Developing a patient-centred patient-reported outcome measure (PROM) for cognitive rehabilitation after stroke: the Patient-Reported Evaluation of Cognitive State (PRECiS) scale

A thesis submitted to the University of Manchester for the degree of Doctor of Philosophy in the Faculty of Medical and Human Sciences

2015

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Word Count: 41, 080

List of Abbreviations

ACT NoW	Assessing Communication Therapy in the North West
ADL	Activity of Daily Living
ANOVA	Analysis of Variance
AST TULIA	Apraxia Screen - Test of Upper Limb Apraxia
BASIC	Brain and Spinal Cord Injury Center
CCT	controlled clinical trial
CLAHRC	Collaboration for Leadership in Applied Health Research and Care
COAST	Communication Outcomes after Stroke
COMET	Core Outcome Measures in Effectiveness Trials
COPE	Carers of Older People in Europe
COREQ	Consolidated criteria for reporting qualitative research
DH	Department of Health
EADL	Extended Activity of Daily Living
FAST	Frenchay Aphasia Screening Test
GAD7	Generalised Anxiety Disorder Scale
G-MASTER	Greater Manchester Assessment of Stroke Rehabilitation
GM-SAT	Greater Manchester Stroke Assessment Tool
GP	General Practitioner
IAPT	Improving Access to Psychological Therapy
ICC	Intraclass correlation coefficient
ICF	International Classification of Functioning, Disability and Health
IN-CAM	Canadian Interdisciplinary Network for Complementary and Alternative Medical Research outcomes database
JLA	James Lind Alliance
MADYSS	Macclesfield and District Young Stroke Society
MoCA	Montreal Cognitive Assessment
NELFT	North East London Foundation Trust

NHS	National Health Service
NIHR	National Institute for Health Research
OPSYRIS	Organisation for Psychological Research in Stroke
PCRN	Primary Care Research Network
PHQ9	Patient Health Questionnaire Scale
PIC	Participant Identification Centre
PiiAF	Public Involvement Assessment Framework
PPI	Patient and Public Involvement
PRECiS	Patient Reported Evaluation of Cognitive State
PROM	Patient Reported Outcome Measure
PROQOLID	Patient-reported outcome and quality of life instrument database
R&D	Research and Development
RCTs	randomised controlled trials
ROM	registry of outcome measures
RUG	Research User Group
SA	Stroke Association
SD	Standard deviation
SIGN	Scottish Intercollegiate Guidelines Network
SIPSO	Subjective Index of Physical and Social Outcomes
SIS	Stroke Impact Scale
UK	United Kingdom
VAS	visual analogue scales
WHO	World Health Organisation

Thesis Abstract

Emma Patchick. The University of Manchester

Abstract of Thesis submitted for the degree of Doctor of Philosophy. June 2015

Developing a patient-centred patient-reported outcome measure (PROM) for cognitive rehabilitation after stroke: the Patient-Reported Evaluation of Cognitive State (PRECiS) scale

Cognitive difficulties can persist for months and years after stroke and adversely impact confidence, mood and functional recovery. Stroke survivors, carers and healthcare professionals collectively agree that improving cognition is the number one research priority for life after stroke. Future research should include measurements of outcome that service users deem important. Patient reported outcome measures (PROMs) are a means of gaining patient perspectives that can be standardised for use in a trial. PROMs should be developed with service users to incorporate their priorities but people with cognitive difficulties are often systematically excluded from the development and use of PROMs.

Study 1 used qualitative interviews (N=16) to explore stroke survivor perspectives on the important and measureable impacts of persisting cognitive problems. The results of this study generated requirements for a PROM that related to conceptual underpinning and face validity of a measurement tool.

Study 2 was a systematic review of existing PROMs related to cognition. 20 Identified PROMs were critically appraised against the requirements generated in the qualitative study. No existing PROMs were identified that met all of the qualitative study review criteria.

The next stage described in chapter 3, was to develop a new PROM that: utilised the strengths of existing tools; met qualitative study requirements; and was refined through consultation with different stakeholders, prioritising feedback of stroke survivors with cognitive difficulties. The result of this work was the Patient Reported Evaluation of Cognitive State (PRECiS) scale.

Study 3 was a psychometric study with stroke survivors (N=164) to test PRECiS in a large sample. Quantitative and qualitative data were collected on acceptability, feasibility and other psychometric properties of validity and reliability.

PRECiS demonstrated good acceptability to stroke survivors and performed well psychometrically. Future validation work required for PRECiS is described in discussion chapter 4. Subject to further validation work, PRECiS may be particularly useful for pragmatic trials of cognitive rehabilitation after stroke.

Lay Abstract of Thesis

Emma Patchick. The University of Manchester

Abstract of Thesis submitted for the degree of Doctor of Philosophy. June 2015

Developing a patient-centred patient-reported outcome measure (PROM) for cognitive rehabilitation after stroke: the Patient-Reported Evaluation of Cognitive State (PRECiS) scale

After a stroke, many survivors can experience difficulties thinking and understanding. They may have problems with memory, perception, problem-solving, planning, attention, and language. These problems are known as cognitive difficulties. Cognitive difficulties after stroke can affect confidence and mood as well as the ability to recover. Stroke survivors themselves can tell us about the impact of cognitive problems and whether a treatment has worked for them. Their opinions on treatment can be collected using questionnaires known as Patient Reported Outcome Measures. However, people with cognitive difficulties are rarely involved in designing and using these questionnaires.

The first study in this thesis involved interviews with 16 stroke survivors. They talked about the important impacts of cognitive problems. These interviews were used to make recommendations about what a questionnaire should include and how it should look.

The second study identified 20 existing questionnaires and compared them against the recommendations from the first study. None of the 20 questionnaires matched all of the recommendations, so a new questionnaire was needed.

Stroke survivors then helped to develop and refine this new questionnaire that was named: the Patient Reported Evaluation of Cognitive State (PRECiS) scale.

The final study tested PRECiS with a large number of stroke survivors with cognitive difficulties. Stroke survivors were positive about PRECiS and could complete it well, with support from a researcher. PRECiS seems to be a reliable questionnaire. That is, we get similar results when people complete PRECiS at two separate times. It also has other good qualities that we look for when testing questionnaires.

More work is needed to improve PRECiS and this is discussed in the final chapter of the thesis. PRECiS may be a useful questionnaire to help us understand whether treatment works for people. We hope it will help improve the lives of people with cognitive difficulties after stroke.

Declaration

No portion of the work referred to in the thesis has been submitted in support of an application for another degree or qualification of this or any other university or other institute of learning.

Copyright Statement

Acknowledgements

Firstly, I would like to thank my supervisors, Audrey Bowen and Andy Vail, for the guidance and support they have provided. They are a fantastic team and I couldn't hope for better mentors. I also thank my advisor, Anne Hesketh for her support.

I am grateful to colleagues from the Greater Manchester Collaboration for Leadership in Applied Health Research and Care (CLAHRC), whom I had the pleasure of working with whilst carrying out the qualitative study (study 1 in this thesis); particularly Maria Horne and Kate Woodward-Nutt.

I would also like to thank the Stroke Association who both funded the PhD and supported recruitment to studies. The Stroke Association is a fantastic charity and they have been a privilege and a pleasure to work with.

The research could not have been completed without the stroke survivors and carers who took part in the research studies and advised me as collaborators and consultees. I am grateful for their participation. I am also very grateful to all the General Practitioners and Healthcare Practitioners who support recruitment to my studies: they are too many to mention here but I thank you all. In particular, I would like to thank the excellent team at the Community Rehabilitation Team within the North East London NHS Foundation Trust (NELFT): Alison Wood; Sue Patterson and Jacki Mundy. Without your support as a late-recruited research site for the psychometric study, I would not have exceeded my target recruitment figure.

Finally, I would like to thank my partner and parents for knowing and saying exactly what I needed to hear to help me through difficult times. All my merits stem from your unconditional love and encouragement and I could not feel more blessed. Thank you.

A note about the format of the thesis

This thesis is presented in alternative format and permission has been obtained to do so. Three studies within this thesis are included in a format suitable for publication in a peer reviewed journal. Sections are also included in line with traditional thesis format.

The incorporation of publication-style sections leads to overlap with other sections of the thesis and some duplication is inevitable: this is acknowledged within the guidelines for alternative format theses.

The pages of the studies presented in publication format are not included in the pagination sequence of the thesis, as per the University of Manchester guidelines for presentation of an alternative format thesis.

1 Introduction

This chapter provides an introduction to the impact of cognitive difficulties experienced after stroke and, with reference to existing substantive reviews and clinical guidelines, summarises the evidence base for cognitive rehabilitation. More detail on cognitive impairment - including theoretical models and neuroanatomy - is described in detail elsewhere (Lezak, 2012; Rapp, 2001) and is not central to this thesis. The purpose of this background is to highlight the current priority to reduce the impact of cognitive difficulties after stroke and the need for trials to include outcomes of importance to stroke survivors. Patient-centred outcomes for cognitive rehabilitation are discussed; including consideration of their strengths and weaknesses. Considerations related to evaluating and developing outcome measures provide a rationale for the studies that have been carried out in this thesis.

1.1 Cognitive difficulties after stroke

Stroke is a leading cause of neurological disability in the United Kingdom (Adamson, Beswick, & Ebrahim, 2004). The National clinical guideline for stroke (2012) for England and Wales recognises that some cognitive loss is probably present in most survivors early after stroke but cognitive impairment can also persist for many months and years after stroke (Barker-Collo et al., 2010; Douiri, Rudd, & Wolfe, 2013; Schaapsmeerders et al., 2013).

Cognition is not a unitary concept: as indicated by the variety of neuropsychological assessment methods highlighted by Lezak (2012), it includes multiple domains that collectively facilitate complex mental processes: allowing individuals to understand; recognise; remember; and generally act upon the environment in a constructive way. Stroke can impair multiple cognitive domains that can be broadly categorised as follows:

- Attention. A domain which is itself multi-faceted and includes: alertness; selection of stimuli; dividing attention between several stimuli; and sustaining attention or concentrating (Levitt & Johnstone, 2001). Attention can be considered a 'starting point' or mediator for cognition since stimuli must be attended to on some level before processing and understanding can occur (McCabe et al., 2010). Attentional deficits have negative impacts on functional ability (Hyndman, Pickering, & Ashburn, 2008; Robertson et al., 1997).
- Memory. Involves the ability to encode, consolidate and retrieve information; influencing the recall of past events and making of new memories (Skeel & Edwards, 2001). Memory impairments can be related to a general reduction in functional ability

for everyday tasks, even after factors such as age and stroke severity are taken into consideration (Wade, Parker, & Langton Hewer, 1986).

- Perception. Involves the integration of the inputs received from the sense organs (e.g. eyes, nose, ears) in order to comprehend the world. Disorders of visual perception receive the most research attention (Mapou & Spector, 1995) and include issues with object recognition (visual agnosia); facial recognition (prosopagnosia); and visuospatial ability that can effect safe navigation of the environment. Perceptual impairments can negatively impact the execution of activities of daily living (ADLs) and functional abilities (Donnelly, Hextell, & Matthey, 1998; Mercier et al., 2001).
- Unilateral spatial neglect. A disorder whereby a patient fails to "notice" stimuli presented to the side opposite a brain lesion that can sometimes be mistaken for hemianopia (a visual field deficit) (Patel & Taylor, 1999). Disorders of neglect may lead sufferers to only eat food on one side of the plate; only groom one side of the body; bump into neglected objects. Neglect can impact patient safety and function of high-order activities such as cooking and driving (Lincoln, 2012; Wyness, 1985).
- Apraxias. Disorders that effect the ability to execute skilled movement that cannot be accounted for by "weakness, incoordination, sensory loss, poor concentration or inattention to command" (Geschwind, 1975). People with apraxia may be unable to perform actions on request (such as waving goodbye) but will perform those actions automatically in natural contexts (such as waving goodbye when leaving a room) (Koski, Iacoboni, & Mazziotta, 2002; Poeck, 1986). Apraxias can effect limb movements and speech. However, given the lack of high quality research into treatment of apraxia of speech (West et al., 2005), it receives little attention in this thesis. Motor apraxia has received more research attention and, although it is difficult to diagnose and treat (West et al., 2008) it does appear to negatively impact everyday function (Foundas et al., 1995; Hagmann, 1998).
- Executive dysfunction. Describes a broad body of mechanisms that are necessary for "appropriate, socially responsible, and effectively self-serving adult conduct." (Lezak, 2012). Executive functions include such actions as planning; initiation; problem solving; and organisation; and self-correction (Chung et al., 2013). Executive dysfunction may effect engagement with rehabilitation and is associated with poor social functioning (Lincoln, 2012).
- Aphasia is a communication disorder that leads to partial or total loss of the ability to articulate ideas or comprehend spoken and/or written language. Stroke is the most common cause of aphasia and it can have severe consequences; restricting social

participation and impacting negatively on personal relationships (Brumfitt, 1998; Hilari et al., 2010; Hodges, 2007).

