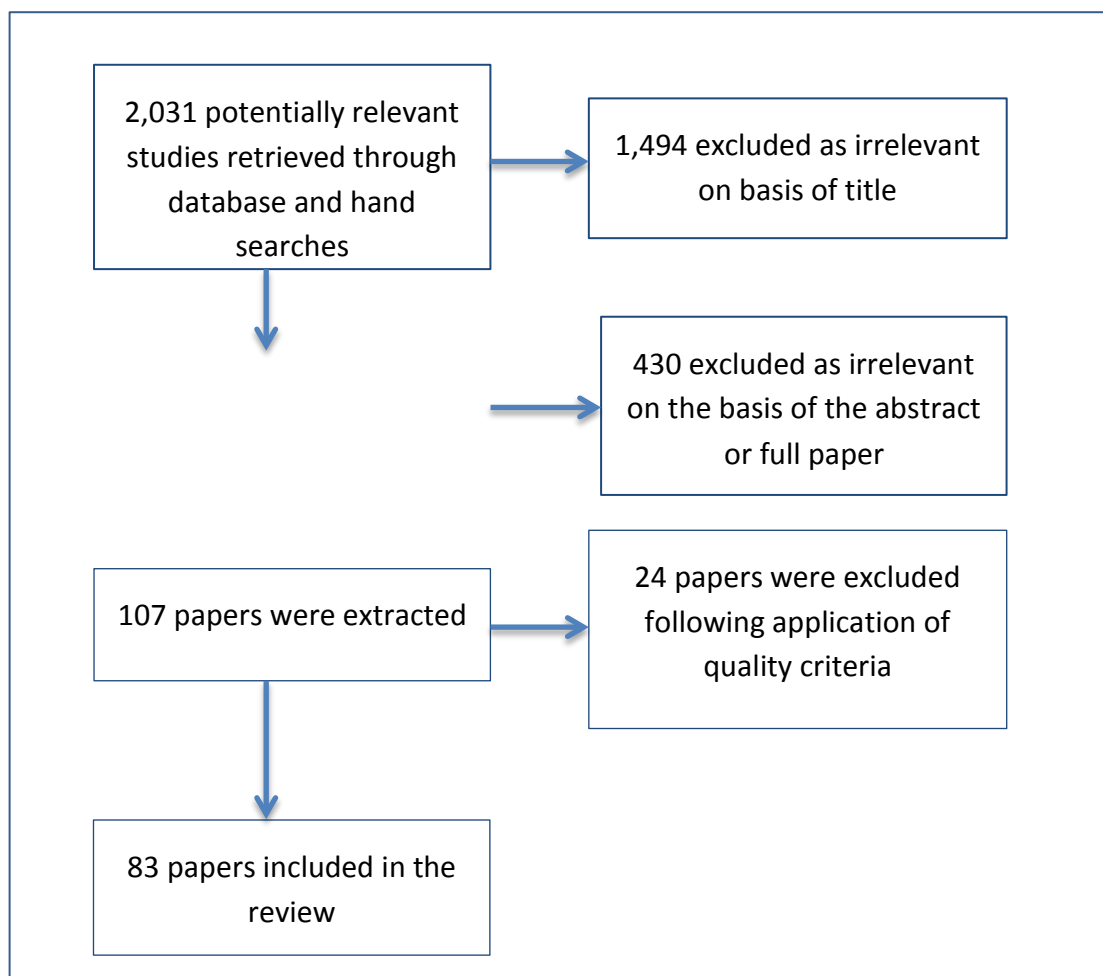
While several quality appraisal tools exist (e.g., the Critical Appraisal Skills Programme http://www.casp-uk.net/ref), the diversity of the extracted papers meant that no single tool could be effectively used (Woodall et al. 2011). A simple five-level classification was devised ranging from 'robust' to 'too poor to include' (see Table 2).

Table 2: Criteria for critically appraising research

Our Rating	Description	NICE Rating	NICE description (NICE 2006)
Robust	Good paper (robust, valid, transferable etc.)	++	Review or study fulfils all or most of the NICE criteria; where criteria have not been fulfilled, the conclusions are still thought very unlikely to alter if the study were replicated.
Adequate	Adequate paper (a few problems, unlikely to be transferable, sample small etc.).	+	Review or study fulfils several of the NICE criteria; those criteria not fulfilled or adequately described are thought unlikely to alter the conclusions if the study were replicated.
Limited	Poor paper (methods used not appropriate, not transferable, sample too small etc.).	-	Review or study fulfils few if any of the NICE criteria; the conclusions are thought likely or very likely to alter if the study were replicated.
Too poor to include	Extremely poor paper, considered too poor for inclusion		
Query	Reviewer unsure of rating or wanted a second opinion from another member of the review team		

The data extraction form designed for the review included the following sections: the type of publication, study design, population, facilitators or barriers to engagement of seldom-heard populations, cost analyses, use of proxies and quality assessment (see Figure 2 in the Appendix for full list of information extracted).

A combination of mapping and narrative synthesis was adopted to summarise the findings from the papers reviewed (Popay et al. 2006, NICE 2006). Nine variables (drawn from the extraction form) were mapped: type of publication, study design, study population, method of data collection, method of analysis, economic cost implications, use of proxies, relationship between service user and proxy and quality appraisal. The thematic analysis concentrated on information extracted about facilitators, and barriers including ethical issues around research participation, and the use of proxy respondents. Text from the data extraction form was imported into NVivo for analysis and organised into emerging themes. The analysis was carried out by two team members with comments on the resulting synthesis by the wider team.

Findings

Mapping the evidence

The aim of the mapping was to provide summary information on the characteristics of the studies that would complement the thematic analysis of barriers, facilitators and ethics, and to provide an overview of the extent and comprehensiveness of the literature.

The type of publication was categorised into primary research, review and opinion piece. Primary research studies were further coded into quantitative, qualitative or mixed-methods papers. The majority of the studies extracted were primary research (82 per cent, n = 68). Of these, 35 per cent (n = 29) were quantitative, 43 per cent (n = 36) qualitative and four per cent (n = 3) mixed-methods papers. Twelve per cent (n = 10) were literature reviews while six per cent (n = 5) were opinion pieces.

The primary research study design was coded as reported by their author(s). If no study design was explicitly reported, a judgement was made by the individual extracting the relevant paper. The distribution of papers according to study design is summarised in Table 3.

Table 3: Distribution of primary research papers in the review by study design

Study Design	Extracted Paper Number (68)	Percentage
Survey	23	34
Qualitative study	32	47
Ethnography	2	3
Experimental design	8	12
Mixed designs	3	4

Six categories were used to describe methods of data collection: structured questionnaire (including those administered face-to-face), semi-structured interview, focus group, and systematic and non-systematic review. The category 'other methods' included less commonly used qualitative methods such as photographic participation, participant observation and nominal group technique. Mixed-methods studies combined two or more data collection methods (see Table 4).

Table 4: Data collection methods in primary research studies and reviews

Methods of Data Collection	Extracted Papers Number (78)	Percentage
Interview	16	20
Questionnaire	23	30
Focus group	10	13
Mixed-methods	16	20
Other methods	3	4
Non-systematic review	9	11
Systematic review	1	1

The number of study participants ranged from two to over 2,000, with the majority of the papers including users as well as carers or proxy respondents. For example, 16 papers (19 per cent) explored the use of proxies as well as users, assessing the levels of agreement between users and their proxies. Three papers did not report the number of participants while, in two of the papers, this data was not clear. Table 5 shows number of participants in the primary research papers.

Table 5: Primary research papers by number of participants

Number of Participants	Extracted Papers Number (68)	Percentage
20 or fewer	18	24
21-50	14	19
51-100	16	22
101-500	8	11
more than 500	7	7

Table 6 summarises the population groups on which the papers focused. The majority of papers focused on people with intellectual disabilities, with older people including those with dementia making up the second biggest group (20 per cent). Some research on other populations was found, but in general these only accounted for a small number of the total sample of papers reviewed. A more detailed breakdown of those study populations in our retrieved papers can be found in the Appendix (see Table 9).

Table 6: Study populations in reviewed papers

Focus of the paper	Extracted papers (Number)	Extracted papers (Percentage)
People with intellectual disabilities	41	49
Older/dementia	17	20
Mental health	9	10
BME	7	8
Deprivation	3	3
Other	10	11
Total	87	100

*The total number of papers exceeds 83 as four papers focused upon more than one population.

Of the 83 papers included, only 19 (23 per cent) included a discussion of the economic or resource implications of carrying out research with groups who experience barriers to participating in research studies or implementing methods to facilitate greater participation.

Just over a quarter of papers within this review were evaluated as robust: that is, the review or study was valid and transferable (see Table 7).

Table 7: Critical appraisal rating by type of publication

Critical appraisal rating	All papers	Qualitative	Quantitative	Not primary research	Mixed design
Robust	23 (27%)	6 (17%)	13 (45%)	4 (27%)	0 (0%)
Adequate	44 (53%)	21 (58%)	13 (45%)	8 (53%)	2 (67%)
Limited	16 (19%)	9 (25%)	3 (10%)	20 (3%)	1 (33%)

The majority of the papers described methods used when trying to conduct research with seldom-heard populations. Only eight studies set out to compare different strategies or methods for involving hard-to-reach or engage groups (Allison et al. 2003, Lloyd et al. 2008, Mathers 2008, McKeown et al. 2010, Murphy and Cameron 2008, Pawson et al. 2005, Rooney et al. 2011, Rugkasa and Canvin 2011). Similarly, only nine studies were designed from the outset to investigate reasons for participation or non-response (Cameron and Murphy 2007, Harkins et al. 2010, Howard et al. 2009, Lowton 2005, Oliver et al. 2002, Tallon et al. 2011, Ulivi et al. 2009, Williams et al. 2007, Woodall et al. 2011).

The effectiveness of recruitment methods was rarely evaluated explicitly, and reporting of different types of non-response was patchy, even if the response rate was particularly low: e.g., when only 26 per cent of potential participants responded to their invitation (Lowton 2005). Some studies usefully differentiated between refusals and 'no-shows' (Cambridge and McCarthy 2001, Cameron and Murphy 2007) and some noted withdrawals, as well as refusals (Cameron and Murphy 2007). However, only six papers considered the likely rationale behind non-participation (Cameron and Murphy 2007, Lloyd et al. 2008, Oliver-Africano et al. 2010, Sheikh et al. 2009, Tallon et al. 2011, Williams et al. 2007). Of these, only one study directly followed up those that did not wish to participate (Williams et al. 2007), while a second carried out secondary analysis to assess if there were particular groups that did not respond (Tallon et al. 2011).

Findings from the narrative synthesis

The narrative synthesis of the barriers and facilitators discussed within the literature produced themes that broadly follow the research pathway. As many of these themes cut across the seldom-heard groups, the research pathway – rather than individual populations – is used as our framework for organising the data.

- Assumptions of researchers, overarching research design and ethical procedures

- Sampling, recruitment and gaining consent

- Data collection
- Analysis and interpretation

Assumption of researchers, overarching research design and ethical processes

Researcher assumptions were noted to significantly influence research design and lead to the exclusion of certain groups of people (Nind 2009, Proctor 2001) These assumptions create barriers at different points in the research process. Assumptions are often made early on about the efficacy of including particular groups of people because their views are not possible to access, valid or worth considering, or that they are not coherent or lucid enough to express a view (Coucill et al. 2001, Nind 2009, Proctor 2001). For example, people with intellectual disabilities may not be included as they do not 'fit' with researchers' ideas of what a participant should be and how they should respond (Aldridge 2007). Similarly, a study comparing US and British researchers' attitudes to the inclusion of minority populations in research found evidence of some stereotyping and prejudice among British researchers (Sheikh et al. 2009). There is also concern relating to fragmented accounts (for example, in research with people with dementia), and the need for gaps and inconsistencies to be interpreted by researchers (Lloyd et al. 2006, Proctor 2001). This again relates to assumptions about what data should look like; less conventional forms of data such as seemingly incoherent accounts are challenging for researchers.

The narrative synthesis identified some ways of reducing some of the barriers encountered in the early stages of research design. At a general level, flexibility across the research

process has been identified as a core facilitator in ensuring the involvement of seldom-heard groups (Rugkasa and Canvin 2011, Tuffrey-Wijne and Davies 2007, Wilson et al. 2010). Gilbert (2004) suggested that study designs should allow, and plan for, the use of different methods with different groups of people within the same study. In terms of specific designs, the use of cluster randomised trials, or a Zelen design,[1] were recommended for improving 'in-trial' participation of people with intellectual disabilities.[2] Both methods overcome the barrier of patient or clinician preference as to treatment, and some researchers have argued that such designs should be used to improve the participation of people with intellectual disabilities in trials (Oliver-Africano et al. 2010).

A key recommendation for facilitating appropriate and inclusive research design was to include stakeholders (e.g., people with dementia or intellectual disabilities, mental illness, minority ethnic communities) in reference or consultation groups to contribute to the design and on-going oversight of projects. Such involvement, it is argued, can aid study design, recruitment, data collection and the discussion of findings (Howard et al. 2009, Jones 2008, Lloyd et al. 2008, McKeown et al. 2010).

The need to gain ethical approval can shape research design in ways that may obstruct the inclusion of particular groups (Allbutt and Masters 2010, Clegg 2004). Study protocols approved by funding bodies and ethics committees restrict the ways researchers can respond to shifting positions, and there is some tension between the approach and assumptions underpinning the workings of local research ethics committees and the assumptions underpinning the research (Allbutt and Masters 2010, Gates and Waight 2007). Inflexible procedures - such as specified frameworks, standard information sheets in advance, an opt-in rather than opt-out approach to recruitment, restrictions on the number of reminders that researchers can send out and, in some cases, a ban on following up non-participation – can create challenges in research with some groups (McKeown et al. 2010, Williams et al. 2007). People with intellectual disabilities are not necessarily independent decision makers: they are interdependent with their carers. If ethical approval processes do not include negotiation with carers, individuals can be isolated from their social system, leading to exclusion from research participation (Clegg 2004).