In addition to these types of cognitive impairments, there may be a general loss of self-awareness or insight relating to the existence or severity of all deficits (anosognosia) (Callahan, 2001; Narushima, Moser, & Robinson, 2008). Anosognosia can reduce motivation to engage in therapy and lead to an inability to set realistic goals or apply useful compensatory strategies (Ownsworth & Clare, 2006; Port, Willmott, & Charlton, 2002).

Other common psychological consequences of stroke include mood disorders, such as depression and anxiety (Brown, O'Leary, & Barlow, 2001; House, 1987). They are mentioned here as they often co-exist with cognitive issues (Barker-Collo, 2007; Kauhanen et al., 2000) and may interact with one another in ways that could influence rehabilitation approaches (Mateer, Sira, & O'Connell, 2005). In fact, the Canadian Best Practice Recommendations for Stroke Care (2010) acknowledge the importance of considering mood and cognitive impairments together: "Post-stroke patients should also be screened for depression, since depression has been found to contribute to cognitive impairment in stroke patients."

Reported prevalence rates of cognitive impairments vary according to different methods of assessment. For example, prevalence rates for post-discharge attentional impairments in stroke survivors have been estimated at 51% (Hyndman, et al., 2008) and 92% (Stapleton, Ashburn, & Stack, 2001). There has been much research measuring cognitive impairment post-stroke (Hochstenbach et al., 1998; Lesniak et al., 2008; Tatemichi et al., 1994) but, as highlighted by Cumming et al (2013) no consistent one-size-fits-all "profile" has emerged; stroke survivors can have a variety of deficits across multiple domains of cognition. Therefore, describing cognitive impairments by domain is an over-simplification, but it is also a reflection of the design of most rehabilitation studies that tend to focus on a single impairment.

1.2 Understanding impact of cognitive problems

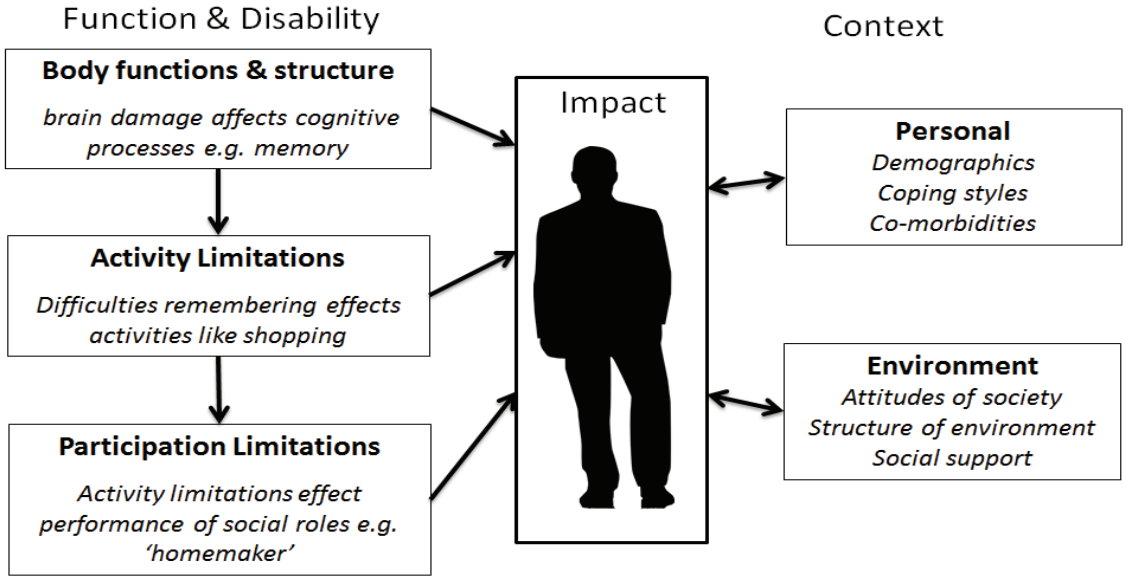
Some of the ways in which specific cognitive impairments are known to effect stroke survivors have been highlighted in section 1.1. Studies exploring statistical relationships between diagnosis and outcomes suggest that cognitive impairments adversely impact how well people participate in overall rehabilitation and recover functionally after stroke in the long term (Barker-Collo, et al., 2010; British Psychological Society, 2002; Claesson et al., 2005; Patel et al., 2002).

Qualitative studies have also been used to explore long-term impact; gaining stroke survivors and carer stories to document the lived experience of stroke. McKevitt et al (2004) review

qualitative studies in stroke; including studies that have been carried out to document the longer-term impact of the condition. They suggest that impact is generally described in terms of loss: of activities; abilities; personal characteristics; and independence. However, whilst some of the studies included in the review explore cognitive impairment (Grant, 1996; Parr, 2001), others have excluded people with cognitive impairments (Lister, 1999; Pound, Gompertz, & Ebrahim, 1998) and/or emphasised physical function when discussing impact (Ellis-Hill, Payne, & Ward, 2000; Kitzmuller, Haggstrom, & Asplund, 2013; Mumma, 1986). The tendency to exclude people with cognitive impairments from qualitative studies has been highlighted and is generally related to the challenges gaining narratives from those with expressive difficulties (Lloyd, Gatherer, & Kalsy, 2006; Paterson & Scott-Findlay, 2002).

The impacts of stroke and cognitive difficulties can be conceptualised according to a model developed by the World Health Organisation (2001): the International Classification of Functioning, Disability and Health (WHO ICF). The WHO ICF (2001) provides a framework for describing and understanding functioning and disability. It recognises that a disease such as stroke, effects bodily functions and structures (e.g. the brain and nervous system), which leads to impairments (such as the loss of cognitive function). These impairments can affect an individual by limiting their execution of activities and their participation in life situations and roles. There are contextual factors that will influence the effect that changes have on an individual: environmental factors that make up the "physical, social and attitudinal environment in which people live and conduct their lives" (WHO, 2001); personal factors related to the individual such as age, gender, co-morbidities, and coping styles. A representation of the WHO ICF model as it could be applied to understanding post-stroke cognitive difficulties is shown in Figure 1.1.

Figure 1.1 Representation of the WHO ICF model with examples related to cognition in italics



Within this framework, there is not a straightforward relationship between impairment and impact: “Diagnosis alone does not predict service needs, length of hospitalization, level of care or functional outcomes” (WHO, 2002). Rather, the impact of a condition (that can be articulated in terms of effected bodily structures; activities; and participation) is a result of a dynamic interaction between that condition and contextual factors specific to the individual (personal and environmental factors). This emphasises that in order to manage / rehabilitate cognitive issues, it is important to identify and understand person-specific contextual factors.

1.3 Consensus on prioritising cognitive problems in life after stroke

The purpose of this section is to highlight some of the evidence that influences the rationale for this thesis.

Wolfe et al (2008) analysed the evidence underpinning the recommendations in the National Stroke Strategy for England (Department of Health, 2007) and identified ten priority areas for stroke research based on gaps in the evidence base. Three of these ten priorities are relevant for the rationale of this thesis and they are:

- To estimate the longer-term needs of patients. There is reference to the lack of reported interventions for long-term support and a specific recommendation for qualitative research to identify patient priorities and their determinants in the longer term;
- The need for evaluation of the effectiveness of rehabilitation interventions in the post-acute / long-term;
- The importance of developing comprehensive outcome measures.

There is an emphasis here on chronicity and gaps in knowing how to improve life after stroke. The James Lind Alliance (JLA: http://www.lindalliance.org/) took a rigorous approach to identifying research priorities for life after stroke by consulting with stroke survivors, informal caregivers, and health professionals as well as searching relevant literature (Pollock et al., 2012). The number one identified priority was improving cognition and the number two and three priorities were respectively: helping people come to terms with the long-term consequences of stroke; and finding ways to help people recover from aphasia.

The Stroke Association carried out a survey with 799 stroke survivors who were at least one year post-stroke to explore their unmet needs. Almost half of the respondents reported problems with their mood and cognition that they felt had not been appropriately addressed (McKevitt et al., 2010). This finding is echoed in work that explored the feasibility of using a new tool to assess unmet needs of stroke patients within the context of a six month review

appointment (Rothwell et al., 2013). The new tool is called The Greater Manchester Stroke Assessment Tool (GM-SAT)(Rothwell, et al., 2013). Of the 137 stroke patients included, the most frequently identified unmet needs related to fatigue and cognitive issues: memory, concentration and attention. Recent work carried out with service users, carers and healthcare providers across 12 Greater Manchester localities also suggests that long-term support is not meeting the needs of stroke survivors with cognitive problems or their carers (Woodward-Nutt et al., 2013).

1.4 Cognitive Rehabilitation

1.4.1 A definition of cognitive rehabilitation

Rehabilitation can be described as an active, educational, problem-solving process that aims to maximise the functioning of an individual in their environment and reduce the overall impact of a health condition (Stucki, Cieza, & Melvin, 2007; Wade, 1992). Cognitive rehabilitation is not provided by a single health profession and typically involves psychologists, occupational therapists and speech and language therapists (Lincoln, 2012).

Rehabilitation approaches can be broadly categorised as related to restitution of function or optimisation / compensation of function (Lincoln, 1992). Initial assessment to determine the exact nature and impact of problems, as well as residual strengths, helps inform rehabilitation approaches (Lincoln, 1992; Prigatano et al., 1984).

Strategies to restore function rely on the ability of the brain to repair and recover after injury. They may involve the repetition and practice of impairment- or activity-specific exercises that attempt to re-establish connections in the brain (Robertson & Murre, 1999; Wilson, 1998). Repair and recovery may be more likely if the brain damage is milder (Robertson & Murre, 1999) and factors such as age, socioeconomic status and amount of pre-stroke disability may be important factors to influence recovery (Cramer, 2008).

Lincoln (2012) suggests that optimising function relies on intact cognitive skills and is sometimes used once attempts to restore function have been completed. It involves working with people with cognitive problems – and their families – to give information, teach compensation strategies and provide environmental aids that aim to help overcome and/or adjust to issues; ultimately reducing the impact of a health condition (Johnstone & Stonnington, 2001; Wade & de Jong, 2000).

1.4.2 The evidence base for cognitive rehabilitation - future directions

Bowen and Patchick (2013) reviewed literature to summarise knowledge about / recommendations for cognitive rehabilitation post stroke, including: Cochrane systematic reviews on post-stroke cognitive rehabilitation (Bowen et al., 2013; Bowen et al., 2011; Brady et al., 2012; Chung, et al., 2013; das Nair & Lincoln, 2007; Loetscher & Lincoln, 2013; West, et al., 2008); and National clinical guidelines / recommendations for stroke produced by several Westernised countries(Intercollegiate Stroke Working Party, 2012; Lindsay, et al., 2010; National Stroke Foundation, 2010; Scottish Intercollegiate Guidelines Network (SIGN), 2010). The book chapter (Bowen & Patchick, 2013) is included in Appendix 1 of this thesis (from page 136). It gives examples of interventions that have been used to treat cognitive difficulties after stroke and comments on service organisation within the UK National Health Service (NHS) required to deliver these.

There is considerable uncertainty about how best to rehabilitate cognitive problems; many of the recommendations in Clinical Guidelines for stroke are based on consensus as opposed to research evidence. Rehabilitation studies have tended to focus on a single cognitive impairment and, whilst this approach is important to help understand specific impairments, it would also be valuable to carry out high-quality pragmatic research that reflects rehabilitation in practice: with stroke survivors who have complex neuropsychological profiles, including impairments in multiple cognitive domains.

Cognitive rehabilitation research has tended to use impairment-focused outcome measures (van Heugten, Gregorio, & Wade, 2012) and the need to include outcomes deemed important by service users in future research has been identified (Bowen & Patchick, 2013; Cicerone et al., 2011).

1.5 Patient-centredness

UK policy regarding the National Health Service (NHS) has increasingly highlighted the importance of patient-centredness: putting patients and their families at the forefront to ensure that the design, delivery and evaluation of healthcare services are responsive to the needs and priorities of service users (Department of Health, 2004, 2005a, 2008b). The content of patient-centred care will depends on the circumstances, needs and preferences of the individual receiving care within a given care setting (Goodrich, 2008; Mead & Bower, 2000; The Health Foundation, 2014). However, one of the identified barriers to delivering patient-centred care is a lack of patient-centred outcome measures (Lawrence & Kinn, 2012; Mead & Bower, 2000).

1.6 Patient-centred, patient-reported outcome measures (PROMs)

Long and Dixon (1996) describe a spectrum of patient-centredness in outcome measures. At one end is the 'patient-defined' outcome, which is based on individualised information supplied directly from the patient related to: the outcomes of importance to them; how they might weight relative importance of those outcomes; and how far each of the outcomes has been achieved. Patient-defined outcomes may be particularly challenging to produce for individuals with severe disability or cognitive/communication difficulties (Carr & Higginson, 2001; Cruice et al., 2005; Macduff & Russell, 1998). At the opposite end of the patient-centred spectrum are 'professionally-defined' outcome measures that are based on clinicians rating the patient through assessment or observation (Long & Dixon, 1996).

A patient-reported outcome measure falls somewhere in the middle of that spectrum: completed using information supplied directly by the patient, without interpretation by others, but with a pre-defined structure (Fitzpatrick et al., 1998; Long & Dixon, 1996). These standardised instruments can be referred to as patient-based or patient-reported outcome measures (PROMs; the terminology that will be used in this thesis). It can be argued that PROMs using standardised structures, by their very nature, may fail to take into account the specific priorities of individuals (Carr & Higginson, 2001; Donovan, Frankel, & Eyles, 1993). However, PROMs that are developed with relevant patient groups may be viewed as sufficiently patient-centred to be an acceptable compromise for the practical requirement of facilitating group comparisons in research trials (Fitzpatrick et al., 2006; Long & Dixon, 1996).

1.7 Consideration points when using PROMs

PROMs are increasingly being advocated by the Department of Health to improve information available on the effectiveness of treatments (Department of Health, 2008a). The use of PROMs to gain patient perspectives on the perceived effect of a rehabilitation intervention may be particularly important when dealing with chronic conditions - such as residual cognitive difficulties after stroke. This is because rehabilitation in chronic conditions tends to focus on optimising function to improve outcomes related to the social, psychological and emotional impact of conditions; aspects that users are a priori best placed to comment on (Bowling, 2005; Fitzpatrick, et al., 2006; Meldahl, Acaster, & Hayes, 2012). Some general considerations about PROMs that are relevant when considering their use with cognitively-impaired individuals are presently outlined and their implications summarised.

1.7.1 The cognitive demands of answering questions

Streiner and Norman (2008) highlight the cognitive processes required when answering questions (including those included in a PROM) for any individual: understanding the question relies on interpretation of the wording and syntax used in the question; recalling the relevant attitude, behaviour or belief to answer the question relies on memory functions; responding using pre-defined structures relies on understanding and interpreting the response categories available (see 1.7.5 for implications).

1.7.2 Self-awareness and masking

As highlighted in section 1.1 above, cognitive impairment is often associated with reduced awareness about the nature and severity of issues. Even in the absence of actual reduced insight into issues, people with neuropsychological impairment may attempt to 'mask' the severity of their issues (Carlsson, Moller, & Blomstrand, 2004; Lincoln, 2012). This can lead to concerns about how far patients are able and/or willing to reliably report on the impact of their illness and complete PROMs (Brooks et al., 1990; Riemsma et al., 2000; Sbordone, Seyranian, & Ruff, 1998).

1.7.3 The effect of mood states

There is evidence to suggest that an individual's transient mood state has an influence on how they evaluate their overall health status and other important aspects of their lives when questioned (Duncan et al., 1999; Isen, 1984; Salovey & Birnbaum, 1989; Svendsen et al., 2012). Positive mood states may bias positively whilst negative mood can bias judgements in less predictable ways; ultimately meaning that transient mood states may influence the test-retest reliability of PROMs (Hanita, 2000). This could be relevant for any PROM but is particularly important to consider here, given the close relationship of mood and cognition (see 1.1 above).

1.7.4 Relationship of subjective cognitive difficulties with objective measures

Whilst some studies have shown that individuals with perceived or subjective cognitive difficulties are also impaired according to more objective neuropsychological tests (Davis et al., 1995; Lincoln & Tinson, 1989; Wendel et al., 2008), other studies have not demonstrated meaningful correlations between subjectively reported cognitive difficulties and observable or measureable cognitive impairments (Aben et al., 2011; Horner, Harvey, & Denier, 1999; Lamb et al., 2013; Prouteau et al., 2004). This has implications for the ways in which PROM data might be clinically interpreted (see 1.7.5).

1.7.5 Summary of PROMs

There are issues with PROMs that mean careful consideration must be given to their development, use and interpretation. PROMs provide a subjective measure of impact that is relevant to the individual functioning within the context of their personal environment; completion relies on their ability to comprehend and answer questions. There are areas in which bias may be introduced in respondents' answers. The lack of a clear relationship between objective cognitive impairment and subjective cognitive difficulty emphasises that PROMs should not be used as a substitute or shortcut for clinical assessment; PROMs provide a different perspective to that of the clinician and consideration must be given to whether one or both are required in any given context.

Many PROMs have not been designed for use with cognitively-impaired participants (Riemsma, et al., 2000) and moreover, people with cognitive impairments have been routinely and systematically excluded from participation in the development of PROMs (Dawson et al., 2010; Fitzpatrick, et al., 1998). If PROMs are to be used, alongside other measures of outcome, to evaluate the effectiveness of interventions for cognitive rehabilitation, it is particularly important that they are developed for and with this patient group.

1.8 Principles of PROM development

Streiner and Norman (2008) highlight the almost paradoxical situation in the general landscape of health measurement tools that there are simultaneously too many and not enough. That said, one of the first stages in tool development is to identify and critically appraise existing tools. Several high-level documents now exist that discuss review criteria when appraising tools (Fitzpatrick, et al., 1998; Scientific Advisory Committee of the Medical Outcomes Trust, 2002; Streiner & Norman, 2008). By logical extension these criteria inform the development of new tools; should a review of existing tools highlight a need.

The contextual reasons *why* measurement is required and *what* measurement is attempting to achieve will inform the relative importance of different review and psychometric criteria; a trade-off between criteria is often required (Mead & Bower, 2000). This may be particularly relevant in a trial, when there are specific trial questions and ultimately a judgement required to determine the fit between these questions and the content of available instruments (Fitzpatrick, et al., 1998). As Wade (1992) points out:

> "*Validity and reliability are not necessarily absolute but relative. One thing is often overlooked – will the measure provide the information needed? This depends upon formulating a clear-cut question at the outset.*"

Discussing properties of a measurement tool therefore requires a context. The information provided in sections 1.1 to 1.7 has been included to provide some context. Specifically:

- There is a need for high quality trials on the topic of cognitive rehabilitation to improve the evidence base and inform clinical guidelines;
- There are a variety of rehabilitation approaches to address every level of disease impact described by the WHO ICF (WHO, 2001). All are designed with the aim of improving function / reducing the impact of health problems and their effectiveness should be assessed with respect to those aims;
- More research is required to improve the lives of stroke survivors in the chronic phase post-stroke and the impact of cognitive and communication issues appears to be a priority for these individuals;
- There is also an argument that trials should include comprehensive interventions suitable for people who have difficulties in more than one cognitive domain;
- PROMs provide an insight into patient perspectives on outcome that, whilst not without their limitations, are often overlooked in stroke trials and arguably should be included in future trials.

To summarise, a patient-centred PROM would be a worthwhile outcome measure to include in trials of comprehensive cognitive rehabilitation in stroke. There are properties that are important to consider when evaluating or developing a PROM to be used for this purpose and these are overviewed presently followed by a rationale for the studies carried out within this thesis.

1.8.1 Acceptability and feasibility

Acceptability refers to how acceptable target users find the measure to complete and is an essential requirement of a PROM - some have argued it is the most important feature (Fitzpatrick, et al., 1998; Gibbons & Fitzpatrick, 2012). If a PROM is not acceptable it can impact data quality and, more importantly, might potentially lead to patient distress. Acceptability is often inferred quantitatively with some general consensus on assessment criteria (Fitzpatrick, et al., 2006; Hilari et al., 2003; Long et al., 2008): there should be less than 10% missing data; there should be no items with more than 80% endorsement at the top or bottom ends of the response scale (floor/ceiling effects); and 75% of items should not have skewed responses. Other aspects to consider when assessing acceptability might relate to the reading and comprehension level of items and the time taken to complete (Scientific Advisory Committee of the Medical Outcomes Trust, 2002). Acceptability should also be assessed directly by seeking

patient views on questionnaires and their items at early stages in the development and testing of a measure (Fitzpatrick, et al., 1998).

Feasibility is similar to acceptability but refers to the burden or difficulty from the perspectives of the administrator, as opposed to the respondent (Fitzpatrick, et al., 2006; Scientific Advisory Committee of the Medical Outcomes Trust, 2002). Time to complete and process as well as any training required for administrators will influence how feasible the measure is to administer within different contexts. Mode of administration is also an aspect to consider in this regard; with face to face administration being more time- and resource-consuming than postal or telephone formats (Scientific Advisory Committee of the Medical Outcomes Trust, 2002).

Ultimately, aspects of acceptability and feasibility may be adaptable and how they influence assessment of existing tools and development of new ones is a balancing act. For example, face to face administration is more resource intensive but it may also lead to higher response rates and there may be some influence on the type of information that respondents disclose when questioned in person (Gibbons & Fitzpatrick, 2012; McHorney, 1996). These are all aspects to be balanced within the context of how a measure will be utilised.

1.8.2 **Validity**

Validity refers to how far the instrument measures what it purports to measure. It is not a fixed property of a measure, but is assessed in relation to a specific purpose or setting (Fitzpatrick, et al., 2006; Scientific Advisory Committee of the Medical Outcomes Trust, 2002). Streiner and Norman (2008) highlight how a measure does not inherently 'have' different *types* of validity, but that there are different *processes* of validation that build up a picture of validity within different contexts and scenarios.

Criterion validity is often referred to in the literature (Streiner & Norman, 2008) and involves the measure being compared to a 'gold standard' pre-existing measure. For example, if a new method of measuring body temperature were developed, we might assess criterion validity by comparing its outputs to those from a digital thermometer. However, given the complexity of assessing PROMs and their context-specific meaning, the importance of assessing criterion validity may be moot if no 'gold standard' exists.

In their review on how to evaluate PROMs for use in clinical trials, Fitzpatrick et al (2006) argue that face, content and construct validity are the most relevant and important aspects of validity for the use of PROMs. Face and content validity respectively refer to whether the instrument appears to measure what it intends to and whether it adequately covers the range of aspects within the subject matter. These aspects of PROMs are inspected through examining actual content and development processes so cannot be readily measured statistically (Fitzpatrick, et

al., 2006; Streiner & Norman, 2008). Face validity may be related to acceptability and an important source of evidence is the process used to develop the questionnaire in the first place.

Construct validity can be tested more empirically by exploring quantitative relationships that measures have with one another. This is particularly relevant for PROMS that tend to measure more abstract concepts that cannot be directly observed (Bowling, 2005). It involves hypothesising about how the construct measured in a particular PROM would relate to constructs measured by other tools. There may be other variables that moderate the relationship between two constructs so construct validity is not always straightforward to assess; if hypothesised relationships are not shown, this could be as much a problem with the hypothesis as it is with the measurement tool(s) (Streiner & Norman, 2008). The process of confidently understanding the validity of any tool, including PROMs requires a build-up of evidence.

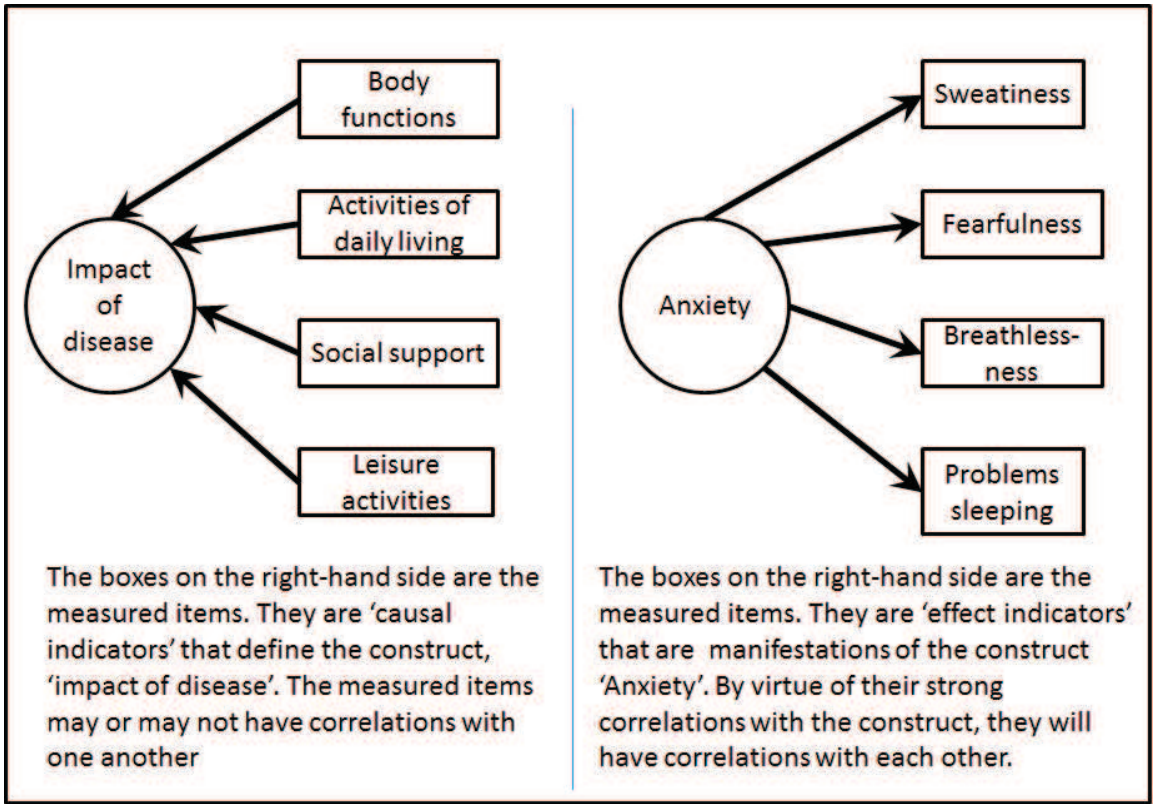
1.8.3 **Reliability**

Reliability is a property that is concerned with whether an instrument produces results that are reproducible, internally consistent and free from error as far as possible (Fitzpatrick, et al., 1998). It is a complex property, but one that is appealing to test developers given that it can be explored more empirically than validity. However, reliability is again not a fixed property of a particular measure, but depends on the context in which it is used, including the characteristics of respondents using it (Streiner & Norman, 2008; Wade, 1992).