The narrative synthesis identified some specific recommendations about ethical issues at particular stages of the research pathway, and these are discussed in more detail later in

---

[1] Where patients are randomised to either the treatment or control group before giving informed consent.

[2] In cluster randomisation the different arms of a trial are randomised through social clusters (e.g, hospital or general practice). This is thought to minimise recruitment barriers (individuals are familiar with the setting) found in standard randomised control trials, as well as sample contamination as those in a similar cluster receive the same treatment. The Zelen design is more controversial, as it involves randomisation prior to gaining consent. Similarly, it places slightly more pressure on those in the control arm as they are included in an observation study, rather than the more simple 'usual treatment'.

this review. However, general ethical recommendations were found in relation to research with on-line communities (Brownlow and O'Dell 2002) , which were found to facilitate inclusion in research for some seldom-heard groups (e.g., people with autism), but bring their own methodological challenges, such as the invasion of privacy with observation of conversations in real-time chats or on listservs.

Sampling, recruitment and gaining consent

Defining the research population

Some minority populations – such as travellers – are not easy to define and it is impossible to determine the – sample size (Brown and Scullion 2010, Mathers 2008, Oliver et al. 2002). Problems also arise when minority populations - such as Black or Asian – are grouped together, obscuring the experiences of particular groups, such as people of 'mixed race' or less visible ethnic groups, such as migrant workers (Garland et al. 2006). Other groups – such as people with intellectual disabilities and dementia - are also diverse, and questions remain about whether the views of those who can speak for themselves can be extrapolated to those with severe disabilities and communication difficulties. A further challenge is whether potential participants define themselves as part of the research population.

Finding, sampling and engaging seldom-heard groups

Finding participants can be problematic. In health and social care, initial approaches to potential research participants are often made through, or with the help of, professionals in the statutory, independent or private sector, and this was often found to be the case in the literature reviewed (Abbott et al. 2005, Allbutt and Masters 2010, Allison et al. 2003, Barr et al. 2003, Boyden et al. 2009, Cambridge and Forrester-Jones 2003, Cambridge and McCarthy 2001, Cameron and Murphy 2007, Hancock et al. 2003, Lloyd et al. 2008, Lowton 2005, Rugkasa and Canvin 2011). Qualitative and small-scale research typically used an unstratified two-stage sampling design. Selected service providers or clinicians were recruited to take part or facilitate recruitment, prior to any sampling of service users. Relying on the organisations' 'local knowledge' or eligibility criteria to screen or nominate potential participants, users were then further identified through a convenience or volunteer process (Abbott et al. 2005, Allbutt and Masters 2010, Allison et al. 2003, Boynton et al. 2004, Duckett and Pratt 2001, Dye et al. 2007, Gates and Waight 2007, Gordon et al. 2007, Gosden and Kirkland 2008, Jones 2008, Ulivi et al. 2009).

Recruitment and sampling can be hampered by a wide range of factors:

- Inaccurate or missing records, demolished and untraceable addresses, misunderstandings and the different settings in which people live (Harkins et al. 2010, Parry et al. 2001).

- A tendency to use existing 'groups' of people with intellectual disabilities as participants for ease of recruitment, leading to rehearsed answers (Kaehne and O'Connell 2010).
- Finding people with intellectual disabilities who live at home or are not using services (Veenstra et al. 2010).
- A lack of interest by potential participants in the research area; it may not be a priority in their lives or they may not feel it will benefit other people or themselves (Gilbert 2004, Harkins et al. 2010, Rugkasa and Canvin 2011, Williams et al. 2007, Woodall et al. 2011).
- Participants' health status. They may experience illness, pain and fatigue, may be reluctant to take time off work to take part, or feel that they may experience harm through participation (Andrews 2005, Cameron and Murphy 2007, Davies et al. 2010, Lloyd et al. 2006, Lowton 2005, Williams et al. 2007).
- Perceptions of research participation as onerous and demanding, or being put off by certain study-related tasks such as a reluctance to give information about themselves, physical assessment such as being weighed, or concern about side effects (Abbott et al. 2005, Harkins et al. 2010, Williams et al. 2007, Woodall et al. 2011).
- A fear of stigmatisation (Veenstra et al 2010)
- Forgotten appointments and difficulties understanding the study information (Abbott et al 2005).

Turning to facilitators, some papers included suggestions about how to increase participants' willingness to take part in research. These suggestions were primarily generic rather than population-specific. For example, where possible, it is recommended that researchers should identify any benefits for future patients because this taps into people's desire to help others (Howard et al 2009, Tallon et al. 2011, Lowton 2005). An important caveat or reservation about this approach concerns research that has no personal benefit for participants and mainly appeals to people's willingness to help: the Mental Capacity Act recommends that research which does not have direct benefits for the individual participant should only be conducted if the research provides knowledge about cause, treatment or care.

Increased time is often needed for negotiating access, contacting potential participants, gaining consent, developing appropriate information sheets, data collection and analysis. Time and resources are needed to conduct the often complex negotiations required to obtain approvals, agreement and co-operation from organisations or individual gatekeepers, such as carers or family members, or health and social care providers. Personal contact and meetings were reported as necessary: for example, agreement to take part from a care home manager was 'seldom achievable just by letter' (Zermansky et al. 2007 p258). One study recruiting a small sample of people with schizophrenia found that following initial

contact, an average of 10 visits per recruit was necessary to obtain written consent (Abbott et al. 2005).

Three studies found that using more than one recruitment strategy, concurrently or sequentially, increased the recruitment rate (Harkins et al. 2010, Lloyd et al. 2006, Rugkasa and Canvin 2011). There were some contradictory findings about which types of strategies were most effective. Rugkasa and Canvin (2011) found that paid bi-cultural researchers and self-referral following an intensive information 'marketing' campaign were more effective than asking workers in community organisations to facilitate contact and recruitment.

The use of flexible and multiple recruitment strategies to improve the accessibility of research may have implications for the nature of the resulting sample. Rugkasa and Canvin (2011) found that the type of strategy used affected the type of participants recruited: for example, those who responded to advertising tended to be service users rather than carers. 'Activists' in the field of mental health on a voluntary or paid basis and or those with higher educational attainment were also more likely to respond to advertising. Participants recruited through community groups included people from more diverse socio-economic backgrounds than the self-referral route. Those recruited by the paid recruiters included more participants born outside the UK than those recruited through the other methods.

Other studies recommended using grassroots organisations or existing service provision in the community to recruit black and minority ethnic participants (Boyden et al. 2009, Garland et al. 2006). However, there are some caveats: Rugkasa and Canvin (2011) found that several participants recruited by gatekeepers withdrew soon after. The authors speculate that they may have felt unable to refuse such a request, or that the study had not been appropriately explained. Rooney et al. (2011) suggested that sampling and recruitment of participants should take into account linguistic as well as ethnic backgrounds to maximise the diversity of the sample recruited from minority ethnic communities.

Information sharing and the circulation of targeted marketing material were identified as helpful recruitment techniques. Posters, leaflets, advertisements in newsletters, specialist publications or local press and mass mailings have all been used to promote awareness of research and self-referral (Fenge 2010, Harkins et al. 2010, Rugkasa and Canvin 2011, Ulivi et al. 2009).

Some studies recommended the use of face-to-face contact to improve recruitment in research projects, while recognising that this method will not be specific to seldom-heard groups (Rugkasa and Canvin 2011). Some evidence of this was available from studies on the recruitment of the oldest-old (aged 85 and over), with Davies and colleagues (2010) reporting that a letter from the GP caused confusion and that better practice was to accompany such a letter with one from the research team and then follow up with direct telephone contact or a home visit. This method is resource-intensive and in this study involved up to nine telephone calls or several visits per participant (Davies et al. 2010).

In terms of reaching socio-economically disadvantaged groups to participate in health research, recruitment through face-to-face canvassing in disadvantaged areas, as part of a community development approach, was found to be more successful than a social marketing campaign using direct mail methods (Harkins et al. 2010).

The use of incentives to promote recruitment was discussed in some of the review papers. There is debate around the ethics and benefits of payment for participation which – although not specific to seldom-heard populations – is of relevance (Roberts et al. 2004). In research with some groups or communities (e.g., black and minority ethnic communities), not paying participants may be perceived as entrenching power relationships, maintaining inequalities between the researcher and the researched (Rugkasa and Canvin 2011). However, one concern identified was that the promise of financial reward may exert a sense of coercion or obligation, bringing into question the 'voluntary' nature of participation (Rugkasa and Canvin 2011). A further worry is that payment does not support the identification and inclusion of the seldom-heard but rewards those participants wishing to be involved but who may need that extra 'nudge'. A more practical difficulty is that any payment made may impact on participants' eligibility for benefits (Jones 2008).

In research with mental health service users, Howard et al. (2009) and Woodall et al. (2011) recommended that researchers should proactively pre-empt and address any fears or misunderstandings, and stress the value of the research in terms of its potential to help future patients. It is suggested that combining these proactive approaches with a robust research process (convenient appointment times, respect, support and delivering feedback) will ensure more effective recruitment and participation (Tallon et al. 2011).

Communication

A lack of effective communication about research with potential participants has been highlighted as a barrier to engagement in various studies, particularly with people who may have difficulties communicating. Problems include unclear information sheets, too much information, and misunderstandings about the implications of taking part in research (Abbott et al. 2005, Finlay and Lyons 2001, Howard et al. 2009, McKeown et al. 2010, Ulivi et al. 2009). Inadequate communication about the research can also exacerbate the uncertainty or anxiety about taking part, especially around issues such as possible side effects, and thus hamper recruitment (Rooney et al. 2011).

Little research focused on the most effective strategies or techniques to communicate study information to specific populations. However, as is the case in any research, clear and understandable communication is a core facilitator throughout the research process. This is perhaps most evident in research with ethnic minorities, particularly people who do not have English as their first language. The use of different languages and formats with people from BME communities, both in the recruitment and fieldwork stages, was found to facilitate engagement: for example, consent obtained through audio rather than written recording, and project information sheets provided in audio format and a range of

languages (Allison et al. 2003, Lloyd et al. 2008, Rooney et al. 2011, Rugkasa and Canvin 2011). Nevertheless, any translation of supporting information, was noted to require considerable pre-testing, on-going development and back-translation to ensure accuracy and reliability (Allison et al. 2003). We return to communicating study information in the section on consent.

There is some debate about whether diagnostic terms (e.g., dementia, schizophrenia) should be avoided in recruitment materials to support the recruitment of people who, along with their relatives, may be unhappy with particular labels or diagnoses (McKeown et al. 2010). Woodall et al. (2011) suggest that, while researchers should use language carefully, they should also seek to engage with potential participants' understandings of their illness.

There is also some evidence that methods designed explicitly to include people with communication difficulties can still obstruct full engagement. For example, the Talking Mat tool (see below) has been found to be distracting for some people with intellectual disabilities, and the tool itself can lack clarity (Murphy et al. 2005). Using such technology to help people understand the nature of research and their potential involvement in it may therefore carry some challenges.

Settings

Inaccessible research venues or a lack of relevant support such as transport or childcare can create barriers to participation (Andrews 2005, Rooney et al. 2011, Tuffrey-Wijne and Davies 2007, Woodall et al. 2011). Cambridge and McCarthy (2001) found that 20 per cent of people who agreed to take part in their focus groups failed to turn up, largely because of transport and support arrangements. Some people may be reluctant to have researchers coming to their homes (Williams et al 2007, Jones 2008, Gates and Waight 2007).

Timing

For people with long-term conditions, the point at which people are approached within the illness trajectory can affect participation (Tallon et al. 2011). For example, Woodall et al. (2011) found that the timing of the approach of their study of people with mental health issues (during an inpatient stay) was given as the reason for refusal. Views about what was a better time to be contacted by researchers were mixed but, in general, it appeared that people preferred to be contacted once the main problem was over (Woodall et al. 2011). In a study involving adults with cystic fibrosis, it was found that when people were well enough to work they were reluctant to take time off work to participate in research (Lowton 2005).

Trust

Trust is a central dimension to enabling participation in research. Often people do not trust the person or institution asking them to take part in the study (Andrews 2005, Brown and Scullion 2010). Some studies found that past negative experiences with the health services or broader institutions create a culture of mistrust and suspicion about the purpose of the study (Harkins et al. 2010, Tuffrey-Wijne et al. 2008). Trust also relates to research fatigue;

people are weary of researchers targeting particular communities, such as traveller communities, and then disappearing (Brown and Scullion 2010).

Rugkasa and Canvin (2011) found that cultural issues create barriers to inclusion. A cultural distance between researchers and the researched group can create misunderstandings between them (Proctor 2001). This can lead to a lack of trust and scepticism arising from a failure to respect cultural and religious sensitivities (Rooney et al. 2011).

On an interpersonal level, some people may think they are not able to answer questions, or provide the 'right' answer (Proctor 2001). They may feel uneducated in comparison to the researcher. Similarly, they may have concerns as to the level of privacy or confidentiality, worrying about results being reported back to carers or care managers (Proctor 2001, Young and Chesson 2006, Ulivi et al. 2009).

To overcome barriers around trust with people with intellectual disabilities or communication difficulties, research has stressed the importance of focusing on ways of developing rapport and building on-going trusting relationships (Nind 2009, Tuffrey-Wijne et al. 2008). A further study suggested that (with the benefit of hindsight) it would have been productive to have built reciprocity into the relationship from the outset by planning for potential joint working and collaboration with participating BME organisations (Rugkasa and Canvin 2011).

Gatekeepers and supporters

Gatekeepers – including GPs, care managers, support workers, carers and parents – in protecting or selecting potential participants can act as a barrier to participation (Atkinson and Flint 2001, Cambridge and McCarthy 2001, McNally 2002, Oliver-Africano et al. 2010, Rugkasa and Canvin 2011, Tuffrey-Wijne et al. 2008, Zermansky et al. 2007, McKeown et al. 2010, Howard et al. 2009). The co-operation of gatekeepers will depend on the relationship they have with potential participants, how they perceive the research, and their judgement about who should be involved (McKeown et al. 2010). Allbutt and Masters (2010) report being surprised by the numbers and layers of gatekeeper involvement in their research involving mental health users. Despite having the approval of the regional management, the site manager refused to allow front-line staff to contribute to the study because they had not had particular training. These authors suggest that current ethical processes reinforce the gatekeeping role of front-line staff and managers.

GPs exclude patients for various reasons including poor health or literacy, patients who are new or unknown to them, patients who are perceived to be problematic for various reasons (e.g. addiction, chronic illness, etc.) or because of a perceived tension between the randomisation process and their aim of providing the best possible care (Howard et al. 2009, Parry et al. 2001 Tallon et al. 2011). The ambivalence of clinicians in intellectual disability services to research participation has been related to over-protection and a fear of blame if something goes wrong (Oliver-Africano et al. 2010).

There is also some evidence that supporters or advocates can negatively influence people's involvement during the research process through their assumptions or perceptions of service users' ability to take part (Kaehne and O'Connell 2010, Llewellyn 2009). Care managers, carers and parents may work from a medical model of disability which creates a culture of dependency and hinders research participation. Supporters can censor the views and the information shared by participants, particularly in research that concerns organisations and policies (Llewellyn 2009).

One study that explored the reasons for poor recruitment by trial staff recommended that researchers pay particular attention to educating clinicians about the reasons for a randomised control trial, the eligibility criteria, the concept of the control group and clinical equipoise, and the role of randomisation (Howard et al. 2009).

Mental capacity and gaining consent

The difficulties inherent in gaining informed consent and the associated issues of ethical approval were another key barrier to recruitment. Potential participants may be excluded because researchers find it challenging to determine whether or not people have capacity to give informed consent, or fear contravening potential participants' legal and ethical rights (Donnelly 2004, McKeown et al. 2010, Oliver-Africano et al. 2010). There is clearly a tension between protecting vulnerable people and allowing people who may not be able to give informed consent access to research participation. Dye et al. (2004) point out that not all aspects of consent are of equal importance. For example, understanding that participation is voluntary is more important than understanding the research protocol. The demands of ethics committees to engage with layers of approval from different people or institutions can also be obstructive. In one study, the ethics requirement was consent from the patient, assent from the primary carer and clinical agreement from the clinician. All three were needed for successful 'recruitment' (Oliver-Africano et al. 2010). Another study highlights a 'catch 22' scenario where researchers have to ask for the mental health service user's consent to approach their psychiatrist to ascertain their capacity, before they can invite the person to take part in non-therapeutic research (Ulivi et al. 2009).

Williams et al. (2007) noted that ethics committees currently require researchers to use 'opt-in'[3] rather than opt-out consent processes[4] where at all possible. However, Williams et al. argue that opt-in approaches for research that involves minimal contact (e.g. questionnaires, health care record consultation) promoted sample bias.

A widely-used technique for gaining informed consent, particularly in qualitative research, is that of 'process consent', where negotiation between the researcher and participant is carried out at different stages of the process to ensure on-going agreement to involvement

---

[3] Where people are contacted initially and asked whether they want to take part before questionnaires are sent or data collected from health practitioners.

[4] Where people are informed about the research and have to let researchers or other contacts know that they do not want to take part.

(Cameron and Murphy 2007, Jones 2008, Lloyd et al. 2008, McKeown et al. 2010, Nind 2009, Tuffrey-Wijne et al. 2008, Wilson et al. 2010). It is argued that capacity to consent to take part in research should be re-contextualised so that capacity is always viewed as a continuum, or matrix of decision-making, in which the individual is supported by a variety of factors (Dye et al. 2007). In this way, the focus is on the adequacy of the process rather than the ability of the individual. Warner et al. (2008) cautioned against the use of standard cognitive tests (e.g., mini-mental state exam) to identify capacity to consent, while Ulivi et al. (2009) recommended educational initiatives to improve and support capacity to consent.

To be accessible to those with communication difficulties, consent information will need to be 'individualised' to the language, memory and attention capacity of the participant and provided in different forms: written, signed, pictorial, oral or electronic (Boyden et al. 2009, Gates and Waight 2007, Oliver-Africano et al. 2010, Wilson et al. 2010, Young and Chesson 2008). The use of large fonts, highlighting of keywords, short sentences, simple language, white space, symbols and the repetition of information is recommended, especially for people with cognitive impairments (Wilson et al. 2010, Cameron and Murphy 2007). One study focusing on people with intellectual disabilities used the accessible language principles developed by Mencap in their illustrated study letter (Cameron and Murphy 2007). New technologies can be effective, with digital presentations of the research process. A mock research interview was developed and used in one study to support the consent process for people with intellectual disabilities (Mathers 2008, Pawson et al. 2005). An effective mechanism allowing appropriate consent to be achieved in different cultural communities was audio recording of patient information in different languages (Lloyd et al. 2008, Pawson et al. 2005, Rugkasa and Canvin 2011). In one study, recordings were provided in advance, allowing potential participants to listen to them in their own time. Audio-recorded consent was then obtained face-to-face immediately before the interview (Lloyd et al. 2008).

Use of third parties to help in decision-making[5]

Witnesses, 'supporters' and carers have been used to help potential participants come to a decision, to offer a view as to the user's capacity, or to verify or co-sign consent (Boyden et al. 2009, Nind 2009, Ulivi et al. 2009, Young and Chesson 2008). However, there is a lack of guidance about how they should be involved (McKeown et al. 2010). It is argued that verbal consent is acceptable when combined with a witness's signature or mark of consent (Wilson et al. 2010). A translator, speech and language therapist, and/or someone who is familiar with the individual's communication may need to be present, or a researcher skilled in using individualised and alternative communication techniques and in recognising how a person says 'yes' or 'no' (Wilson et al. 2010, Cambridge and Forrester-Jones 2003, Cameron and Murphy 2007, Donnelly 2004). For people with intellectual disabilities, face- to-face

---

[5] It is important to note here that the Mental Capacity Act (2005) was introduced in the middle of the period of the review. As such, earlier papers referred to issues around the use of proxy consent, which has now been replaced by consultee agreement.

explanation is essential to pick up both verbal and non-verbal signals (Cameron and Murphy 2007).

Several papers comment on the importance of considering health or social care professionals' knowledge and practice in supporting and respecting the potential participants' right to choose, arguing that some further education or training for professionals may be necessary (Cambridge and Forrester-Jones 2003, Cameron and Murphy 2007, Nind 2009). Some researchers have additionally sought agreement of a relative or carer, even where the participant had given informed consent. Similarly, allowance was made for the possibility of participants providing assent rather than consent (Oliver-Africano et al. 2010). Proxy consent or consultee agreement for those with severe intellectual disabilities has been regarded by some commentators as a necessary compromise (Aldridge 2007, Nind 2009), while the agreement by family members or staff has sometimes been used in place of consent or assent by older care home residents (McKee et al. 2002, Whelan et al. 2009).

Data collection

There were a number of barriers and facilitators found relating to how data was collected. Some of these applied across different research designs and methodologies, and some were specific to particular data collection methods.

The focus of the research literature was primarily on involving people with intellectual disabilities. Most commonly, the barriers related to the use of questionnaires, surveys, interviews and focus groups to ascertain participants' views.

Communication difficulties experienced by participants are one of the most important considerations in designing data collection methods. Problems in communication may relate to difficulties in cognitive, memory and language use (Lloyd et al. 2006, Ross and Oliver 2003, Tuffrey-Wijne et al. 2008, Wilson et al. 2010, Fraser and Fraser 2001). Lloyd et al. (2006), in their study of people with expressive language difficulties, highlighted a lack of insight or awareness, meaningless responses, poor temporal orientation, disordered speech patterns, dwindling vocabulary and a lack of focus, as issues related to communication difficulties. Some people, particularly those with intellectual disabilities, may have trouble understanding the concept of research or abstract conceptualisations (Tuffrey-Wijne et al. 2008, Young and Chesson 2008). Perceived or actual difficulties in understanding can lead to research involving only the more articulate or to a reliance on staff to interpret the experiences of people with intellectual disability, when people themselves are competent social actors (Lloyd et al. 2006, Nind 2009).

A further area relating to communication and data collection with people with cognitive disabilities is acquiescence, an issue that has long been recognised. Cambridge and Forrester-Jones (2003) reported that acquiescence was a key issue for people with more severe levels of intellectual disability, even where they did have some communication skills. The role others, including researchers, supporters, or proxies, play in interpreting,

translating or responding on behalf of participants, can obstruct the involvement of people with severe intellectual disability in research. Antaki et al. (2002) used conversation analysis to explore how care staff delivered a questionnaire-based interview to residents with intellectual disabilities. They found that deviations were made from the questionnaire script, largely because the interviewers were trying to be helpful. These deviations influenced responses and, in some cases, answers were constructed by the care staff.

One issue that was identified as going across almost all research methods used in the studies was that of time. In addition to time needed to recruit people, time was also an important factor in data collection. For people with intellectual disabilities in particular, time is an essential factor in facilitating involvement: providing the space necessary for people to express themselves and to provide on-going support throughout the process (Cambridge and Forrester-Jones 2003, Fraser and Fraser 2001, Kaehne and O'Connell 2010, Tuffrey-Wijne and Butler 2010). The need for face-to-face support in survey research again means more time needed (Finlay and Lyons 2001). The methods used can create additional time pressures. For example, observation can be an effective method to include people with intellectual disabilities, but involves considerable time.

People can experience fatigue when completing surveys and interviews and so need regular breaks, therefore extending the time taken to collect the data. It can take longer to find out information from people with cognitive and communication difficulties due to the need to ask questions more than once and perhaps in different formats, as does allowing people the time to formulate and express their views (Davies et al. 2010). Sustaining involvement in research can be a challenge (Lloyd et al. 2008, Woodall et al. 2011). Tension may arise between the additional time needed to conduct the research (Davies et al. 2010) and maintaining the interest of participants (Fenge 2010).

Barriers in surveys and questionnaires

Surveys and questionnaires hold specific challenges for some groups. For example, only ten out of sixty residents of care homes were able to complete the Schedule for the Evaluation of Individualised Quality of Life-Direct Weighting (SEIQOL-DW) in a study on older people (McKee et al. 2002). Questions around time, quantitative judgements, direct comparison questions, abstract concepts and generalised judgements can be challenging for people with cognitive impairments, including people with intellectual disabilities and people with dementia. Other problems with questionnaires include complex, negatively phrased questions, acquiescence, multiple choice, irrelevant questions and validity issues. It has been suggested that people with intellectual disabilities are too heterogeneous in terms of personal history, expressive and receptive language and cognitive ability for a single questionnaire to be valid for the whole population (Finlay and Lyons 2002).

Structured questionnaires have been successfully used with some seldom-heard populations. Nevertheless, there are caveats in their application. Perhaps the greatest concern is that researchers simply transfer instruments validated in the general population

to seldom-heard groups. For example, a study that pilot-tested the psychometric properties of the Quality of Life Assessment Schedule (QOLAS) with dementia patients highlighted a number of population-specific issues: participants with dementia needed appropriate prompting and have the scoring options for each construct repeated; there was a tendency for users to respond using 'words' (e.g., slight problem) rather than the corresponding number; use of an overall visual analogue score (VAS) limited responses as users and carers wished to separately indicate physical as well as mental health (Selai et al. 2001). In another study, the only appropriate way to fully involve older people with physical disabilities was to change the mode of administration of a particular tool (SF-36), from postal self-completion to face-to-face interviews (Seymour et al. 2001).

Barriers in interviews and focus groups

Some of these problems also occur with face-to-face interviewing. Complex questions, subject-object questions, and open questions have all been shown to make understanding and responding more difficult for people with intellectual disabilities (Finlay and Lyons 2002, Gilbert 2004, McNally 2002). In their meta-synthesis of qualitative interview research with people with expressive language difficulties (e.g. people with brain injury, dementia, intellectual disability etc.), Lloyd et al. (2006) highlight a limited understanding of complex grammatical phrases and abstract concepts, together with a lack of insight, awareness, meaningful response, poor temporal orientation and acquiescence. Inarticulateness (linked to low self-esteem as well as language skills) can lead to limited responsiveness in interviews. It has been argued that people with severe intellectual disabilities may only be able to provide reactions, rather than views (Nind 2009).

The use of focus groups brings its own set of difficulties (Nind 2009). The cognitive and emotional demands of reflecting on other people's arguments and engaging opposing views can be particularly challenging for people with intellectual disabilities (Kaehne and O'Connell 2010). These authors note that focus groups with people with intellectual disabilities can resemble one-to-one interviews in practice, and may not achieve the aims of a focus group. In addition, issues of compliance, censoring, conformity and contamination arise where supporters are also attending the focus group (Llewellyn 2009). Some authors conclude that focus groups are not a suitable method for some people with intellectual disabilities who may have additional sensory and communication needs (Barr et al. 2003, Fraser and Fraser 2001).