Internal consistency of a scale is one means of assessing reliability and it relates to how far the items within a tool are measuring a similar construct. Cronbach's alpha is one of the most commonly used statistics to denote internal consistency and the Scientific Advisory Committee (2002) advise that reliability coefficients should exceed 0.70 for group comparisons and 0.90 for individual comparisons. This is because the degree of reliability required in order to compare groups is lower than that required to assess and compare individuals. These are thresholds that are endorsed by Fitzpatrick et al (1998) but the authors also highlight that employing statistical thresholds indiscriminately can lead to absurdities since, for example, a high internal consistency is achieved by a tool that contains virtually identical items and is therefore of little value. Another important point against the strict implementation of numerical thresholds for internal consistency is discussed by Streiner (2003) and presented pictorially in Figure 1.2 overleaf: PROMs often explore aspects of health where the measured items are chosen because they are thought to collectively define the construct of interest, such as ‘impact of disease.’ In this situation, the measured items can be described as “causal indicators” collectively contributing to the construct of interest, but they may or may not have a

relationship to each other. This is different to situations in which the items are chosen as direct manifestations of the underlying construct, such as the items included in measures of the construct 'anxiety'. These items can be described as “effect indicators” collectively used to describe or demonstrate the underlying construct. The measured items should be correlated with one another and it may or may not be important to include them all: with high inter-correlations, one item could be substituted for another and potentially omitted overall. Figure 1.2 attempts to show the difference between these two situations.

Figure 1.2: different relationships between items and constructs determine the relative importance of internal consistency, as described by Streiner (2003)



If a measure includes items that are chosen for defining the construct without necessarily being correlated with one another, internal consistency would legitimately be lower than when effect indicators are selected. It is important therefore to consider the appropriateness of established thresholds for measures of internal consistency (Streiner, 2003).

Internal consistency is also closely linked with unidimensionality. In the example shown in figure 1.2, anxiety is used as an example of a unidimensional construct that manifests intercorrelated and internally consistent measureable items. However, the example of anxiety may itself be more complex than shown. If a theoretical model of anxiety proposes different components (e.g. cognitive and behavioural (as per Hamilton (1959)) then it may itself be multi-dimensional and conceptualised as having causal indicators, as per the ‘impact of disease’ example. Unidimensionality is achieved if all the items are tapping different aspects of the same

underlying trait and not different parts of different traits (Streiner & Norman, 2008). Unidimensionality is most commonly tested statistically through factor analysis; a method to describe the variability in scores among a number of measured items through identifying a smaller number of factors. These factors are statistically calculated so that the first factor accounts for the maximum amount of variability in the scores across a sample of participants who have provided data on the measure and its items (Streiner & Norman, 2008, p. 409-414). Like most of the psychometric qualities described thus far, internal consistency and unidimensionality are not straight-forward concepts. As per the example of anxiety described in Figure 1.2, it can be conceptualised as a single attribute or as one that has different components that might still be intercorrelated and may still demonstrate unidimensionality. Streiner and Norman (2008) point out that demonstrating unidimensionality is "not an all or nothing concept." : some researchers have suggested that if the first statistically identified 'factor' accounts for at least 20% of the total variance of scores, it can be considered unidimensional; others have suggested 40% as a threshold; still others have suggested that it is the ratio of variances between identified factors that is important. When discussing data analysis that relies on assumptions of scale unidimensionality, Hill et al (2007) show that data can appear unidimensional in one sample but multi-dimensional in another. They give advice that "it is less important for the [analysis] model to be perfect than it is for it to be useful" (Hill, et al., 2007).

Reproducibility is another aspect of reliability and it is assessed in test-retest paradigms to see whether an instrument gives the same results in different administrations if there are no underlying changes in the respondent (Bowling, 2005). There is again room for error here though; the assumption that there are no underlying changes needs to be assessed. Streiner and Norman (2008) advocate the importance of considering the length of time between administrations when considering underlying change. For example, individuals with chronic impairment are assumed to be more stable than those in the acute phase post-injury and therefore underlying change may be less likely to occur over short time spans. Fitzpatrick et al (1998) also suggest a means of checking underlying changes would be to simply ask respondents at second administration. Test-retest reliability is typically examined using correlation coefficients and a minimum standard of 0.70 is advocated (Fitzpatrick, et al., 1998; Streiner & Norman, 2008). Low correlations may mean that the test is unreliable or that the test is reliable but the construct being measured has changed. Streiner and Norman (2008) point out that the act of measurement itself may influence results at a second administration: by prompting individuals to think about the construct being measured; or by sensitising respondents to issues. As well as checking correlations between two administrations, Bland and Altman (1986) also advocate the importance of looking beyond correlation coefficients (influenced by sample size and data variability) and assessing actual agreement between scores. To generate Bland-Altman

plots, the mean score for a participant across two administrations is plotted on the X-axis of a graph, with the difference between scores for that participant plotted on the Y-axis. These plots visually show outliers as well as any patterns of differences between the measurements (e.g. those who score highly on first administration consistently scoring lower on second administration).

1.8.4 Sensitivity to change

Sensitivity to change refers to the ability of a measurement tool to pick up meaningful change in underlying health states when they have occurred (Fitzpatrick, et al., 1998) e.g. after an elective rehabilitation intervention. Whilst it is included as a property in its own right, it is actually part and parcel of validity and has a relationship with reliability (Streiner & Norman, 2008). On a conceptual level, a measurement tool would a priori detect change in a context if there was confidence in its reliability and validity within that context. Both the Scientific Advisory Committee (2002) and Fitzpatrick et al (1998) highlight that, in order to critically appraise tools against this criterion, there are no strict rules of what statistical methods should be used or what thresholds should be met for analyses. Rather, there should be discussion about this property demonstrating evidence that a) a change of importance has occurred in respondents; and b) a corresponding change has occurred within the measurement score.

1.9 Summary & rationale for studies

This introduction has attempted to make the case that there is a need for trials that assess the effectiveness of rehabilitation interventions addressing multiple persisting cognitive difficulties after stroke; and that these trials would usefully include a patient-centred PROM as an outcome.

In order to consider what is required of a PROM, some core psychometric properties of PROMs have been discussed in section 1.8. Care has been taken to emphasise that each property is complex and context-specific and cannot be categorically judged by reference to empirical data alone. As Fitzpatrick et al (1998) state, critical appraisal of a PROM, "remains to some extent a matter of judgement and as much an art as a science."

A review of existing PROMs and, if required, the development of a new PROM, would usefully start by understanding service users' views on important aspects that should be included in tools. This would guide the "art" of critical appraisal and would build on gaps in the qualitative and PROM development literature that suggest that people with cognitive difficulties are under-represented in these fields (see 1.2 and 1.7.5).

1.10 Aims and objectives of the studies in this thesis

Overall, the purpose of the research described in this thesis was to identify a patient-centred, patient reported outcome measure (PROM) for trials of comprehensive cognitive rehabilitation in stroke.

This was achieved in stages:

The first stage (Study 1: Qualitative Study, from page 44) was to understand service user perspectives on the important and measureable impacts of persisting cognitive problems. This was to generate requirements for a PROM that could influence both a critical appraisal of existing tools and, if necessary, the development of a new tool.

In the next stage (Study 2: Systematic Review of PROMs, from page 62), the aim was to identify whether any existing tools, perhaps from other neurological conditions, satisfied the requirements for a PROM derived from the first stage. No tool did fully meet the user-defined recommendations.

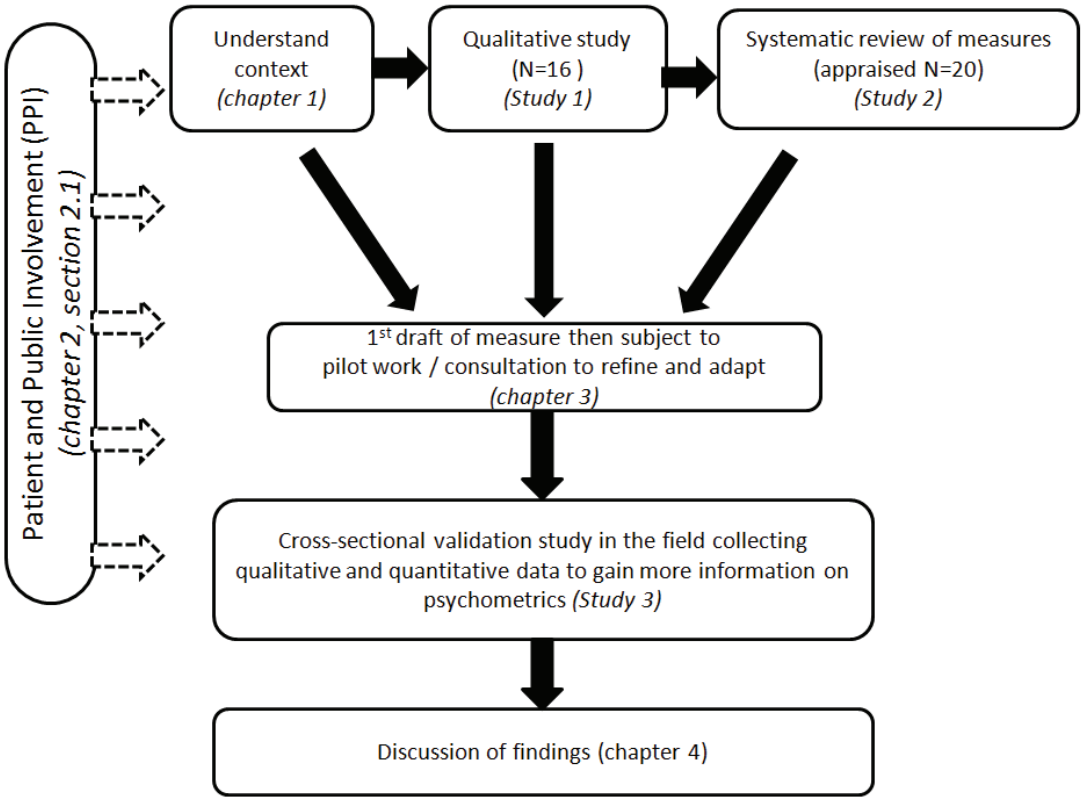
Chapter 3 (Tool development, from page 78) then describes the process of developing a new PROM to meet user-defined recommendations that utilised feedback from different stakeholders.

Finally, the new tool was field tested in a large sample in order to gain quantitative and qualitative data on its acceptability, feasibility and other psychometric properties (Study 3: Psychometric Study, from page 90).

2 Research Methodology

This chapter provides rationale for the methodologies that were used to meet the aims of the research and identify a patient-centred, patient reported outcome measure (PROM) for trials of comprehensive cognitive rehabilitation after stroke. A sequential process was utilised in this thesis; where results of one study influenced questions asked and methods used in subsequent studies. Creswell at al. (2004) describes an 'instrument development design' as an exploratory sequential design utilising both qualitative and quantitative methods in a mixed approach that strengthens, rather than divides, enquiry. As well as the specific quantitative and qualitative studies that were carried out in different stages of the research, there was an active partnership with stroke survivors and their carers utilised throughout all stages. Figure 2.1 shows how the different studies and methodologies form the thesis.