Facilitators for surveys and questionnaires

For people with intellectual disabilities, face-to-face support was often found to be essential, and it has been proposed that any 'tick-box' administration should be supported by digitally recording the interaction to ensure nuances can be captured (Fang et al. 2011, Finlay and Lyons 2001, 2002, Pawson et al. 2005, Perry and Felce 2002). Questions often need to be simplified (e.g. Likert-type scales reduced), and made visual (e.g., smiley faces) while the use of different coloured stickers can help participants identify priorities (Finlay

and Lyons 2001, Nind 2009, Schmidt et al. 2010, Young and Chesson 2006, Gordon et al. 2007). Show-cards were found to be helpful to illustrate topics covered through open questions (Mindham and Espie 2003). Throughout the administration of any standardised tool, it was recommended that validity of responses should be tested through: repetition of difficult questions; use of reverse wording and nonsense questions to test for acquiescence; and notating any conflicting information for later discussion (Finlay and Lyons 2001).

In terms of postal questionnaires, a meta-analysis found the numbers of returned questionnaires can be improved through direct personalised mailing: addressing the individual by name (e.g., Mrs Smith) rather than by any generic appellation (e.g., Dear Participant). Adding hand-written signatures of the researchers can further increase responses. The size of the impact of this strategy on the proportion of questionnaires returned is predicted to be between four and 10 per cent, depending on the baseline response proportion when using neither intervention (Scott and Edwards 2006). Response rates among South Asians were also found to be increased through the pragmatic (yet expensive and time-consuming) technique of an interviewer visiting those individuals who did not respond to the postal questionnaire (Allison et al. 2003). Nevertheless, it remains unknown whether more than one postal reminder or a third mailing of a questionnaire improves the response rate for self-completion questionnaires with seldom-heard groups, as there was no analysis around the effectiveness of this method to increase response rates in the articles reviewed.

Facilitators for interviews and focus groups

Recommendations for involving individuals with intellectual disabilities in interviews and focus groups emphasise the importance of avoiding complex or abstract topics and conceptual or time-orientated questions (Lloyd et al. 2006, Murphy and Cameron 2008, Nind 2009). Open discussion is unlikely to be effective, and verbal questions need to be asked in a direct style, with clear sentence structure and ordered to progressively focus down on any issue (Gilbert 2004, Lloyd et al. 2006, Nind 2009). Nevertheless, direct questioning was not found to be suitable for all groups. Some people with mild or moderate dementia are said to find discussion, rather than questions, less confusing or worrying (McKeown et al. 2010). Preparation for focus groups and interviews was found to be important. Fraser and Fraser (2001) advise that moderators of focus groups should come prepared to communicate with participants with speech and language problems, as well as prepared to deal with potentially dominant participants and those with repetitive speech.

A variety of techniques have been developed to support communication with people with intellectual disabilities during interviews (Nind 2009). These include using:

- A combination of written, oral and visual stimulus including pictures, photographs and drawings (Mathers 2008, Nind 2009, Pawson et al. 2005, Young and Chesson 2008);

- Activities where participants produce craft, tell social stories, take photographs, cut up pictures from magazines or use drawings and photos to produce large-scale canvas collages (Aldridge 2007, Gates and Waight 2007, Gilbert 2004, Gosden and Kirkland 2008, Mathers 2008, Nind 2009, Pawson et al. 2005, Young and Chesson 2008);
- Digital slideshows of photographs to elicit interview talk and prompt discussion (Mathers 2008);
- Talking Mats – a technique developed to support people without speech, allowing comparison of ideas through the use of pictures and symbols (Brewster 2004, Nind 2009, Murphy et al. 2005). It is recommended that a series of Talking Mat interviews are carried out, each created immediately after a communication exchange to promote greater immediacy and salience, reduce the level of abstraction, and develop a cumulative picture that should be more representative of participants' views (Brewster 2004).

In some studies, other people were used to support communication within an interview or focus group setting. The evidence as to the extent and type of support that seldom-heard groups might welcome was not conclusive. Some commentators argue that ensuring effective participation of people with intellectual disabilities requires an 'interpreter' – a professional or family member – to ensure individuals' views are appropriately conveyed (Pawson et al. 2005). However, such support is perceived by other commentators as 'gatekeeping': negatively influencing people's involvement and silencing views that may be contrary to existing policies (Brewster 2004, Kaehne and O'Connell 2010, Llewellyn 2009, Nind 2009). Some mental health users are clear that they wish to participate on their own behalf in any focus group or interview, despite welcoming friends and relatives to provide a support network throughout the research process (Ulivi et al. 2009).

Contact or meetings with participants before interviews take place is recommended. These can be used to discuss the research process, break down social barriers, confirm attendance, identify any access problems, or ask about any reasons for non-attendance (Andrews 2005, Proctor 2001, Lloyd et al. 2006).

The majority of 'good practice' advice is generic, and commentators recommend: being flexible about the timing and location of data collection; conducting interviews in familiar settings (Barr et al. 2003, Boyden et al. 2009, Young and Chesson 2006); interviewing older people in their own homes to minimise transport barriers (Davies et al. 2010); visiting research participants at a time of day convenient to them (McKeown et al. 2010, Young and Chesson 2006); and preparing alternative ways of asking questions (Boyden et al. 2009, Fraser and Fraser 2001). Nevertheless, some of these practices are noted to have additional benefits for specific populations. For example, the use of familiar settings for focus groups or interviews has been described as having the advantage of minimising the potential distraction of a novel environment for people with intellectual disabilities (Nind 2009).

Duration and number of interviews

Shorter interviews or focus group sessions are recommended for research involving people with intellectual disabilities, the oldest old (people aged 85 plus) and people with dementia (Cambridge and Forrester-Jones 2003, Cambridge and McCarthy 2001, Davies et al. 2010, Proctor 2001). Collection of baseline data from mental health service users has been split over two visits to minimise burden (Abbott et al. 2005), and repeated interviews with older people or people with intellectual disabilities are recommended to help build up information (Brewster 2004, Gilbert 2004, Davies et al. 2010), develop relationships (Jones 2008) and enable participants with to express themselves and respond to individual questions (Kaehne and O'Connell 2010, Nind 2009).

Interviewer characteristics and group composition

Some researchers have sought to use interviewers or focus group facilitators from a similar background to that of participants. One study allowed participants to choose the language spoken and gender and ethnicity of the interviewer (Rugkasa and Canvin 2011). Other commentators argue that it is the ability to speak to people in their own language – either by the researcher or through the use of an interpreter – that is important, rather than the matching of ethnic background or gender (Rooney et al. 2011). Perry and Felce (2004) found that people with intellectual disabilities can be trained and supported to be involved in data collection using face-to-face questionnaires without response bias. Cambridge and McCarthy (2001) used one male and one female facilitator in 'mixed' focus groups with people with intellectual disabilities to ensure participants did not feel marginalised or intimidated by the gender of the facilitator. In a qualitative study involving South Asians with asthma, the need to consider the group composition of focus groups in terms of gender segregation was important (Sheikh et al. 2009).

Proxy respondents

One research adaptation that has been used to assess the experiences of people with communication difficulties, including those with intellectual disabilities and those with dementia, has been the use of proxy respondents. From the articles reviewed here drawing on work published in the last ten years, it is clear that the views about who is 'uninterviewable' have changed, particularly in relation to the types of people who are included in qualitative research. However, challenges remain and there are still people who are considered 'unable' or 'unsuitable' to take part in research, or for whom methods have not yet been developed to support engagement. For example, although Young and Chesson (2006) carefully selected tools and approaches to support the inclusion of people with intellectual disabilities, they could not support the involvement of people with severe mental health problems and challenging behaviour. Similarly, Hancock et al. (2003) reported that out of a sample of 101 older people with mental health issues, 14 could not be interviewed because of severe dementia, chronic schizophrenia or severe depression.

People with severe dementia have also been excluded from research seeking subjective views through the use of standard self-report quantitative measures (Hoe et al. 2007).

Quantitative health and social care research often seeks 'proxy' information for seldom-heard groups from various sources. Typically, family carers, clinicians or social care staff – such as care workers or managers – are used to gather information about particular groups, including people with dementia, autism and mental health issues (Beadle-Brown et al. 2009, Bryan et al. 2005, Hancock et al. 2003, Hoe et al. 2007, McKee et al. 2002, Schmidt et al. 2010). The use of proxies raises the question of whether the proxy-generated information can be regarded as 'equivalent' to or in agreement with the viewpoint or 'self-ratings' of patients or service users, or should be treated as an alternative perspective. It remains unclear how much concordance there is between proxies and those they care for, though some evidence suggests sufficient overall correlation to support the use of proxies for people with severe intellectual disabilities, and people with aphasia.

Carers of people with dementia have been found to have different perceptions of the quality of life of people they care for, and it is recommended that researchers should treat them as offering different viewpoints rather than a substitute (Hoe et al. 2007). Selai et al. (2001) asked carers to think about how they (i.e., the carer) perceived the quality of life of the patient with dementia, rather than how they thought the patient perceived their own quality of life. However, comparisons between patients' ratings with those given by carers have produced considerable overlap and agreement (Selai 2001, Hilari et al. 2007). Among people with severe intellectual disabilities and autism, proxy and self-ratings have been found to be significantly correlated (Beadle-Brown et al. 2009). Key-workers' ratings of depression in adults with mild intellectual disabilities have been shown to be reliable, temporally stable, and have a high degree of agreement with self-ratings (Gordon et al. 2007).

Not all studies reviewed, however, produced evidence that positively supported the use of proxies to collect information about people with intellectual disabilities. For example, a study comparing staff proxy concordance with the scores of people with intellectual disabilities who were supported to complete a structured interview tool found that ratings by people with intellectual disability were not significantly correlated with staff scores (Perry and Felce 2002).

In relation to the quality of life of people with dementia, a comparison of caregivers and self-report information[6] found that carers rated the cared-for person's quality of life lower than the cared-for person did (Hoe et al. 2007). The Pleasant Events Schedule – Alzheimer Disease (PES-AD) has been found to be an appropriate and reliable proxy measure when

---

[6] Using the Quality of Life in Alzheimer's Disease Scale (QOL -AD), the Mini Mental Health State Examination (MMSE), the Cornell Scale for Depression in Dementia, Neuropsychiatric Inventory (NPI) for neuropsychiatric symptoms of AD, the AD Co-operative Study - Activities of Daily Living Inventory (ADCS -ADL) and the Hospital Anxiety and Depression Scale (HADS).

used by care home staff to measure the quality of life of residents in one nursing home, but not the Apparent Emotion Rating Scale (McKee et al. 2002).

Some work compares responses of different proxies, both in terms of how they each compare with other measures and how far each agrees with patient self-report. For example, a study of the construct validity of clinicians' and caregivers' 'proxy' reports of the health-related quality of life (as measured through EQ-5D) of patients with dementia concluded that, while there were some differences, both provided valid sources of information in terms of being associated with clinical measures of disease severity (Bryan et al. 2005). Research focusing on agreement between care-givers and older people with mental health problems reported general agreement between the number of needs reported by people themselves and by staff and family carers (Hancock et al. 2003). The only user group where some differences were found was for people with dementia, who reported fewer needs than their proxies reported. In contrast, Coucill et al. (2001) found that agreement on health-related quality of between older people and proxies did not differ by severity of dementia.

The most important factor in ensuring that there is proxy and participant agreement, or that the proxy respondent gives a reliable account of the experiences of the person they support, is the relationship or understanding between the proxy respondent and the participant (Coucill et al. 2001, Gordon et al. 2007, Schmidt et al. 2010). The evidence provided as to 'who' should act as a proxy respondent supports the use of a range of individuals: informal carers, keyworkers and clinicians as well as wider friends.

The use of proxies can sometimes also create barriers to engagement. McKee et al. (2002) found that proxies were unwilling to rate the emotional state of participants, even with behavioural guidance in the instrument. Questions are also raised over the knowledge and understanding proxies have in order to respond on the part of participant (McKeown et al. 2010). The magnitude of difference between proxy and person with intellectual disabilities can be influenced by cultural differences, as well as the degree to which the proxy knows the person with intellectual disabilities (close and regular contact) and the severity of disability (Schmidt et al. 2010). Proxies may find it difficult to divest themselves of their own views (Nind 2009). Perry and Felce (2002), on their study of quality of life of people with intellectual disabilities, found no rationale for using staff as proxy respondents on subjective issues.

Recommendations around strengthening quality of life information collected from proxies include the use of 'proxy' information from more than one source (Bryan et al. 2005), and the collection of objective measures in addition to subjective measures (Beadle-Brown et al. 2009).

Clearly, the issue of using proxies is far from straightforward. The literature on the use of proxy respondents has been analysed in more detail elsewhere (Smith and Malley 2012).

Observation

Observation is an alternative way of collecting information about the experiences of people when data cannot be collected directly from them or through proxies. It has been used effectively to evaluate the quality of life and wellbeing of older people with dementia as well as people with intellectual disabilities (Gilbert 2004, Jones 2008, Mathers 2008, McKee et al. 2002). Observation can be supported by instrumentation but is, however, labour-intensive (Tuffrey-Wijne et al. 2010). For example, the Dementia Care Mapping observation tool and the Mood, Interest and Pleasure Questionnaire (MIPQ) have been found to be appropriate and reliable for collecting quality of life information or for measuring affect (McKee et al. 2002, Ross and Oliver 2003). Research on observation is considered in more detail elsewhere (Mansell 2011).