Figure 2.1 a representation of this thesis



The patient and public involvement (PPI) that fed into various aspects of the research is described first in this chapter. Participant selection and ethical considerations relevant to primary data collection studies are then described. The methodological approaches used in each of the studies / chapters are then described at a level of providing a rationale for the methodology used. The specific methods employed and the results of studies are the remit of each individual chapter.

2.1 Patient and Public Involvement (PPI)

INVOLVE (www.invo.org.uk) is a UK national organisation that promotes and supports PPI in the NHS and in health research. INVOLVE explain PPI in research as a process performing research 'with' or 'by' members of the public; often those with experience of the condition under investigation. Members of the public are actively involved in the research process as opposed to being participants in primary data collection (INVOLVE, 2012). INVOLVE outline three approaches to involving users that often overlap: consultation (involving asking for views to inform decision-making); collaboration (involving partnerships where decision-making is shared); and user-controlled research where users actively direct and manage research. These approaches can be utilised at all stages in the research cycle: from identifying and prioritising research topics through to monitoring and evaluating research in practise (INVOLVE, 2012).

Patients and the public provide a unique perspective and can help improve many aspects of research including: ensuring the methods used are suited to target participants; creating accessible materials; improving credibility of findings (Staley, 2009). Systematic reviews to assess the effectiveness of PPI in improving research have highlighted an absence of robust evidence (Boote, Telford, & Cooper, 2002; Mockford et al., 2012). However, it is important to acknowledge that a lack of evidence does not mean a lack of effectiveness. The challenges of assessing impact formally (e.g. a lack of tools and funds to do so), are likely contributors to the lack of evidence (Barber et al., 2012; Mockford, et al., 2012). A multi-disciplinary team of researchers and service users have recently developed the Public Involvement Assessment Framework (PiiAF) (Popay, Collins, & PiiAF study group, 2013); a means of exploring and assessing impact of PPI. However, even with the right tools to assess impact, it is not universally accepted that PPI is beneficial for research: resistance to the use of PPI tends to be related to the time, cost and training implications as well as an underlying concern that PPI diminishes the control or influence of researchers (Beresford, 2007; Boote, et al., 2002; Ong & Wood, 2005). However, despite these pockets of resistance, the value of well-structured PPI has been shown in stroke studies (Boote et al., 2015; Boote et al., 2014) and involving users in research is seen as a mark of quality (Department of Health, 2005b). There is also anecdotal evidence of positive impact of PPI (Barber, et al., 2012) which is supported on a personal level from my own perspective; having previous experience working closely with the Research User Group (RUG) of the ACT NoW study (Assessing Communication Therapy in the North West (Bowen et al., 2012)). The ACT NoW RUG was a group of service users including stroke survivors with communication problems and their carers who met regularly to shape many aspects of this complex, pragmatic trial including: training researchers; developing recruitment materials; and developing the Communication Outcomes after Stroke (COAST) scale (Long, et al., 2008).

One single user group was not utilised within this PhD. In the first instance, World Stroke Day in October 2011 (shortly after this PhD was started) gave me the opportunity to gain advice from an expert patient adviser on the direction of the research. It was suggested that, once I had a clearer idea of the direction I was likely to take, I would be welcome to attend the regular meetings of the community stroke group that this expert patient adviser was also a member of. As the direction of the research evolved, PPI was utilised at discrete stages of the study and would therefore primarily fall under INVOLVE description of 'consultation' and also aspects of 'collaboration', since decision-making was shared (INVOLVE, 2012). The consultation and collaboration supported: decision-making; designing methods and materials; and interpretation of findings. This was particularly pertinent for: Study 1: Qualitative Study; the developmental stages of the tool (see chapter 3: Tool development), as well as to inform the protocol and patient information and consent materials used in Study 3: Psychometric Study.

2.2 Study Participants

For the primary data collection studies it was important to recruit participants who adequately represented the target population for the measure, to help ensure patient-centredness (Fitzpatrick, et al., 2006; Long & Dixon, 1996).

This research was particularly interested in adults (18 years plus) at the stage in the clinical pathway that is often called ‘life after stroke' (see Introduction section 1.3). Spontaneous recovery after stroke is generally thought to occur within the first three months (Robertson & Murre, 1999), although cognitive deficits can demonstrate spontaneous gains ongoing to six months (Cramer, 2008) and recovery can be considered ongoing for years. 'Life after stroke' encompasses attempting to return to life activities and participation after a stroke and learning to accept and adjust to residual disabilities. For the purpose of this thesis, stroke survivors were considered as being in the 'life after stroke' stage of the clinical pathway if they were a minimum six months post stroke and living in community settings.

Individuals who are months or years post-stroke may not be actively involved in treatment and so identifying them for recruitment was a challenge. The National clinical guideline for stroke (2012) chapter on longer-term care advises that a healthcare review should take place six months post-stroke to determine whether further rehabilitation interventions are warranted. The Greater Manchester Stroke Assessment Tool (GM-SAT) (Rothwell, et al., 2013) has been designed as a means of standardising the six month review process and ensuring that relevant issues are explored as required. This tool includes specific questions about unmet needs in cognition and so it was a potential means of identifying research participants by targeting individuals who did report needs in these areas and for whom cognitive rehabilitation at this

time would potentially be offered, subject to local service provision (Woodward-Nutt, et al., 2013). Collaborations were forged with teams and individuals carrying out six month reviews in the Greater Manchester localities using the GM-SAT - as well as those providing community-based rehabilitation interventions (e.g. clinical psychologists and occupational therapists) to support recruitment to studies. In those cases, healthcare professionals would provide brief study information to potentially eligible participants and invite them to self-refer to the research team if they were interested in finding out more about the study.

Another means of recruiting stroke survivors was through postal mail outs from General Practitioners (GPs) that utilised stroke registers and invited potentially eligible individuals to self-refer to the research team. Recruitment drives were also carried out at stroke community groups across Greater Manchester, including those run by the Stroke Association and voluntary support groups. The purpose of relevant study for recruitment was outlined initially at a recruitment drive. Members could then approach the researcher to volunteer as potential research participants if they wished. In addition, brief information leaflets about the study could be left with groups for members to peruse and self-refer in their own time. These recruitment methods are critiqued in the final discussion chapter four (e.g. see discussion section 4.1.4).

Another core requirement for selection of stroke survivors was the need to recruit those with cognitive and communication difficulties. As discussed in the introductory sections of this thesis (see 1.2 and 1.7.4), there is not necessarily a correlation between measureable cognitive impairment and subjective reports of difficulty in function due to cognition. The important factor here is that individuals who self-report cognitive difficulties are likely to be end users of any developed tool since they are likely to seek support for their difficulties. As such, primary means of assessing difficulty due to cognition was based on self-report: stroke survivors were recruited if they felt they had unmet needs in cognition and communication and that their daily lives and functional abilities were compromised because of these issues. This approach meant that those with serious self-awareness issues who did not acknowledge their difficulties at all were less likely to be recruited as they might not self-refer. Conversely, participants might perform at a level to 'pass' cognitive assessments but still be eligible if they reported difficulty. Cognitive assessments were still useful as a means of describing the sample and, for the psychometric study, to explore relationships/lack of relationships between assessment scores and perceived impact ratings. A full in-depth neuropsychological assessment requires experts such as neuropsychologists and can be long and tiring for stroke survivors who may fatigue easily (Lezak, 2012). As such, the main means of describing cognitive difficulties objectively was through screening procedures that could be reasonably carried out within the context of a home visit for data collection with a non-expert researcher. The screening tools used to assess

cognition are described in relevant study methodologies (see Study 1: Qualitative Study and Study 3: Psychometric Study) and, as mentioned, participants were *not* excluded on the basis of threshold scores from these cognitive screening tools (i.e. they were included if they passed screening tests as long as they self-reported cognitive difficulties).

2.2.1 A summary of inclusion criteria for participants in Study 1 and Study 3

- Adult (18 years plus), at least 6 months post-stroke;
- Living in community settings;
- With self-reported cognitive difficulties;
- An ability to understand and communicate in the language of English, given that the interviews were to be conducted in English and the tool was to be developed in the English language. All forms of communication were encouraged to facilitate the inclusion of people with aphasia or dysarthria;
- Capacity to give informed consent:
  - Participants were facilitated to provide this; the researcher had experience supporting the understanding of people with stroke and severe cognitive problems. Cognitive problems could potentially influence capacity but, as per the Mental Capacity Act Code of Practice (Office of the Public Guardian, 2007), these individuals were presumed able to make a decision unless it could be established that they lacked capacity. As per the recommendations of the code of practice: the study information and consent forms were available in accessible 'aphasia friendly' formats; a significant other was consulted where available regarding the likely preferences of the service user; and there was at least 24 hours built in to the consent procedure to ask the service user questions to verify their ability to retain information and weigh up pros and cons to make a decision.

2.3 Ethics

NHS Research Ethics Service approval and R&D approval at Participant Identification Centres (PIC) sites was obtained for the primary data collection studies (Study 1: Qualitative Study and Study 3: Psychometric Study). Studies were conducted in accordance with the British Psychological Society (2010) guidelines regarding the protection and welfare of participants; informed consent; right to withdraw; and confidentiality. Good Clinical Practice training was undertaken by researchers (www.gcp.epigeum.com).

Primary data collection involved interviews and questionnaire completion / assessment and there were no direct risks from taking part. However, participants were engaging with the

research in the context of illness and potential vulnerability and may become upset when recalling and talking about their experiences. Considerations were made about the potential for studies to cause emotional distress to participants or reveal unmet needs. Links were forged with stroke community groups and NHS community rehabilitation services across Greater Manchester to allow referrals to be made to these services when unmet and distressing needs were disclosed by research participants. Participants' GPs were not informed when individuals were recruited to studies as standard. However, participants were sometimes recommended to make appointments with their GP after taking part. In these cases, participants had the option to consent to researchers sending a letter to their GP; confirming their participation in the study and that a recommendation had been made to book an appointment. Procedures were also in place if participants made criminal or other disclosures requiring action without consent. If disclosures occurred, the researcher would discuss with the participant in the first instance and disclose the duty to share information. Discussion with supervisors would determine the appropriate course of action.

2.4 Study 1: Qualitative Study

Published in Health Expectations (Patchick et al., 2014), the aim of this study was to understand service user perspectives on the important and measureable impacts of persisting cognitive problems. This was to generate requirements for a PROM, which could influence both a critical appraisal of existing tools and, if necessary, the development of a new tool. Qualitative methods were used for this study as they take a subjective approach using non-numerical data to describe, interpret and understand lived experiences and perspectives (Peters, 2010).

Qualitative data collection methods that require engagement from the participants (i.e. not based on observation alone) are collected through either in-depth interviews or focus groups. Interviews are preferred in the context of a safe, private environment for generating personal accounts in order to explore details of complex processes such as impacts (Brédart et al., 2014; Ritchie & Lewis, 2003). They are particularly useful for individuals who may have difficulty travelling to a group location due to disabilities and if people have cognitive difficulties that may hinder their ability to contribute in a group context (Ritchie & Lewis, 2003). PPI with community stroke groups also advised that if the goal is to explore the impact of cognitive problems and encourage patients to think about ways in which rehabilitation services should be evaluated, in-depth qualitative interviews should be carried out.

Supporting communication and expression is an important pre-requisite for gaining useable and useful data through interviews (Brédart, et al., 2014). PPI led to agreed definitions and pictorial aids for cognition and the interview schedule (see Appendix 2 from page 161). Interview

questions can be on a spectrum from open (e.g. “can you tell me about ..”) to closed (e.g. “when did you have a stroke?”). A mixture gives flexibility between gaining patient stories in their own words but also keeping the interview on track and moving it forward (Ritchie & Lewis, 2003). Closed questions, using prompts and cues are also useful with individuals who may tire easily or have difficulty expressing themselves (Brédart, et al., 2014). A semi-structured approach to interview was informed by PPI using a flexible set of open and closed questions to explore: how cognitive issues affect individuals; how this changes over time; and how change or effect/impact might be measured.

2.4.1 Sampling

Qualitative research does not seek to include a large, representative sample of a population (Ritchie & Lewis, 2003). Instead, purposive sampling targets recruitment of individuals who have specific characteristics that might influence their responses to address the research aims appropriately (Patton, 2002; Peters, 2010). Purposive sampling strategy is described in Study 1: Qualitative Study. The number of people included was driven by data saturation; when a point is reached where researchers are no longer hearing or seeing new information (Ritchie & Lewis, 2003). Data saturation is a tricky concept as it depends on a number of variables including: the complexity of data (influenced by the sampling strategy); the experience of the researcher(s); the number of individuals identifying themes (which may influence how quickly consensus is reached) (Guest, Bunce, & Johnson, 2006). Guest (2006) explored a dataset of 60 interviews and found that after 12 interviews, almost all of the themes that were reported had already been identified and they suggested that meaningful over-arching themes and interpretations were possible to identify after just the first six interviews. Given the broad variety of cognitive issues and the complex nature of impact we wished to explore in interviews, we anticipated a sample size of around 20 to 25 participants would be desirable and achievable.