Analysis and interpretation

Fewer difficulties were reported relating to analysis and dissemination. Validity was raised as a problem as researcher assumptions can feed into interpretations and analysis of qualitative findings, but this is a challenge within qualitative research more generally (Lloyd et al. 2006). Arguably, there is potentially a greater requirement for researcher interpretation when people have high support needs. There is a tension between a flexible study design to enable the participation of particular groups and maintaining sufficient academic rigour to ensure validity. Accessible outputs that include pictures rather than text or simplified language are difficult to place in established peer-reviewed journals (Nind 2009, Tuffrey-Wijne and Butler 2010).

Where there were discussions relating to analysis and dissemination, recommendations focused on appropriate research practices. The use of triangulation of multiple sources of evidence, such as joint interviews with carers, or cross-referencing to sources of information such as daily logs and plans, was recommended by Gilbert (2004), as this can help to validate participants' responses. However, Gilbert also warned against the level of interpretation required in doing this. It was also suggested that any analysis should involve a reflexive and rational method to ensure the consideration of issues of power and allow robust inter-rater reliability (Murphy and Cameron 2008, Proctor 2001).

Recommendations as to effective dissemination to seldom-heard groups emphasises the necessity of on-going individual and community engagement after data collection (Rooney et al. 2011) and the provision of imaginative and alternative formats. For example, multi-media presentations that include sound, pictures, signing and textured representations are recommended for people with visual impairment (Duckett and Pratt 2001). Specific recommendations for people with intellectual disabilities include the provision of 'plain facts', use of audio recordings, community feedback reports, workshops, leaflets, photographs of individual Talking Mats and media coverage (Brown and Scullion 2010, Cameron and Murphy 2007). One study organised a public exhibition of the work produced

by the research with people with intellectual disabilities that had the advantage of providing feedback and marking the end of the project in a celebratory way (Mathers 2008).

Discussion

The evidence to answer the three research questions was variable. In this section we discuss some of the key findings, highlight the gaps and inconsistencies in the literature and reflect on issues of validity.

What do we know about whose views and experiences are excluded from research and how often does this happens in health and social care research?

The research reviewed predominantly focused on four categories as being difficult to reach or engage in research: intellectual disability, older adults, in particular those with dementia, mental health conditions and minority ethnic groups. Almost half the papers reviewed focused on those with intellectual disabilities. There were a number of groups on which no research was found: for example, self-funders, homeless groups, lesbian, bisexual, gay or transgender groups. None of the papers looked at the prevalence of the exclusion of these or other groups. This is largely because it is difficult to know which groups of people were not reached by recruitment strategies, and there are ethical and practical issues around trying to follow-up people who may have been reached but who did not respond to being recruited. Studies which included, for example, people with intellectual disabilities or older adults did not necessarily consider how representative their sample was in terms of other risk factors for exclusion such as ethnicity.

Why are some people's views and experiences not represented in some research related to health and social care?

There were many reasons found why the views and experiences of certain groups may not always be represented in research on health and social care of people with long-term conditions. We have summarised the reasons why the views and experiences of certain groups may not be represented in research under three broad headings relating to: assumptions; definition and recruitment of these groups; and population characteristics.

Assumptions

A key barrier to inclusion was the assumptions made by various people (researchers, gatekeepers, ethics committees, funders and so on) involved at each stage of the research process, including research design, ethics and recruitment. These assumptions fed into who was considered a competent research participant and who was not. These assumptions are embedded in what 'conventional' research practice looks like, and often in the practices of ethics committees. These assumptions may shift as the effects of the Mental Capacity Act, which argues that capacity should be assumed until proved otherwise, filter through.

Defining and recruiting seldom-heard groups

There were many issues raised with regards to reaching those who were seldom heard. Among those who accessed health and social care services, finding relevant participants was easier, and barriers usually came later in the research pathway. However, under-use of services by those from ethnic minorities and those with milder levels of intellectual or physical disabilities living with their families mean that these groups are often harder to identify and contact, as are those who fund their own social care, those who are homeless and those from travelling communities.

One disadvantage that was found in going through services in order to recruit participants is that professionals, community groups or service providers can act as gatekeepers and make decisions about whether to involve particular people, or can either wittingly or unwittingly sabotage the recruitment process by not communicating the research to those they represent.

The sampling strategy chosen can also impact on recruitment: often a convenience sample was used which meant that samples were not necessarily representative of the population and could also result in some samples being over-researched as they were the easiest to contact.

Characteristics of specific populations

Our particular interest is the inclusion of seldom-heard groups in the evidence base on people with long-term conditions. There were three core characteristics which appeared to impact on participation in research whether at the recruitment stage, at consent stage or during the data collection stage. Where people had cognitive impairments or difficulties with communication, the challenges of successful recruitment and involvement were substantially greater. In addition, language and cultural differences had an impact. Finally, the presence of physical or sensory disabilities, along with the specific characteristics of particular long-term conditions, could play a role not only in whether or not people with long-term conditions agreed to take part in research but also in whether they remained part of the sample during the data collection phase.

What methods are there for facilitating people's views to be heard?

The involvement of stake holders

Including stakeholders in the design process and in the testing of measures, letters, interview schedules is strongly recommended. Involvement of a user reference group can operate as a counter-balance to researchers' assumptions which can constrain involvement of seldom-heard populations. The review provided little guidance as to the 'best structure' or model of involvement which, whatever model is used, has time and resource implications. However, outside of the literature reviews there is guidance and advice available through organisations such as Involve (www.invo.org.uk).

Allowing additional time

Additional time across the research pathway is needed to facilitate inclusion. Of the 83 papers extracted for this review, authors of 54 papers highlighted the need for more time to be allocated to the research process. Little specific guidance is provided about time-frames, but this is not surprising given the different foci of the papers (the different populations and outcomes sought), as well as the different skill levels of the researchers involved. There was no overt discussion about how researchers could work effectively with research funding or commissioning bodies in setting-up appropriate time-frames around research projects. The majority of commissioned research has a time-limit of between two and three years. Ensuring the inclusion of seldom-heard communities could take a considerable portion of that time.

Flexibility

Flexibility is needed, again across the research pathway: flexibility in recruitment strategies, methods used and the way in which these methods are applied. Triangulation of methods is a key strategy. For those individuals with severe cognitive or communication difficulties, such data gathering should include a combination of different types of communication aids and stimuli to promote participation: such as using drawings, photo and objects, not just verbal questions and show-cards.

The use of proxies (where necessary)

The use of proxy respondents is sometimes necessary to ensure the inclusion of some people. There were differing conclusions as to the effectiveness of this approach, but the narrative synthesis suggested that there were some existing scales that showed good concordance between proxies and participants. Slight shifts in practice, such as asking the proxy respondent to think about how they perceived the quality of life of the participant, rather than how they think the patient perceives their own quality of life, is recommended. The relationship – or understanding – between the proxy respondent and the participant is important. They should be in regular contact.

Limitations of the evidence base

It is clear that there are no clear answers to the challenges of including seldom-heard populations in research. The recommendations for facilitating the involvement of seldom-heard groups appear somewhat prosaic, often focused more towards generic good research practice than the identification of effective and innovative methods, techniques and processes for particular groups of people. Few clear recommendations emerged, and in many cases the research from the last decade does not appear to have substantially added to the knowledge from earlier research, especially related to involving people with intellectual disabilities or other cognitive impairments.

Gaps in the knowledge base

The mapping and narrative synthesis of the papers indicated a lack of evidence in three core areas: non-participation or non-response, cost-effectiveness and guidance or information on a number of seldom-heard population groups.

Non-participation and non-response

The validity of reported outcomes from standardised questionnaires and interviews depends on two factors: that the administration of the data collection encompasses all relevant individuals, and that there is limited missing data across the different questions and scales. In this review, we explored the former rather than latter question. As discussed, many of the extracted papers discussed relevant techniques around recruitment, but surprisingly few papers explicitly presented the numbers and characteristics of individuals that did not respond to any invitation, refused participation at recruitment, or dropped out of any study; nor any rationale that resulted in fewer participants. It is not clear from the UK literature reviewed why such core data remains underreported. It could be argued that one reason is publication bias (Foxcroft and Smith 2008, McGauran et al. 2010, Siddiqi 2011). For example, a systematic review that explored study publication and outcome reporting bias found strong evidence of association between significant results and publication (Dwan et al. 2008). Where a study has high levels of refusals, drop-outs or general non-response, outcomes may well be equivocal and thus the likelihood of publication lowers. Alternatively, such a lack of reporting could be as a result of our own review bias, our inclusion criteria (e.g., UK only) and search terms. The focus of this review included only outputs in peer-reviewed journals, while the breadth of this review precluded exploring wider than the UK (these further areas of enquiry will be discussed in the recommendations). Nevertheless, our search terms were comprehensive and included words such as 'participation', 'response', 'sample' and 'selection bias' to allow for appropriate identification and extraction.

The lack of reporting of non-participation and 'drop-out rates may also relate to research ethics guiding the actions of researchers or a lack of time and funding to do so (O'Reilly et al. 2009, Ramcharan and Cutcliffe 2001, Stalker et al. 2004). Ethics committees micro-manage the submitted structure and process of many research projects, dictating the process of recruitment (opt-in rather than opt-out), the structure and content of consent documentation and the number of reminders allowed to be sent out. There is often a blanket ban on following up the rationale behind non-participation. This is a contentious area; individuals should not be harassed nor called to account for their non-participation (Garrard and Dawson 2005). Nevertheless, unless we begin to fully understand the rationale behind non-participation it will be difficult to ensure full inclusion in research.

Cost-effectiveness

Appropriate engagement requires greater resources. Skilled and experienced researchers are necessary to develop and apply appropriate methods; additional staff to support

participants (before and during the research), and a more flexible time-frame is needed to conduct and disseminate the research. Additional costs include translators and interpreters, incentives, transport or childcare provision (McKeown et al. 2010, McKee et al. 2002, Cambridge and Forrester-Jones 2003, Davies et al. 2010, Fenge 2010, Lloyd et al. 2008, Young and Chesson 2008). Only two papers provided, or attempted to provide, some idea of the cost of carrying out research with marginalised groups. One paper concentrated on the impact of recruitment, arguing that owing to the multiple recruitment techniques necessary, the cost per recruited participant was £11,000 (Oliver-Africano et al. 2010); while the second estimated that trial research in care homes would cost approximately three times the amount of running a similar trial with older people in their own homes (Zermansky et al. 2007).

Seldom-heard populations

In shaping our review, the search terms included a range of groups or populations for whom there was some prior evidence (either empirical or anecdotal) of exclusion. The majority of papers reported on working with people with intellectual disabilities or other cognitive disabilities, such as those with dementia or frail older people. There was less evidence about other groups which may not all necessarily be hard to include but in practice were not explicitly identified, including lesbian, bisexual, gay or transgender groups, black and minority ethnic groups, travellers, survivors of abuse, and those individuals facing severe economic and social deprivation. Only six papers focused on people with severe mental health problems. There was very little research focusing on physical and sensory disabilities. There was also a lack of engagement with the multi-faceted nature of people who straddled different groups, such as ethnic minority and intellectual disability. There was also very little research related to people who fund their own health and social care and no guidance as to how to reach these people.

Limitations of the review

A limitation to the review is the exclusion of the international literature that has explored the research participation of seldom-heard groups (e.g. Pinto et al. 2007, Skaff et al. 2002, Williams et al. 2001) and techniques around proxy response and non-response (e.g. Coucill et al. 2001, Rogler et al. 2001). The inclusion of only UK papers may mean that the findings and conclusions may not transfer to other policy and practice contexts. However, a brief review of some of the papers excluded illustrate similar issues and themes in research in the USA, Australia and the Netherlands (McDonald et al. 2009, Iacono 2006, Dalemans et al. 2009, Lennox et al. 2005, Evenhuis et al. 2004), as well as reported in a review of the international literature by Cleaver et al. (2010). Key barriers that emerged from these papers were gaining consent and a lack of time. Similarities with our findings were apparent in the papers reviewed.

A further limitation of this review is the relatively narrow focus of UK studies on a small number of seldom-heard groups. Finally, the quality of the studies included in this review

was variable. Of the 83 papers, only 23 were 'scored' as robust. As with much research in health and social care, future evaluation needs to concentrate on appropriately measuring (rather than merely assessing) involvement of seldom-heard groups across the research process.

Implications and recommendations for policy and research practice on long-term conditions

For the purposes of the QORU research agenda, we are concerned with health and social care of people with long-term conditions. Although, this review focused somewhat more widely than just long-term conditions and included some groups without long-term conditions, the findings from this wider analysis apply to those with long-term conditions. These findings reinforce the argument that most research underpinning the evidence-base of policy and practice tends to favour easily-accessible groups and those that are (at the very least) able to understand standardised interviews and communicate their thoughts and wishes (Linehan et al. 2009). If we are to appropriately identify those for whom different forms of health and social care based practice is most suitable, there must be a move from institutional exclusion to early inclusion of such groups in any research process. The idiopathic nature of seldom-heard groups requires concurrent and multiple recruitment and data collection techniques, robust but adapted data collection, and innovative multi-method analysis. Increasing the time and resources available for recruitment and for data collection where triangulation of information is needed will affect the cost of the research, the time-frame of the research, possibly changes in the ethical review process, and the focus of researchers themselves.

The necessary flexibility has implications for policy and practice on collecting information about people's experiences of and outcomes from receiving health and social care. A variety of standardised tools are currently being used to collect data on health and social care outcomes in the UK (Department of Health 2011): Patient Reported Outcome Measures (PROMS) of specific conditions or procedures; the EQ-5D global measure of health-related quality of life; and the Adult Social Care Outcomes Toolkit (ASCOT) measure of social care-related quality of life. PROMs are currently completed for only four procedures at present (hip and knee replacements, groin hernia and varicose veins), although there are plans to extend this. Most, if not all of the seldom-heard groups identified by the review would have difficulties in completing such instruments, although no research exists to show how such measures might be made more accessible to groups who have traditionally not been included in previous research.

The EQ-5D could also not be self-completed by many of the populations we explored and, although available in proxy form, further research is needed to recognise that multiple actors will need to complete the same measure (formal and informal carers, clinicians, relatives) to ensure that the functional and physical status as well as the psychological and social health is correctly recorded (Bryan et al. 2005). Such adopted tools could be used for

some of the populations explored in this review. Nevertheless, if outcomes are to be appropriately collected and all views represented rather than excluded, there will need to be a multi-level approach: melding and cross-referencing visual, biographical and story-telling methods with the more standardised outcomes.

Another issue with EQ-5D is that some key outcomes of services for long-term conditions are not assessed, such as confidence and ability in managing symptoms, medication and lifestyle or adjustment, self-esteem, sense of stigma and so on. In addition, some EQ-5D items which are very important for those with long-term conditions are collapsed and simplified in a way that is not helpful for this group. Currently, the possibility of developing a generic PROM for long-term conditions that takes account of these weaknesses is being considered by QORU. However, the development of such a measure of outcome will need to take into account the barriers and facilitators found in this review.

ASCOT can be self-completed, but it is more applicable to marginalised groups though the use of an observation tool (ASCOT CH3, http://www.pssru.ac.uk/ascot/) as well as a four-level face-to-face interview schedule. The former, although currently limited to residential care homes, provides the opportunity to gather the experiences of people with cognitive and communication impairments, while the latter, although too complex for many of the marginalised groups highlighted within this review (Gordon et al. 2007), provides a means to collect proxy data. However more research is needed to clarify the validity and reliability of each element of the ASCOT tool and to provide guidance as to how best to choose and weight proxy responses. The Adult Social Care survey (http://www.ic.nhs.uk/services/social-care/social-care-collections/user-surveys/user-survey-guidance-2010-11) combines some elements of EQ-5D with ASCOT to measure outcomes and experiences of those receiving adult social care. A version for people with learning disabilities was developed and tested, but to be accessible to people with communication difficulties required fewer response options and simpler phrasing which in some cases changed the direct comparability between the two versions of the survey. In addition, it is not permitted to change the EQ-5D questions and so it was not possible to make these questions accessible for people with communication difficulties.

This review was both a response to some of the work described above, but also designed to inform the development of the field of measuring outcomes in health and social care. Overall, it found that there were very few concrete consistent recommendations for how to involve seldom-heard groups of people with long-term conditions in research. The majority of recommendations relate to what might be considered good practice in general, but which appear to be more important for those in the seldom-heard groups explored here. There were a few strategies which appeared to improve recruitment and then to improve participation of people once they had been recruited. Although these findings related to research, they also relate to any form of consultation.

- Researchers need to be very aware of their own assumptions about who can take part in research and design their research to be inclusive, rather than excluding people who might need more time or support to take part.

- Researchers should involve user reference groups or individual user consultants in designing the study, including deciding on recruitment strategies, on the methods of data collection, and on the adaptations needed to measures in order to involve as many people as possible. Asking users to comment on information sheets, for example, can be a useful and very simple way to ensure that they are understandable and will encourage people to take part. In the UK, the Department of Health has been leading the way with the Public Patient Involvement initiative, and most research grant-awarding bodies want to know how users have been or will be involved in designing and monitoring research. However, more could be done to ensure that research is inclusive of people from the groups focused on here.

- When working with people from seldom-heard groups, more time and resources should be allowed in order to recruit people to studies: having the flexibility to have a face-to-face meeting with prospective participants to provide further information, develop rapport and answer questions. This might be individually or through existing groups and meetings, although the latter on its own might limit recruitment to those who are able and motivated to attend meetings/social or community groups etc.

- Using more than one recruitment strategy appears to be needed in order to ensure that as many people as possible know about the research and are in a position to consent to taking part.

- Information materials should be clear and accessible to the target population: e.g. available in translated form if needed, or in easy-read version, with photos or symbols etc. However, flexibility is needed so that materials can be individualised, especially in the case of people with communication difficulties, by those who might be helping them to understand the research and decide whether to take part. Clear information about the benefits of participation is needed for those who are acting as personal or nominated consultees.

- More time and resources are also needed during the course of the data collection: for example, being able to do an interview over several shorter sessions within the person's own home or other familiar environment may be more likely to elicit a positive response to an invitation to participate than being asked to attend a two-hour focus group in an unfamiliar environment which they might require transport to reach. .

- Researchers and others involved in gathering data need the skills and knowledge to interview or conduct focus groups with those some seldom-heard groups, especially those with cognitive, communication or cultural differences. Training in alternative forms of communication or cultural sensitivities may be needed.

- Triangulation of data collection methods is often needed, especially where it is not possible to get a self-report measure, or the reliability of people's self-report might

be in question due to cognitive or communication impairments. This could involve combining, for example, a self-report measure with a perspective taken from someone who knows the person well withobservation of what happens to the person or information taken from records.. Observation can be critical to getting reliable information about the lived experience of people with severe and profound intellectual disabilities.

- When using proxy respondents, asking them their own views about an individual's experiences is easier for them to answer than asking them what the individual would say if they were filling in the survey or responding to the interview themselves.

- When working with different groups of people, including different seldom-heard groups, it may be necessary to use different ways of obtaining the same information in each group. Surveys often have to be simplified and combined with pictures or other media to allow those with communication or cognitive impairments to understand them. The question format might need to be the same – e.g. three or five response options rather than seven. This can cause difficulty in ensuring that the same data is actually being collected in different groups if it is important that the information is all processed and analysed together. So when different versions of the same thing (e.g. translated, simpler language, interview rather than survey etc) are used, researchers should make the effort to map across the measures to establish reliability. For example, how closely related are the results from self-report measures, observation, proxy measures etc. Many of the existing measures as noted above require validation and need to be tested and mapped across different populations.

- Researchers should report more about the characteristics of people who participate so that it is possible to know whether those from seldom-heard populations are included and then to consider how representative their sample is in terms of the whole population studied.

- Although there is currently a lack of research on different groups and the issues for research participation, it is clear that when working with people from ethnic minority groups it is important not to just group people together as one: e.g. BME group. The diversity between different ethnic groups is substantial and can affect the success of recruitment and involvement.

- Ethical approval processes need to recognise the need for and allow flexibility to respond to the individual needs of people taking part, while at the same time balancing the need to protect people's interests. It would also be helpful if there was more acceptance of the possible role proxy respondents/informants might play in ensuring that the experiences of all groups can be represented within research, even when people cannot express their views and opinions directly.

Implications and recommendations for future research

- While systematic data are hard to establish, the group that seem most widely and systematically excluded from the evidence base of health and social care of people with long-term conditions are those with severe cognitive impairments, physical or sensory disabilities that result in communication difficulties, and those with mental health problems. People who belong to several different seldom-heard groups – for example, people with intellectual disabilities and dementia – are particularly at risk of exclusion.

- There has been limited progress in developing methods to systematically include these groups in recent years:

    - More research is needed on the use of proxy respondents, in particular, research that focuses on who are suitable respondents, what type of questions they can reliably respond to, in what type of situations they should be used and how these responses map across to alternative methods, such as observation.

    - Similarly, more research is needed on whether and how to adapt existing measures for particular groups: e.g. EQ-5D, PROMS, ASCOT. There are very few adaptation studies which that look at relationships between the adapted measure and original measure, as well as research validating the relationships of different versions of any measure across different groups.

- There is a need for more research exploring the use of alternative techniques for eliciting responses: e.g. photo elicitation or Talking Mats etc.

    - There is a need for research around capacity and consent issues, such as better ways to assess capacity and who are the most appropriate consultees.

- More research is also needed on a range of other seldom-heard populations, in particular those who are homeless or from travelling communities, and those from a range of different minority ethnic groups.

- The role and attitudes of gatekeepers has also been identified in the literature as an area for further research: for example, GP and patient attitudes to participation in mental health research, or the role of the NHS and voluntary organisations in supporting or hindering research participation.

- Research on the comparative cost implications and effectiveness of recruitment and data collection methods which promote more inclusive research is needed. Allowing more time and multiple methods is resource-intensive, but it is not clear from any of the research how resource intensive it is and what the return on the investment is with respect to more inclusive and representative evidence.

- There is a need for more in-depth information about who does not respond to surveys and why people drop out of research studies. Because of the ethical and often pragmatic difficulties of following up non-response or drop out, this probably needs to be done as part of large-scale surveys and studies.

Appendix

Tables developed from the mapping data

Table 8: Number of papers extracted by year of publication.

Year	Extracted papers n = 83	%
2001	9	9.8
2002	7	7.6
2003	6	6.5
2004	7	7.6
2005	6	6.5
2006	4	4.3
2007	10	10.9
2008	7	7.6
2009	8	8.7
2010	13	14.1
2011	6	6.5

Table 9: Detailed study populations in reviewed papers.

Population	Extracted papers (Number)	Extracted papers (Percentage)
BME	5	6.0
Dementia/older people	16	19
Deprivation	3	3
General population	3	3
LBGT	1	1
LD/PWID	39	47

Population	Extracted papers (Number)	Extracted papers (Percentage)
Mental health	6	7
Travellers	1	12
Victims of abuse	0	0
Other:	9	11
BME and LD/PWID	1	1
BME and mental health	1	1
Chronic aphasia	1	1
Chronic illness	1	1
Huntingdon's disease	1	1
LD/PWID and Mental health/CB	1	1
Older people and mental health	1	1
Visual impairment	1	1
Wheelchair users	1	1
Total	**83**	**100**

Rapid review search terms, databases and extraction table variables.

Table 10: Rapid review search terms

Broad fields	• Community care • Health • Long-term conditions • Personal social services • Social care
Populations (long-term conditions and social care service users)	• Alzheimer's • Anxiet* • At risk (?) • Autis* • Bipolar affective disorder • Carer or informal carer or unpaid carer or caregiver or care giver (not foster not professional) • Chronic condition*/ill* • Cognit* dis*/disab* • Communication difficult* • Dementia • Depress* • Developmental disab* • Dis* • Health condition* • Hearing loss/impair*/deaf • Intellect* dis*/disab* • Learn* dis*/disab* • Learning difficult* • Long-term condition* or long-standing illness • Longstanding ill* • Mental health or mental health problem* • Mental* retard* • Physical* impair* / physical* disab* • Psychosis • Schizophrenia • Sight loss / visual* impair* / blind/Sensory impair* / sensory disab* • Visually impaired or sight loss
Concept of hard to reach, marginalised and vulnerable people: synonyms and related terms	• Asylum or asylum seekers or refugees or immigrants • Bisex* • Black comm* or BME or Ethnic minorit*, Black and minority ethnicity, Asian, BAME, ethni* • Gay • Gyps* • Hard-to-reach • Hidden or invisible • Homeless* • Linguistic minorities

	• Lesbian
	• LGBT
	• Marginal*or under-represented
	• Mental capacity
	• Minority comm* or population or group
	• Seldom-heard
	• Self-funders or privat* funded
	• Transex*
	• Transgend*
	• Traveller*
	• Undeserved
	• Vulnerable or frail
Concept of involvement and engagement across research pathway: synonyms and related terms: Involvement/engagement	• Access • Barriers or challenges • Consult* • Engage* • Include* • Involve* • Inclus* • Participat* • Prox* • Reach
Concept of involvement and engagement across research pathway: synonyms and related terms: Research pathway	• Administration or mode of data collection • Drop-out or withdraw, attrition or retention • Facilitators • Informed consent/consent • Non-respons* or non-participation • Payment or incentive • Population identification • Recruitment • Response • Sample bias or response bias or selection bias • Sampling • Strategies
Concept of effectiveness	• Cost* • Cost-effectiv*

Table 11: Final listing of searched databases

Abstracts in Social Gerontology

Academic Search Complete

ASSIA

British Nursing Index

CAB Extracts

CINAHL

Cochrane Library (Review and methods databases)

EMBASE

ESRC research catalogue

Google Scholar

IBSS

Medline

Psychinfo

Psych Articles

SCOPUS

Science Citation Index Expanded

Social Care on Line

Social Sciences Citation Index

Table 12: Listing of hand searched journals

JARID (1 year)

JIDR (6 months)

BJLD (6 months

Disability and Society (10 years)

Figure 2 Information extracted for each paper and then mapped or analysed.

Author/Title/Date/Publication

Type of publication (primary research, review, opinion piece, etc.)

Aim(s) of paper (and wider study, if applicable)

Country where study took place

Study design (and wider study, if applicable)

Hard to reach or engage population

Sampling/recruitment procedures

Number and characteristics of participants

Details of any relevant theory/concepts used

Methods of data collection (postal survey, face-to-face interviews, telephone interviews, focus groups, observation, mixed-methods etc)

Facilitators/strategies to increase participation (e.g. incentives, alternative formats, reminders, flexible study design)

Barriers to participation

Type of analysis and summary of findings

Ethical issues related to those 'hard to reach'

Economic cost implications (indicate if 'not discussed')

Effectiveness of research approach for involving hard to reach etc (indicate if 'not discussed')

Validity issues and study limitations

Use of proxies (where applicable) – Relationship between proxy and participant (eg. Careworker, daughter, etc) – Level of contact between proxy and participant (eg daily, weekly, twice a year) – Nature of contact (eg. face-to-face, by telephone, writing)

Suggestions for further research work (if relevant)

Most relevant findings for the rapid review

 'Grading' or Scoring of the paper. * 1 = Robust* 2= Adequate *3 Limited *Q (query – add comment to notes)

Notes: e.g. references, keywords, if cannot quality appraise.