2.4.2 Analysis

There are different approaches to analysis of qualitative data and the choice of analysis will be influenced by the approach to sampling and interview (Ritchie & Lewis, 2003). Peters (2010) describes the role of qualitative research within mental health research and her examples influenced the analysis approach utilised in this PhD. Peters (2010) describes measurement tool development and highlights how “rich qualitative data provide terminology that is meaningful to service users that can then be incorporated into question items” (Peters, 2010). Basic content analysis in this PhD study therefore identified terminology that was commonly used by patients

and would inform actual wording of items. The content analysis involved utilising the powerful analysis tools inbuilt to NVivo, a qualitative software analysis package used in this research.

In addition Peters (2010) also describes how qualitative research can be useful for identifying overarching core themes of importance to patients that inform dimensions of a measurement tool. In line with this, the analysis approach in this study was a thematic framework analysis that is championed by Ritchie (2003) and is a means of identifying, synthesising and organising data in a heterogeneous sample using a structured chart showing key themes, concepts and categories. The broad conceptual understanding of the WHO ICF model of impact of a health condition (2001) (see section 1.2) did influence the framework of analysis by providing a conceptual groundwork e.g. understanding 'contextual factors' as a potential theme. However, the framework analysis was ultimately data driven and the methods to achieve framework analysis are described in Study 1: Qualitative Study. The analysis of the qualitative data led to recommendations for a PROM for trials of cognitive rehabilitation after stroke.

2.5 Study 2: Systematic Review of PROMs

In this study, the aim was to identify whether any existing tools, perhaps from other neurological conditions, satisfied the recommendations for a PROM derived from the qualitative interviews and might be used in stroke care and research.

As described in introductory chapter section 1.8, reviewing existing measurement tools is as much an art as a science (Fitzpatrick, et al., 1998). The context of measurement informs the relative importance of different review criteria and will determine whether a tool is fit for purpose (Mead & Bower, 2000).

2.5.1 Identifying tools

Before PROMs could be reviewed, they needed to first be identified. The Core Outcome Measurements for Effectiveness Trials (COMET) Initiative (www.comet-initiative.org) highlighted during their third annual meeting in Manchester that identifying instruments is challenging, despite the existence of databases that aim to bring them together. A method of identifying instruments to review took some consideration.

In the first year of the PhD, randomised controlled trials (RCTs) and controlled clinical trials (CCTs) of cognitive rehabilitation were identified as a means of extracting outcome measures. This approach was initially chosen as a pragmatic means of identifying tools i.e. if nobody is using the outcome measure in trials, why review it? However, it involved the assumption that researchers carrying out trials would be using rigorous standards to select tools. As set out in

the introduction (e.g. section 1.5), literature suggests that this may not always be the case and trials may not be using outcome measures that were deemed important by service users.

Because of this, an alternative approach to identifying patient-centred PROMs for cognition was subsequently used. This approach asked the question, “what tools are out there?” (as opposed to “what tools are trialists using?”); including literature from other neurological conditions that might identify tools that would be ideal for stroke patients, with some adaptations. The search strategy employed for this approach is described in Study 2: Systematic Review of PROMs.

2.5.2 Appraising tools

Guidance for conducting critical appraisal of PROMs is provided by the Scientific Advisory Committee of the Medical Outcomes Trust (2002), the Patient-reported Outcome Measurement Group (Jenkinson, Gibbons, & Fitzpatrick, 2009) and Fitzpatrick et al (2006). These documents break down appraisal of tools into review criteria that cover psychometric properties of: conceptual development; appropriateness and acceptability to users; reliability; validity; precision and sensitivity to change; interpretability; feasibility and administrative burden.

As highlighted in introductory section 1.8 ‘Principles of PROM development’, the relative importance of psychometric properties in review criteria is determined by contextual reasons *why* measurement is required and *what* measurement is attempting to achieve.

Fitzpatrick et al (2006) recommend that face and content validity may be the most relevant aspects of validity for the use of PROMs and, "need to be inspected, literally by examining the questionnaire." This was an important first stage of the tool review process.

The Study 1: Qualitative Study (Patchick, et al., 2014) had generated recommendations for a PROM. These recommendations related to conceptual underpinning of a tool and aspects of face and content validity and, as such, became review criteria against which identified PROMs were first assessed. The assessment was achieved by examination of the tool content and development papers that described what the tools were attempting to achieve in measurement. The method by which the qualitative recommendations became review criteria are described in Study 2: Systematic Review of PROMs.

The means of assessing other psychometric properties such as reliability and construct validity are also described in study 2 and were informed by the guidance documents identified and mentioned above (Fitzpatrick, et al., 1998; Jenkinson, et al., 2009; Scientific Advisory Committee of the Medical Outcomes Trust, 2002).

2.6 Pilot work / Tool development (Chapter 3)

As no appropriate tool was found in the review, the aim of this pilot test stage was to develop and refine a new tool to establish: content and number of items; formatting; initial data on acceptability and interpretability.

The process of developing the tool relied on synthesising all previous work described up to this point as well as utilising PPI and consultation with healthcare professionals and researchers to make refinements and improvement. The process is described in detail in chapter 3 and a critique of the methodology used is given in discussion chapter section 4.3.4.

The result of this work was the Patient Reported Evaluation of Cognitive State (PRECiS) scale that was tested psychometrically. It is described in detail in chapter 3 section 3.11 and included as an appendix to this thesis (appendix 5 from page 170) . PRECiS asked respondents to rate 'bother' associated with any limitations due to cognition.

2.7 Study 3: Psychometric Study

The aim of the psychometric study was to gain further qualitative feedback on PRECiS plus quantitative information on psychometric qualities of the measure in a large sample. This would support finalising the content and format plus any associated manuals.

2.7.1 Collecting data

Quantitative and qualitative data were collected in a mixed methods study. O'Cathain, Murphy and Nicholl (2007) reviewed the use of mixed methodologies in health research and found that there has been a surge in use of this method. They suggest this method capitalises on the strengths of both approaches to enrich one another and help capture the complex nature of health research (O'Cathain, et al., 2007).

A cross-sectional field study was used as it allowed the opportunity to recruit a large number and broad range of stroke survivors. It was beyond the scope of this thesis to devise and implement a cognitive rehabilitation intervention that might help explore whether PRECiS was able to detect change if change had occurred. Sensitivity to change was not an attribute that was to be explored in this study.

2.7.2 Modes of administration

As well as face-to-face, other popular modes of administering tests include telephone, postal and using email / the web. Each method of administration has advantages and disadvantages.

A face-to-face approach ensures you are speaking to the correct person and can verify that they have completed the tool. This is not necessarily possible with other methods of administration such as postal completion (Streiner & Norman, 2008). Face-to-face personal interaction is particularly useful for gaining feedback and identifying areas of misunderstanding that might otherwise be glossed over or left as missing responses if sent by post or if completing by computer. There is also a flexibility afforded through face-to-face administration that can inform how items and/or an administrator guide might be improved (Streiner & Norman, 2008). This may be particularly important in a heterogeneous group of stroke survivors with cognitive issues that impact their ability to process information and/or communicate. A face-to-face approach means that participants can be offered as much support as required.

In qualitative research, the researcher is recognised as an instrument influencing the data collection process, with the ability to influence and potentially bias results (Ritchie & Lewis, 2003). Face-to-face administration of a questionnaire is also potentially open to this aspect of bias or inconsistency in the data collected. However, bias in the sample is also possible with other administrations such as postal surveys where a particular attribute e.g. cognitive difficulty, may mean a person is less likely to respond and send back a questionnaire. A primary disadvantage of face-to-face administration of a tool is the time and cost involved; which can potentially impact the feasibility of using this method. The purpose of this study was to gain a rich dataset of quantitative and qualitative information about the tool and to maximise understanding of how respondents use the tool in test settings. As such, the costs of face-to-face administration were deemed worth the return in terms of allowing flexible and supported administration with this patient group.

2.7.3 Sampling

The stroke survivors recruited for this study have been described in section 2.2. There is a lack of consistency in guidelines related to sample sizes for psychometric testing of a scale. Depending on different sources, aspects of the tool being evaluated and specifics of the study, recommendations range from a minimum of 100 to a minimum of 500 respondents (MacCallum & Widaman, 1999). A common rule of thumb is to aim for five respondents per item (Long, et al., 2008). With 27 items making up the PRECiS scale, the target sample size for this study was rounded up to 150.

2.7.4 Exploring Validity

This study was part of the *process* of validation; to start building up a picture of attributes relating to validity, within context (see introductory chapter section 1.8.2).

There was a practical limit on the number of additional measures that could be included in the study; stroke survivors are likely to fatigue easily and it was therefore desirable to ensure visits could be completed within a maximum two hour window as this was more likely to be accommodated within the context of a morning or afternoon home visit. The process of taking consent, carrying out cognitive screening and collecting data on PRECiS including qualitative feedback, was anticipated to last at least one hour. An influential factor when choosing comparison measures was therefore length of the measure and anticipated time to complete.

As described above in section 2.2, the cognitive screens carried out on participants were useful for describing the sample as well as exploring relationships / lack of relationships between assessment scores and perceived ratings. In addition to these cognitive screens, a measure of overall functional impairment was desired to explore the relationship between levels of impairment and amount of perceived impact. The modified Barthel Index (Collin et al., 1988) was selected as it is one of the most commonly used in stroke trials (Quinn et al., 2009); and shown to have excellent reliability and validity for stroke (Wolfe et al., 1991). It is also simple to administer for a non-expert researcher and was a tool I had some familiarity with from previous research experience.

A simple measure of participants' perceived level of overall independence in function was desired as a means to supplement the researcher-completed Barthel Index. The Nottingham Extended Activities of Daily living (Nottingham EADL) (Nouri & Lincoln, 1987) was specifically developed for use with stroke patients and has been widely validated in the post-acute phase demonstrating excellent reliability and validity (Jenkinson, et al., 2009).

Self-reported measures of cognition often have a significant correlation with low mood (see introductory sections including 1.1). It was therefore important that mood was considered in this study; both for exploring construct validity of the new measures but also as a potential explanatory factor that might influence the relationship between measurable impairment and perceived impact. A systematic review of tools available for mood screening in stroke (Burton & Tyson, 2015) supported decision-making regarding tools to use for these purposes. The Patient Health Questionnaire (PHQ9) (Spitzer, Kroenke, & Williams, 1999) performed well in this review: it had good psychometric properties; was quick and straight-forward to administer; and is freely available for use in research. The PHQ9 also shows evidence of utility with stroke patients in community settings (Lincoln, 2012) and is a recommended tool for screening depression in the Greater Manchester Assessment of Stroke Rehabilitation (G-MASTER) toolkit (Tyson, Burton, & McGovern, 2014). The Generalised Anxiety Disorder (GAD7) (Spitzer et al., 2006) is also recommended in the G-MASTER toolkit (Tyson, et al., 2014) and is often used alongside the PHQ9 as a mood measurement. It is also the tool recommended by the Improving Access to

Psychological Therapist (IAPT) services (Clark & Oates, 2014) and so was chosen for this study as one of the mood assessment tools.

As well as functional ability and mood, a measure of impact was desired as a means of assessing PRECiS scores compared to an existing, validated test of impact. The review of PROMS (Study 2: Systematic Review of PROMs) did not identify any tools that assessed impact of cognition directly (hence the need to develop PRECiS) and so a measurement of overall stroke impact was desired for criterion validity. Whilst it would be hard to argue that there is a 'gold standard' for measuring stroke impact, the Oxford Patient Reported Outcomes Measurement Group (Jenkinson, et al., 2009) have critically appraised many stroke-specific PROMs and recommend the Stroke Impact Scale (SIS) (Duncan, et al., 1999) as it showed good psychometric properties including low floor/ceiling effects. The SIS was therefore selected for exploring validity.

2.7.5 Exploring Reliability

Conceptual and statistical approaches to reliability that were set out in introductory section 1.8.3 were explored within this study and the methodology used is described in Study 3: Psychometric Study.

Whilst the study process and methodology are described in Study 3: Psychometric Study, more detail is given here on the assessment of test-retest reliability as it is only briefly covered in study 3 write up due to word limitations. The purpose of this study was to gain preliminary data on these qualities and whilst a dataset with second visits for all participants might have been desirable, it was not necessary for statistical comparisons. Reducing the number of second visits also reduced burden for stroke survivors taking part and was achievable with one data collector visiting individuals in their homes at times to suit them.

An assessment of test-retest reliability was therefore planned with a stratified sub-sample of 60 participants completing the measure on a second occasion. The sub-sample was stratified to carry out second visits with individuals achieving a wide range of scores on the PRECiS scale e.g. some who report little impact up to those who report very high impact. The first 30 participants were automatically included for test-retest visits. The scores achieved by those first 30 participants were used to calculate range of the lowest 10 scores; the highest 10 scores; and the 10 scores in the 'mid-range'. Then, from participant 31 onwards, second visits were arranged with 10 participants who scored in each of the lowest-, mid- and highest- score range (and who agreed to second visits).

Study 1: Qualitative Study

Development of a patient-centred, patient-reported outcome measure (PROM) for post-stroke cognitive rehabilitation: qualitative interviews with stroke survivors to inform design and content.

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Patchick EL, Horne M, Woodward-Nutt K, Vail A, Bowen A. Development of a patient-centred, patient-reported outcome measure (PROM) for post-stroke cognitive rehabilitation: qualitative interviews with stroke survivors to inform design and content. Health Expectations. 2014;doi: 10.1111/hex.12311.

Development of a patient-centred, patient-reported outcome measure (PROM) for post-stroke cognitive rehabilitation: qualitative interviews with stroke survivors to inform design and content.

Abstract:

Background: Improving cognition is service users' top research priority for life after stroke and future research should include outcomes that they deem important. Patient perspectives on outcomes are collected using patient-reported outcome measures (PROMs). There is currently no patient-centred PROM specific for cognitive rehabilitation trials.

Objective: Inform PROM development by exploring stroke-survivor perspectives on the important, measurable impacts of persisting post-stroke cognitive problems.

Design: Qualitative semi-structured interviews in participants' homes.

Participants: Purposive sample of 16 cognitively-impaired stroke-survivors at least six months post-stroke.

Methods: Interviews used a schedule and communication aids developed through patient consultation. Interviews were transcribed verbatim with non-verbal communication recorded using field notes. Data were analysed using a framework approach to find commonalities to shape the focus and content of an outcome measure.

Results: Participants identified important impacts of their "invisible" cognitive problems, outside of other stroke-related impairments. Cognitive problems exacerbated emotional issues and vice versa. Changes in self-identity and social participation were prominent. Impact was not spoken about in terms of frequency but rather in terms of the negative affect associated with problems; terms like "bothered" and "frustration" were often used.

Conclusions: The results support the development of a PROM specifically designed to address the impact of cognitive problems. It should:

- include items addressing a comprehensive range of cognitive skills;
- ask questions about mood, self-identity and social participation;
- use accessible wording that respondents understand and endorse;
- measure impact rather than frequency.
- Explore perceived impact on carers

Keywords: Stroke; Cognition; Patient-centred; Patient-reported outcome measure (PROM); qualitative; psychometrics

Introduction

Persisting post-stroke cognitive problems are common and include issues with attention and concentration; memory; aphasia; unilateral spatial neglect; perception; apraxia; and executive dysfunction[1]. Cognitive problems exacerbate the long-term burden of stroke, adversely impacting confidence, self-esteem and long-term functional recovery[2].

The National clinical guideline for stroke[3] for England and Wales recommends treating cognitive problems comprehensively, but more research is required to inform best-practice interventions. Stroke survivors, caregivers, and health professionals collectively agree that improving cognition is the number one research priority for life after stroke[4]. Cochrane reviews of cognitive rehabilitation trials conclude that future research should use outcomes that are deemed important by service users.[5-6]

Patient perspectives on outcome are often overlooked in stroke trials[7]. Trials need a 'baseline' for individual comparison of outcome and typically use a measure of impairment or function for these purposes. However, the most ecologically valid 'baseline' for assessing change would be pre-morbid performance[8]. These baseline data are rarely available and, by definition, cannot be obtained retrospectively. Given these difficulties obtaining meaningful baseline data, a potential alternative – that is arguably more relevant to service users - is to gain patient perspectives on perceived effect of an intervention. This often involves patient-reported outcome measures (PROMs).

Dawson[9] has discussed the importance of using appropriate, validated PROMs for a given specified purpose (in this case, evaluating a cognitive rehabilitation intervention) but goes on to advise that: "a patient's inability to understand a questionnaire, for reasons of impaired cognition or difficulty with the language in which it is available, should constitute an exclusion criterion." People with cognitive problems that influence understanding and expression are often routinely excluded from participation in the development and use of PROMs: so despite being patient-reported, PROMs are not necessarily appropriate, inclusive or patient-centred[10].

The authors are not aware of a patient-centred PROM that would be suitable for use with cognitively-impaired stroke survivors to evaluate trials of post-stroke comprehensive cognitive rehabilitation. One of the most commonly used PROMs in this area is the Cognitive Failures Questionnaire[11]. The Cognitive Failures Questionnaire uses complicated language, is heavily loaded towards memory issues and, as service users were not involved in its development, it is not a 'patient-centred' measure.

A recent systematic review of stroke literature identified only three patient-centred outcome measures for stroke[12]: The first is the Subjective Index of Physical and Social Outcomes (SIPSO)[13], which measures social integration after stroke. It focuses on the impact of physical functioning and social/emotional functioning for integration, so is not appropriate for cognition. The Stroke Impact Scale (SIS)[14] is a stroke-specific self-report health status measure. However, like other stroke-specific tools (e.g. Stroke-Specific Quality of Life scale[15]), the SIS includes items related to cognition but does not ask about the impact of cognitive function on social participation and quality of life. The third measure is the Communication Outcome After Stroke (COAST) Scale[16]. This tool explores communication effectiveness for those with aphasia (or dysarthria) following stroke as well as the impact of these problems on life and integration. The COAST does not explore other cognitive impairments so, whilst it is useful for aphasia, it would not necessarily be suitable for trials of comprehensive cognitive rehabilitation.

Another critique of existing PROMs is that they may be too long and tiring for patients with stroke who can fatigue easily (fatigue in itself is likely to contribute to reduced scores) e.g. the European Brain Injury Questionnaire[17] and the Stroke Impact Scale[14], with 63 and 60 items respectively.

The authors believe that there is a need for the development of a patient-centred PROM that specifically addresses the impact of a broad range of cognitive problems after stroke and is developed in collaboration with cognitively-impaired stroke survivors.

Aim

The aim was to inform the development of a comprehensive patient-centred PROM for cognition by exploring stroke-survivor perspectives on the important, measurable impacts of persisting post-stroke cognitive problems.

Methods

Ethics approval was granted by the National Research Ethics Service (reference 12/NW/0663).

Patient and Public Involvement (PPI)

To enhance patient-centredness, prior to data collection service users were consulted as research partners to devise methods and materials used in the research. These service users were all stroke survivors or carers who had experience of cognitive problems and were approached via stroke community groups or were previously known to the researchers.

Through PPI, we agreed that semi-structured interviews (with open questions and closed prompts) would be the preferred methodology for cognitively-impaired interviewees, who may need support processing and communicating information. The decision to interview stroke survivors independently of their carers was agreed through PPI as service users felt that a more open and honest dialogue would be achieved one-to-one.

The interview schedule was refined through pilot testing with cognitively-impaired service users as part of the PPI process. This had the added benefit of providing training for researchers.

Communication aids were developed through PPI to support understanding and expression. Lay pictorial representations of cognition were used to orient users to the discussion topics and cue cards used as ramps for communication, if required. Examples of the supporting aids are provided as supplementary materials[i].

Recruitment

Research participants were recruited between September 2012 and January 2013. The research team visited community stroke groups to inform attendees about the research and invite them to participate, if eligible. Additionally, community healthcare professionals treating stroke survivors in the North West of England gave basic information about the study to their eligible service users and invited them to self-refer to the research team for more information.

Participants were included if they were at least 6 months post-stroke with ongoing cognitive impairment. Cognitive impairment was determined predominantly by self-report; eligible and interested stroke survivors were asked about their problems informally to determine eligibility. Once recruited, the interview explored participant's self-reported cognitive problems in more detail. A cognitive screen was also carried out to gain *indicative* quantitative data on the nature of impairment. Cognitive screening employed the Montreal Cognitive Assessment[18]. This was supplemented by the Apraxia Screen of TULIA[19] and Star Cancellation Test[20] to better detect apraxia and neglect.

Stroke survivors were excluded if they were not pre-morbidly fluent in the English language and/or could not provide informed consent. This was due to the practical requirement to gain narratives from participants (without translation). Hospital in-patients or those living in fully supported care homes were also excluded as their experience of impact may be limited. Those who had been involved in PPI work were excluded from participation in interviews.

[i] Communication aids are supplementary materials in the paper but included as Appendix 2 of this thesis (from page 161)

A purposive sampling strategy[21] - with a sample size determined by data saturation[22] - aimed to recruit participants across the following characteristics:

- Age – adults both below and above 65 years (retirement age)
- Gender – men and women
- Severity of cognitive problems – self-reported cognitive issues, observable issues and screening data described above gave an *indicator* of severity of impairment to drive purposive sampling. We sought to include survivors who achieved a range of scores on these screening instruments, including high scores or 'passes'. We recognised the limitations of screening tools for highlighting the complex and multi-faceted nature of chronic cognitive impairment.

Procedure

The qualitative methodology explored participant meanings and views in a structured way to inform both the conceptual underpinning of any developed PROM as well as its specific content[23]. The use of semi-structured interviews was agreed through PPI and allowed for in-depth exploration of topics that arose[22] to ensure that the derived measure captured information most relevant to patients in accessible terms[24].

EP conducted all stroke survivor interviews one-to-one in participant's homes and independently of carers. Where carers were available and willing, they were also interviewed independently by KWN as part of a wider research programme. Carers were asked about the impact on themselves of stroke survivors cognitive impairments, amongst other things. The results of carer interviews will be reported in a separate paper.

EP had several years of research experience with communication-impaired stroke survivors; including the use of communication aids and carrying out assessments. There was no prior relationship between EP and the participants included in this research.

To facilitate interviews with cognitively-impaired participants, the researchers followed guidelines[25-26] and utilised prior experience of members of the research team; building on previously-developed resources[27]. Interviews typically lasted between 1-2 hours with breaks factored in to overcome fatigue. Examples of the communication aids developed through PPI were used as required to support understanding and expression (see supplementary materials[ii]). Participants were asked open questions initially such as "How do your cognitive problems effect you?" with more closed prompts available (such as how they effect "what you do?" "How you

[ii] Communication aids are supplementary materials in the paper but included as Appendix 2 of this thesis (from page 161)

live?" "How you feel?" etc.). Cognitively-impaired participants may find it difficult to talk in the abstract so these prompts were used to encourage dialogue.

Data Analysis

The goal of analysis was to find commonalities across the interviews to shape focus and content of an outcome measure for future use with a large and heterogeneous sample in a trial.

Interviews were audio-recorded and transcribed verbatim with non-verbal communication recorded using field notes. For example, if interviewees used communication aids to express themselves, this was noted against the recording time to support full capture of survivor story for interpretation and analysis.

A thematic analysis was conducted, using a framework approach[22]. EP repeatedly listened to recordings, alongside transcripts and field notes to achieve immersion in the data and remove any biases or 'knee-jerk' ideas about analysis. EP in collaboration with other members of the research team (KWN, who conducted similar interviews with informal carers of the stroke survivors included here and MH, an expert in qualitative research) then devised a first draft of codes that could be used to describe portions of the data and develop thematic codes that summarised commonalities and differences in the data across participants.

Data immersion began after the first interview was conducted with regular meetings between the research team to reach a consensus on interpretation of the data and to discuss and refine codes. NVivo software was used to attach codes to portions of the data in a way that could be instantly shared between the group to support consensus decision-making. This process also allowed the team to agree when data saturation had been achieved.

Development of thematic charts involved the gradual combination and reduction of codes into overarching themes designed to meet the aims of the research. These were presented visually for the purposes of discussion with the broader research team; to interpret the data and inform recommendations for the necessary qualities of a patient-centred outcome measure for cognitive rehabilitation. An example of the development of one theme is shown in Box 1 in the results section.