References

ABBOTT, M., ARTHUR, A., WALKER, L. & DOODY, G. 2005. The challenge of recruiting people with schizophrenia to a health promotion trial. British Journal of General Practice, 55**,** 634-636.

ALDRIDGE, J. 2007. Picture this: the use of participatory photographic research methods with people with learning disabilities. Disability & Society, 22**,** 1,17.

ALLBUTT, H. & MASTERS, H. 2010. Ethnography and the ethics of undertaking research in different mental healthcare settings. Journal of Psychiatric & Mental Health Nursing, 17**,** 210-215.

ALLISON, T., AHMAD, T., BRAMMAH, T., SYMMONS, D. & URWIN, M. 2003. Can findings from postal questionnaires be combined with interview results to improve the response rate among ethnic minority populations? Ethnicity & Health, 8**,** 63-69.

ANDREWS, J. 2005. Wheeling uphill? Reflections of practical and methodological difficulties encountered in researching the experiences of disabled volunteers. Disability & Society, 20**,** 201-212.

ANTAKI, C., YOUNG, N. & FINLAY, M. 2002 Shaping clients' answers: Departures from neutrality in care-staff interviews with people with a learning disability, Disability & Society, 17, 4, 6, 435-455.

ATKINSON, D. (1988). Research interviews with people with mental handicaps. *Mental Handicap Research, 1*(1), 75-90.

ATKINSON, D. (2005). Research as social work: Participatory research in learning disability. *British Journal of Social Work, 35*(4), 425-434. doi:10.1093/bjsw/bch189

ATKINSON, R. & FLINT, J. 2001. Accessing hidden and hard-to-reach populations: snowball research strategies. Social Research Update, Issue 33.

BARR, O., MCCONKEY, R. & MCCONAGHIE, J. 2003. Views of people with learning difficulties about current and future accommodation: The use of focus groups to promote discussion. Disability & Society, 18**,** 577-597.

BEADLE-BROWN, J., MURPHY, G. & DITERLIZZI, M. 2009. Quality of life for the Camberwell cohort. Journal of Applied Research in Intellectual Disabilities, 22**,** 380-390.

BOYDEN, P., ESSCOPRI, N., OGI, L., BRENNAN, A. & KALSY-LILLICO, S. 2009. Service users leading the way: Focus group methodology in developing accessible information DVDs with people with learning disabilities. Journal of Intellectual Disabilities, 13**,** 183-194.

BOYNTON, P. M., WOOD, G. W. & GREENHALGH, T. 2004. Reaching beyond the white middle class. British Medical Journal**,** 328, 1433-1436.

BREWSTER, S. J. 2004. Putting words in their mouths? Interviewing people with learning disabilities and little/no speech. British Journal of Learning Disabilities. 32, 166-169

BROWN, P. & SCULLION, L. 2010. 'Doing research' with Gypsy–Travellers in England: reflections on experience and practice. Community Development Journal, 45**,** 169-185.

BROWNLOW, C. & O'DELL, L. 2002. Ethical issues for qualitative research in on-line communities. Disability & Society, 17**,** 685-694.

BRYAN, S., HARDYMAN, W., BENTHAM, P., BUCKLEY, A. & LAIGHT, A. 2005. Proxy completion of EQ-5D in patients with dementia. Quality of Life Research, 14**,** 107-118.

CABINET OFFICE, Inclusion Health (2012) http://webarchive.nationalarchives.gov.uk/+/http://www.cabinetoffice.gov.uk/social_exclusion_task_force/short_studies/health-care.aspx

CAMBRIDGE, P. & FORRESTER-JONES, R. 2003. Using individualised communication for interviewing people with intellectual disability: a case study of user-centred research. Journal of Intellectual & Developmental Disability, 28**,** 5- 23.

CAMBRIDGE, P. & MCCARTHY, M. 2001. User focus groups and best value in services for people with learning disabilities. Health and Social Care in the Community, 9**,** 476-489.

CAMERON, L. & MURPHY, J. 2007. Obtaining consent to participate in research: The issues involved in including people with a range of learning and communication disabilities. British Journal of Learning Disabilities, 35**,** 113-120.

CLEAVER, S., OUELLETTE-KUNTZ, H., & SAKAR, A. (2010). Participation in intellectual disability research: A review of 20 years of studies. Journal of Intellectual Disability Research : JIDR, 54(3), 187-193.

CLEGG, J. 2004. Practice in focus: A hermeneutic approach to research ethics. British Journal of Learning Disabilities, 32**,** 186-190.

COUCILL, W., BRYAN, S., BENTHAM, P., BUCKLEY, A. & LAIGHT, A. 2001. EQ-5D in patients with dementia – An investigation of inter-rater agreement. Medical Care, 39**,** 760-771.

CUMMINS, R (1991). 'The comprehensive quality of life scale – intellectual disablity: An instrument under development.' *Australia and New Zealand Journal of Development Disabilities* 17(2): 259-264.

CUMMINS, R (1997). 'Comprehensive quality of life scale – intellectual/cogntive disablity 5th edition: Manual', School of Psychology, Deaking University.

DALEMANS, R., WADE, D. T., VAN DEN HEUVEL, W. J. A., & DE WITTE, L. P. (2009). Facilitating the participation of people with aphasia in research: A description of strategies. Clinical Rehabilitation, 23(10), 948-959.

DAVIES, K., COLLERTON, J. C., JAGGER, C., BOND, J., BARKER, S. A. H., EDWARDS, J., HUGHES, J., HUNT, J. M. & ROBINSON, L. 2010. Engaging the oldest old in research: lessons from the Newcastle 85 study. BMC Geriatr, 10, 64-73

DEPARTMENT OF HEALTH 2011. Transparency in outcomes: a framework for quality in adult social care: A response to the consultation and next steps. Department of Health, London.

DONNELLY, J. 2004. Can adults with cognitive impairment consent to take part in research? Journal of Wound Care, 13**,** 257-62.

DUCKETT, P. S. & PRATT, R. 2001. The researched opinions on research: visually impaired people and visual impairment research. Disability & Society, 16**,** 815-835.

DWAN, K., ALTMAN, D. G., ARNAIZ, J. A., BLOOM, J., AN-WEN, C., CRONIN, E., DECULLIER, E., EASTERBROOK, P. J., VON ELM, E., GAMBLE, C., GHERSI, D., IOANNIDIS, J. P. A., SIMES, J. & WILLIAMSON, P. R. 2008. Systematic Review of the Empirical Evidence of Study Publication Bias and Outcome Reporting Bias. PLoS Clinical Trials, 5**,** 1-31.

DYE, L., HARE, D. J. & HENDY, S. 2007. Capacity of people with intellectual disabilities to consent to take part in a research study. Journal of Applied Research in Intellectual Disabilities, 20**,** 168-174.

DYE, L., HENDY, S., HARE, D. J. & BURTON, M. 2004. Capacity to consent to participate in research – a recontextualization. British Journal of Learning Disabilities, 32**,** 144-150.

EVENHUIS, H., VAN SPLUNDER, J., VINK, M., WEERDENBURG, C., VAN ZANTEN, B., & STILMA, J. (2004). Obstacles in large-scale epidemiological assessment of sensory impairments in a dutch population with intellectual disabilities. Journal of Intellectual Disability Research, 48(8), 708-718.

FANG, J., FLECK, M. P., GREEN, A., MCVILLY, K., HAO, Y., TAN, W., FU, R. & POWER, M. 2011. The response scale for the intellectual disability module of the WHOQOL: 5-point or 3-point? Journal of Intellectual Disability Research, 55**,** 537-549.

FENGE, L.-A. 2010. Striving towards inclusive research: an example of participatory action research with older lesbians and gay men. British Journal of Social Work, 40**,** 878-894.

FINLAY, W. M. L. & LYONS, E. 2002. Acquiescence in interviews with people who have mental retardation. Mental Retardation, 40**,** 14-29.

FINLAY, W. M. L. & LYONS, E. 2001. Methodological issues in interviewing and using self-report questionnaires with people with mental retardation. Psychological Assessment, 13**,** 319-335.

FOXCROFT, D. R. & SMITH, L. 2008. We need to guard against bias. Drug & Alcohol Review, 27**,** 346-348.

FRASER, M. & FRASER, A. 2001. Are people with learning disabilities able to contribute to focus groups on health promotion? Journal of Advanced Nursing, 33**,** 225-233.

GARLAND, J., SPALEK, B. & CHAKRABORTI, N. 2006. Hearing lost voices: issues in researching 'hidden' minority ethnic communities. British Journal of Criminology, 46.423-437.

GARRARD, E. & DAWSON, A. 2005. What is the role of the research ethics committee? Paternalism, inducements, and harm in research ethics. Journal of Medical Ethics, 31**,** 419 – 423.

GATES, B. & WAIGHT, M. 2007. Reflections on conducting focus groups with people with learning disabilities: Theoretical and practical issues. Journal of Research in Nursing, 12**,** 111-126.

GILBERT, T. 2004. Involving people with learning disabilities in research: issues and possibilities. Health & Social Care in the Community, 12**,** 298-308.

GORDON, M. S., SHEVLIN, M., TIERNEY, K. J., BUNTING, B. & TRIMBLE, T. 2007. Correspondence between self-ratings and key-workers' ratings of depression in adults with mild learning disabilities. British Journal of Clinical Psychology, 46**,** 491-495.

GOSDEN, T. & KIRKLAND, J. 2008. A focus group for fathers with learning disabilities: Using a qualitative analysis to develop an understanding of their experiences. Clinical Psychology Forum**,** 191, 20-24.

HAMILTON, C., & ATKINSON, D. (2009). 'A story to tell': Learning from the life-stories of older people with intellectual disabilities in Ireland. *British Journal of Learning Disabilities, 37*(4), 316-322.

HANCOCK, G. A., REYNOLDS, T., WOODS, B., THORNICROFT, G. & ORRELL, M. 2003. The needs of older people with mental health problems according to the user, the carer, and the staff. International Journal of Geriatric Psychiatry, 18**,** 803-811.

HARKINS, C., SHAW, R., GILLIES, M., SLOAN, H., MACINTYRE, K., SCOULAR, A., MORRISON, C., MACKAY, F., CUNNINGHAM, H., DOCHERTY, P., MACINTYRE, P. & FINDLAY, I. 2010. Overcoming barriers to engaging socio-economically disadvantaged populations in CHD primary prevention: a qualitative study. BMC Public Health, 10**,** 391-398

HEAL, L. W., & SIEGELMAN, C. K. (1996). Methodological issues in quality of life measurement. In R. L. Schalock (Ed.), *Quality of life vol.1: Conceptualization and measurement* (pp. 91-104)

HILARI, K., OWEN, S. & FARRELLY, S. J. 2007. Proxy and self-report agreement on the Stroke and Aphasia Quality of Life Scale-39. Journal of Neurology, Neurosurgery & Psychiatry, 78**,** 1072-5.

HOE, J., KATONA, C., ORRELL, M. & LIVINGSTON, G. 2007. Quality of life in dementia: care recipient and caregiver perceptions of quality of life in dementia: the LASER-AD study. International Journal of Geriatric Psychiatry, 22**,** 1031-1036.

HOWARD, L., DE SALIS, I., TOMLIN, Z., THORNICROFT, G. & DONOVAN, J. 2009. Why is recruitment to trials difficult? An investigation into recruitment difficulties in an RCT of supported employment in patients with severe mental illness. Contemporary Clinical Trials, 30**,** 40- 46.

IACONO, T. (2006). Ethical challenges and complexities of including people with intellectual disability as participants in research. Journal of Intellectual & Developmental Disability, 31(3), 173-179.

JONES, S. P. 2008. From marginalization to participation and back again: Difficulties – but for how long? Disability Studies Quarterly, 28. http://dsq-sds.org/article/view/95/95 (Retrieved 20.4.12)

KAEHNE, A. & O'CONNELL, C. 2010. Focus groups with people with learning disabilities. Journal of Intellectual Disabilities, 14**,** 133-145.

Kroese, B. S., Gillott, A., & Atkinson, V. (1998). Consumers with intellectual disabilities as service evaluators. *Journal of Applied Research in Intellectual Disabilities, 11*(2), 116-128.

LENNOX, N., TAYLOR, M., REY-CONDE, T., BAIN, C., PURDIE, D. M., & BOYLE, F. (2005). Beating the barriers: Recruitment of people with intellectual disability to participate in research. Journal of Intellectual Disability Research, 49, 296-305.

LINEHAN, C., WALSH, P. N., LANTMAN-DE VALK, H. M. J. V. S., KERR, M. P., DAWSON, F. & POMONA, I. G. 2009. Are people with intellectual disabilities represented in European public health surveys? Journal of Applied Research in Intellectual Disabilities, 22**,** 409-420.

LLEWELLYN, P. 2009. Supporting people with intellectual disabilities to take part in focus groups: Reflections on a research project. Disability & Society, 24**,** 845-856.

LLOYD, C. E., JOHNSON, M. R., MUGHAL, S., STURT, J. A., COLLINS, G. S., ROY, T., BIBI, R. & BARNETT, A. H. 2008. Securing recruitment and obtaining informed consent in minority ethnic groups in the UK. BMC Health Services Research**,** 8, 68-77.

LLOYD, V., GATHERER, A. & KALSY, S. 2006. Conducting qualitative interview research with people with expressive language difficulties. Qualitative Health Research, 16**,** 1386-1404.

LOWTON, K. 2005. Trials and tribulations: understanding motivations for clinical research participation amongst adults with cystic fibrosis. Social Science & Medicine, 61**,** 1854-65.

MANSELL, J. 2011. Structured observational research in services for people with learning disabilities. SSCR methods review, 10. NIHR School for Social Care Research, London, UK.

MATHERS, A. R. 2008. Hidden voices: the participation of people with learning disabilities in the experience of public open space. Local Environment, 13**,** 515-529.

MCCONKEY, R. (1996). New perspectives on evaluations. In R. McConkey (Ed.), *Innovations in evaluating services for people with intellectual disabilities* (pp. 3-12)

MCDONALD, K. E., KIDNEY, C. A., NELMS, S. L., PARKER, M. R., KIMMEL, A., & KEYS, C. B. (2009). Including adults with intellectual disabilities in research: Scientists' perceptions of risks and protections. Journal of Policy and Practice in Intellectual Disabilities, 6(4), 244-252.

MCGAURAN, N., WIESELER, B., KREIS, J., SCHÜLER, Y.-B., KÖLSCH, H. & KAISER, T. 2010. Reporting bias in medical research- a narrative review. Trials, 11**,** 37-51.

MCKEE, K. J., HOUSTON, D. M. & BARNES, S. 2002. Methods for assessing quality of life and well-being in frail older people. Psychology & Health, 17**,** 737-751.

MCKEOWN, J., CLARKE, A., INGLETON, C. & REPPER, J. 2010. Actively involving people with dementia in qualitative research. Journal of Clinical Nursing, 19**,** 1935 – 1943.

MCNALLY, S. 2002. A survey of self-advocacy groups for people with learning disabilities in an English region. Journal of Learning Disabilities, 2**,** 185-199.

MINDHAM, J. & ESPIE, C. A. 2003. Glasgow Anxiety Scale for people with an Intellectual Disability (GAS-ID): development and psychometric properties of a new measure for use with people with mild intellectual disability. Journal of Intellectual Disability Research, 47**,** 22-30.

MOSER, C. A. 1958. Survey Methods in Social Investigation. Heinemann Educational Books, London.

MURPHY, J., TESTER, S., HUBBARD, G., DOWNS, M. & MACDONALD, C. 2005. Enabling frail older people with a communication difficulty to express their views: the use of Talking Mats™ as an interview tool. Health & Social Care in the Community, 13**,** 95-107.

MURPHY, J. and CAMERON, L 2008. The effectiveness of talking mats with people with intellectual disability. British Journal of Learning Disabilities. 36, 232-241.

NICE 2006. Methods for development of NICE public health guidance

NIND, M. 2009. Conducting qualitative research with people with learning, communication and other disabilities; methodological challenges. National Centre for Review Methods.

NOTA, L, SORESI, S & PERRY, J (2006). 'Quality of life in adults with an intellectual disability: The evaluation of quality of life instrument.' *Journal of Intellectual Disability Research* 50: 371-385.

OLIVER-AFRICANO, P., DICKENS, S., AHMED, Z., BOURAS, N., COORAY, S. & DEB, S. 2010. Overcoming the barriers experienced in conducting a medication trial in adults with aggressive challenging behaviour and intellectual disabilities. Journal of Intellectual Disability Research, 54**,** 17-25.

OLIVER, P. C., PIACHAUD, J., DONE, J., REGAN, A., COORAY, S. & TYRER, P. 2002. Difficulties in conducting a randomized controlled trial of health service interventions in intellectual disability: implications for evidence-based practice. Journal of Intellectual Disability Research, 46**,** 340-345.

O'REILLY, M., ARMSTRONG, N. & DIXON-WOODS, M. 2009. Subject positions in research ethics committee letters: a discursive analysis. Clinical Ethics, 4**,** 187-194.

OUELLETTE-KUNTZ, H (1990). 'A pilot study in the use of the quality of life schedule.' *Social Indicators Research* 23: 283-298.

PARRY, O., BANCROFT, A., GNICH, W., & AMOS, A. (2001). Nobody home?: issues of respondent recruitment in areas of deprivation. Critical Public Health, 11, 305 – 317.

PAWSON, N., RAGHAVAN, R., SMALL, N., CRAIG, S. & SPENCER, M. 2005. Social inclusion, social networks and ethnicity: the development of the Social Inclusion Interview Schedule for young people with learning disabilities. British Journal of Learning Disabilities, 33**,** 15-22.

PERRY, J. & FELCE, D. 2002. Subjective and objective quality of life assessment: Responsiveness, response bias, and resident:proxy concordance. Mental Retardation, 40**,** 445-456.

PERRY, J., & FELCE, D. (2004). Initial findings on the involvement of people with an intellectual disability in interviewing their peers about quality of life. Journal of Intellectual and Developmental Disability, 29(2), 164-171.

PINTO, R. R. M., MCKAY, M. M., BAPTISTE, D., BELL, C. C., MADISON-BOYD, S., PAIKOFF, R., WILSON, M. & PHILLIPS, D. 2007. Motivators and barriers to participation of ethnic

minority families in a family-based HIV prevention program. Social Work in Mental Health, 5, 187-201.

POPAY, J., ROBERTS, H., SOWDEN, A., PETTICREW, M., ARAI, L., RODGERS, M., BRITTEN, N. & WITH ROEN, K. A. D., S. 2006. Guidance on the Conduct of Narrative Synthesis in Systematic Reviews. ESRC.

PROCTOR, G. 2001. Listening to older women with dementia: relationships, voices and power. Disability & Society, 16, 361-376.

RAMCHARAN, P. & CUTCLIFFE, J. R. 2001. Judging the ethics of qualitative research: considering the 'ethics as process' model. Health & Social Care in the Community, 9, 358-366.

RAMCHARAN, P., & GRANT, G. (2001). Views and experiences of people with intellectual disabilities and their families.(1) the user perspective. *Journal of Applied Research in Intellectual Disabilities, 14*(4), 348-363.

ROBERTS, L. M. WILSON, S. ROALFE, A. BRIDGE, P. (2004) A randomised controlled trial to determine the effect on response of including a lottery incentive in health survey *BMC Health Serv Res*, 4, 30

ROGLER, L. H., MROCZEK, D. K., FELLOWS, M. & LOFTUS, S. T. 2001. The neglect of response bias in mental health research. Journal of Nervous and Mental Disease, 189, 182-187.

ROONEY, L. K., BHOPAL, R., HALANI, L., LEVY, M. L., PARTRIDGE, M. R., NETUVELI, G., CAR, J., GRIFFITHS, C., ATKINSON, J., LINDSAY, G. & SHEIKH, A. 2011. Promoting recruitment of minority ethnic groups into research: qualitative study exploring the views of South Asian people with asthma. Journal of Public Health. 33, 604-15.

ROSS, E. & OLIVER, C. 2003. Preliminary analysis of the psychometric properties of the Mood, Interest & Pleasure Questionnaire (MIPQ) for adults with severe and profound learning disabilities. British Journal of Clinical Psychology, 42, 81-93.

RUGKASA, J. & CANVIN, K. 2011. Researching Mental Health in Minority Ethnic Communities: Reflections on Recruitment. Qualitative Health Research, 21, 132 – 143.

SCHALOCK, R. L. (1996). Reconsidering the conceptualization and measurement of quality of life. In R. L. Schalock (Ed.), *Quality of life vol.1: Conceptualization and measurement* (pp. 123-139)

SCHMIDT, S., POWER, M., GREEN, A., LUCAS-CARRASCO, R., ESER, E., DRAGOMIRECKA, E. & FLECK, M. 2010. Self and proxy rating of quality of life in adults with intellectual disabilities: Results from the DISQOL study. Research in Developmental Disabilities, 31, 1015-1026.

SCOTT, P. & EDWARDS, P. 2006. Personally addressed hand-signed letters increase questionnaire response: a meta-analysis of randomised controlled trials. BMC Health Services Research, 6.111-115.

SELAI, C. E., TRIMBLE, M. R., ROSSOR, M. N. & HARVEY, R. J. 2001. Assessing quality of life in dementia: Preliminary psychometric testing of the Quality of Life Assessment Schedule (QOLAS). Neuropsychological Rehabilitation, 11, 219-243.

SEYMOUR, D. G., BALL, A. E., RUSSELL, E. M., PRIMROSE, W. R., GARRATT, A. M. & CRAWFORD, J. R. 2001. Problems in using health survey questionnaires in older patients with physical disabilities. The reliability and validity of the SF-36 and the effect of cognitive impairment. Journal of Evaluation in Clinical Practice, 7**,** 411 – 418.

SHEIKH, A., HALANI, L., BHOPAL, R., NETUVELI, G., PARTRIDGE, M. R., CAR, J., GRIFFITHS, C. & LEVY, M. 2009. Facilitating the recruitment of minority ethnic people into research: qualitative case study of South Asians and asthma. PLoS Medicine [Online]. Available: http://www.mrw.interscience.wiley.com/cochrane/clcmr/articles/CMR-15598/frame.html.

SIDDIQI, N. 2011. Publication bias in epidemiological studies. Central European Journal of Public Health, 19**,** 118-120.

SKAFF, M. M., CHESLA, C. A., MYCUE, V. D. L. S. & FISHER, L. 2002. Lessons in cultural competence: Adapting research methodology for Latino participants. Journal of Community Psychology, 30**,** 305-323.

SOCIAL EXCLUSION TASK FORCE, CABINET OFFICE (2010) Inclusion Health. Improving the way we meet the primary health care needs of the socially excluded. HM Government, London. http://webarchive.nationalarchives.gov.uk/+/http://www.cabinetoffice.gov.uk/media/346571/inclusion-health.pdf

STALKER, K., CARPENTER, J., CONNORS, C. & PHILLIPS, R. 2004. Ethical issues in social research: difficulties encountered gaining access to children in hospital for research. Child: Care, Health & Development, 30**,** 377-383.

TALLON, D., MULLIGAN, J., WILES, N., THOMAS, L., T.J., P., ELGIE, R., SHARP, D. & LEWIS, G. 2011. Involving patients with depression in research: survey of patients' attitudes to participation. British Journal of General Practice, 61**,** 134 – 141.

TUFFREY-WIJNE, I., BERNAL, J. & HOLLINS, S. 2008. Doing research on people with learning disabilities, cancer and dying: ethics, possibilities and pitfalls. British Journal of Learning Disabilities, 36.185-190.

TUFFREY-WIJNE, I., BERNAL, J., HUBERT, J., BUTLER, G. & HOLLINS, S. 2010. Exploring the lived experiences of people with learning disabilities who are dying of cancer. Nursing Times, 106**,** 15-18.

TUFFREY-WIJNE, I. & BUTLER, G. 2010. Co-researching with people with learning disabilities: an experience of involvement in qualitative data analysis. Health Expectations, 13**,** 174-184.

TUFFREY-WIJNE, I. and DAVIES, J. 2007. This is my story: I''ve got cancer: 'The Veronica Project': An ethnographic study of the experiences of people with learning disabilities who have cancer. British Journal of Learning Disabilities. 35, 7-11.

ULIVI, G., REILLY, J. & ATKINSON, J. M. 2009. Protection or empowerment: Mental health service users' views on access and consent for non-therapeutic research. Journal of Mental Health, 18**,** 161 – 168.

VEENSTRA, M. Y., WALSH, P. N., LANTMAN-DE VALK, H. M. J. V. S., HAVEMAN, M. J., LINEHAN, C. & KERR, M. P. 2010. Sampling and ethical issues in a multicenter study

on health of people with intellectual disabilities. Journal of Clinical Epidemiology, 63**,** 1091-1100.

WARNER, J., MCCARNEY, R., GRIFFIN, M., HILL, K. & FISHER, P. 2008. Participation in dementia research: rates and correlates of capacity to give informed consent. Journal of Medical Ethics, 34**,** 167 – 170.

WHELAN, P. J. P., OLESZEK, J., MACDONALD, A. & GAUGHRAN, F. 2009. The utility of the Mini-Mental State Examination in guiding assessment of capacity to consent to research. International Psychogeriatrics, 21**,** 338-344.

WILLIAMS, B., IRVINE, L., MCGINNIS, A. R., MCMURDO, M. E. & CROMBIE, I. K. 2007. When "no" might not quite mean "no"; the importance of informed and meaningful non-consent: results from a survey of individuals refusing participation in a health-related research project. BMC Health Services Research [Online].
Available: http://www.mrw.interscience.wiley.com/cochrane/clcmr/articles/CMR-10704/frame.html.

WILLIAMS, C. L., TAPPEN, R., BUSCEMI, C., RIVERA, R. & LEZCANO, J. 2001. Obtaining family consent for participation in Alzheimer's research in a Cuban-American population: Strategies to overcome the barriers. American Journal of Alzheimer's Disease, 16**,** 183-187.

WILSON, E., POLLOCK, K. & AUBEELUCK, A. 2010. Gaining and maintaining consent when capacity can be an issue: A research study with people with Huntington's disease. Clinical Ethics, 5**,** 142-147.

WOODALL, A., HOWARD, L. & MORGAN, C. 2011. Barriers to participation in mental health research: Findings from the Genetics and Psychosis (GAP) Study. International Review of Psychiatry, 23**,** 31-40.

YOUNG, A. F. & CHESSON, R. A. 2006. Obtaining views on health care from people with learning disabilities and severe mental health problems. British Journal of Learning Disabilities, 34**,** 11-19.

YOUNG, A. F. & CHESSON, R. A. 2008. Determining research questions on health risks by people with learning disabilities, carers and care-workers. British Journal of Learning Disabilities, 36**,** 22-31.

ZERMANSKY, A. G., ALLDRED, D. P., PETTY, D. R. & RAYNOR, D. K. 2007. Striving to recruit: the difficulties of conducting clincial research on elderly care home residents. Journal of the Royal Society of Medicine, 100**,** 258 – 261.