Results

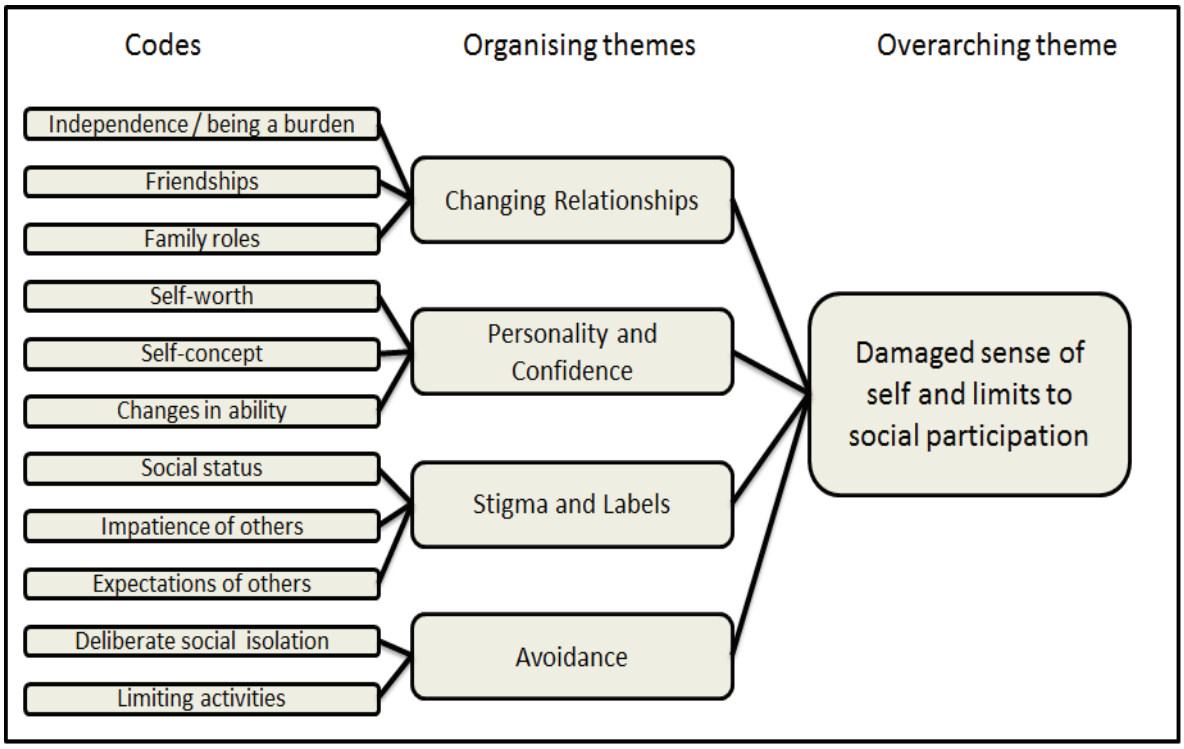
Participant characteristics (demographics and cognitive assessment data) are presented initially with a comment provided on the screening tools used for this study.

Seven themes were developed using the thematic framework approach described (see Box 1 for an example of how one theme was developed). They are described below according to headings:

1. Hidden Problems
2. Focus on cognitive skills, not activities
3. Damaged sense of self and limits to social participation
4. Emotional issues and fatigue
5. Impairment does not equal impact
6. Awareness of cognitive difficulties takes time
7. Perceived level of impact on carers

Information is given to describe each of the identified themes with illustrative quotes in italics. Field notes for non-verbal communication and supplementary information for quotes are inserted in square brackets.

BOX 1: example of the development of the theme 'Damaged sense of self and limits to social participation'



Participants

There were 45 eligible stroke survivors who self-referred to the research team after being invited to take part. Purposive sampling to the point of data saturation led to a total of 16 stroke survivors being interviewed.

Mean age was 58 years (range: 42 to 74) which is relatively young for a stroke population[28]. Time post-stroke ranged from six months to 15 years (mean = 4.5 years). Almost all participants (N=15, 94%) lived with partners and the sample was almost exclusively White British (N=15, 94%). Summary information for each participant is given in table 1.

Participants with a broad range of cognitive impairment were included. Scores on the Montreal Cognitive Assessment ranged from eight to 29 (mean = 22). Despite all participants reporting cognitive deficits that were also observable, four out of 16 (25%) performed at a level sufficient to pass screening tests. Many were clearly employing strategies to do so; for example, mnemonic strategies or deliberate scanning in star cancellation. Others may have passed screens but had obvious impairment that interfered with their cognitively demanding lives.

The use of screening tools that employ cut-offs for 'normal' ranges was often seen as redundant and even offensive, as they do not take into account pre-morbid ability. After testing, one participant commented:

> *"I can't see how you can really measure that [normal ranges]. I mean, [my] friend, he says it himself, he's not very intelligent and he's not very eloquent and he said that I'm now even better than he is. So, I think, when people say, the normal range, on my speech is very good, and that sort of thing, to me, it's not very good" (P08)*

Table 1: Summary of participants

ID	Age	Years post-stroke	Sex	12 years education	Employment	MoCA *	AST **	Star ***
P01	45	12.0	M	Yes	Full time	29	12	54
P02	72	7.2	F	No	Retired	13	8	43
P03	56	10.6	F	Yes	Retired[±]	29	11	52
P04	48	6.9	M	No	Retired[±]	22	10	52
P05	46	1.2	M	Yes	Retired[±]	23	12	54
P06	59	3.0	M	Yes	Retired	18	12	52
P07	72	15.2	F	Yes	Retired	24	12	54
P08	63	3.4	F	Yes	Retired	27	11	52
P09	55	0.6	M	No	Sick leave	25	12	53
P10	74	0.7	M	No	Retired	8	7	25
P11	55	1.2	F	Yes	Retired[±]	25	9	50
P12	54	1.9	M	No	Retired[±]	18	9	37
P13	59	1.1	M	No	Retired[±]	22	12	54
P14	72	1.0	F	No	Retired	16	11	54
P15	42	4.2	M	Yes	Part time	28	12	54
P16	57	1.8	M	No	Retired[±]	24	12	53
Mean	58.1	4.5				21.9	10.8	49.6
SD	10.3	4.6				5.9	1.7	8.0

[±]Retired early; employed at time of stroke

*MoCA = Montreal Cognitive Assessment (total possible score 30). A score of ≥26 is considered 'normal'

**AST = Apraxia Screen of TULIA. (total possible score of 12) A score of <9 is indicative of apraxia

*** Star cancellation test (total possible score 54). A score of <44 is indicative of neglect

Thematic Analysis

Hidden problems

One of the most striking themes was around the hidden nature of cognitive problems. When compared with physical problems, *"invisible"* cognitive problems were felt to be poorly understood by others, including immediate family:

> *"All I'd want more than anything ever is for them [family] to understand that I'm not stupid, I've just got problems." (P12)*

This would often lead to attempted masking of the problems and withdrawal from social situations: *"I just want to cut everybody out."(P14).*

Focus on cognitive skills, not activities

Participants talked about activity limitations due to cognitive difficulties. Activities of perceived importance were varied but what was common across participants was the articulation that different activities relied on the same impaired cognitive skill:

> *"I can concentrate but I'm much more easily distracted, than I was before… I was an avid reader before – and I cannot now effectively read a novel…I couldn't even watch a television programme." (P11)*

Clinically, this does not tell us anything new; but since stroke survivors themselves articulate limitations in this way, it may be valid to ask directly about impactive cognitive limitations (e.g. difficulties with concentration), rather than limitations in particular activities that vary widely between individuals.

Damaged sense of self and limits to social participation

Self-identity could be intrinsically tied to participant's cognitive abilities; being seen and praised as a *"problem-solver"* (P04), *"organised"*(P15), *"capable"* (P13), or *"intelligent"*(P08). Cognitive abilities could have a special significance in this regard and damage could lead to fundamental changes in participant's sense-of-self:

> *"I'd gone from being somebody who was the one who was always there speaking, to be someone who never said anything sat in a corner and so, of course, that's got effects on your personality." (P01)*

Participants also described negative changes in social relationships: *"ordinary people don't want to know us"* (P07). This included relationships with immediate family; losing *"dad confidence"* (P01) or now feeling like a *"burden"* (P02). Difficulties in social contacts with work and friendships were also common across participants.

Emotional issues and fatigue

Cognitive and affective difficulties commonly co-occurred; many participants had low mood or were on medication for depression. Frustration, anxiety and embarrassment were also common emotions associated with cognitive limitations. Cognitive difficulties and negative emotion would often exacerbate one another.

> *"I don't feel confident passing on information... It [getting it wrong] starts making you lack confidence, you see and then you get [more] things wrong." (P03)*

Fatigue was also commonly reported. Fatigue could occur even after very little exertion, but there was a sense that the increased cognitive effort to perform everyday activities would intensify fatigue:

> *"But it's not normal in the sense of you having it [points to EP]…. You are tired because… You've overworked…. I try and…in my head, go asleep. [EP: "So it's that mental effort that can make you quite tired?"]… It must be, it must be that, because I'm not physically, you know…[hand gestures to body and shrugs indicating he is not doing anything physically] It must be my mind." (P13)*

Impairment does not equal impact

The perceived impact of cognitive impairment is mediated by context-specific variables, such as support networks, environmental aids and personality. This was well-captured by the following participant:

> *"How much your brain is damaged is unimportant in terms of how you live your life. So for instance I can say to you, yeah I know I've got brain damage, I know that I don't perform in certain tasks as well as I did but the outcome for me at present is not that bad because [partner] finished work to look after me so I'm really lucky." (P11)*

This lack of a simple linear relationship between impairment and impact echoes the earlier comment on the cognitive assessment scores; even participants with measurably 'normal' cognition can experience significant impacts on their daily life. Impact was typically discussed in terms of how much negative emotion it caused; how much "bother", "upset" or "frustration" it

led to. Cognitive problems were sometimes considered more 'bothersome' by the very fact that they were difficult to see or measure objectively:

> *"I've accepted all my problems with my limbs, and this [points to brain] bothers me more, because people look at you and they expect you to be [alright] and really, you're not." (P15)*

Awareness of cognitive difficulties takes time

The impact of cognitive problems appeared to manifest later in the stroke recovery phase:

> *"It's not just straight away, because it took me three, four years to start thinking [properly] again….at first, your mind is [on], will I be able to sleep tonight." (P02)*

Cognitive problems became more apparent when participants were attempting to return to pre-stroke life including more cognitively demanding activities. The presence of pronounced physical issues may have acted as a barrier to recognising the impact of cognitive problems early on. The cognitive confusion that could be caused by cooking (e.g. ordering actions, following recipes and timing) would not become apparent if participants did not have strength or dexterity to attempt cooking.

Perceived impact on carers

Participants often felt that their cognitive problems impacted on informal carers who had to fill cognitive gaps or rectify dangerous oversights:

> *"I'll cook and I'll leave the gas on… Stupid things. The other day I made a pork chop and set fire to it. Luckily [partner] was in. I put it on but completely forgot about it." (P16)*

The impact on carers could be even more pronounced by a stroke survivor's lack of awareness or memory:

> *"She [partner] can come in here and have a row with me or whatever, because all I have to do is make a bed in the morning… So I forget about it or I have an afternoon sleep and just forget about it, and then after ten minutes of rowing I don't even know I've had a row. But she's the one dealing with all that. It's not fair for her." (P12)*

Stroke survivors were often highly concerned about being a burden due to the effect of their issues on their and loved ones; impact of problems could theoretically be reduced if perceived carer impact was reduced.

Discussion

Qualitative interviews with cognitively-impaired stroke survivors were a challenge but were possible with training, reference to guidance[25-26] and by using the schedule and communication aids that were developed through PPI with this population in mind. Interviewees discussed the specific – and measureable - impacts of persisting cognitive problems that should be included in a PROM evaluating cognitive rehabilitation.

Participants spoke about the specific negative impact of these "invisible" cognitive problems, outside of other stroke-related impairments such as hemiplegia. Emotional issues and fatigue were commonly reported as a result of cognitive problems and appeared to mutually exacerbate one another. This co-existence of issues with mood and cognition is often observed in the literature[29-30]. Therefore an outcome that asks about the specific impact of cognitive problems - particularly on emotion - would be worthwhile for evaluating cognitive rehabilitation trials. Generic or stroke-specific PROMs may not be sensitive enough to measure changes in the impact of cognitive problems in these areas.

Rehabilitation interventions often aim to reduce activity limitations and thus outcome measures typically include information on specific activities[31]. However, the common impacts discussed in this study tended to go beyond activity-specific dialogue. PROM items might therefore be related to cognitive skills (e.g. concentration or remembering) rather than an activity thought to be of importance (e.g. television watching or reading). Items would also usefully explore sense of self and social participation, which were highly impacted by the hidden nature of cognitive problems.

The complexities associated with assessment of cognition[8] reflect the difficulty highlighted in this study and documented in the literature that there is a lack of correlation between levels of cognitive impairment and self-reported impact[32]. The differential impact of cognitive problems (mediated by context-specific variables) means that comprehensive cognitive rehabilitation could theoretically address multiple context-specific variables (for example: support networks; thought processes; compensatory strategies) and reduce perceived impact without actually reducing the underlying problem itself. Theoretically a problem could occur rarely but have high negative impact and vice versa so we argue that it is insufficient to measure outcomes at the level of ‘frequency’ or ‘amount’ of a problem that is assumed to be of importance. Participants typically discussed impact in terms of negative emotion, such as "bother", "upset" and "frustration". “Bother” was the most frequently used emotionally-laden word in interviews and thus could usefully be what is ‘measured’ in a PROM to assess impact. For example: “how much does this problem bother you?” rather than “how often do you experience this problem?”

Participants suggested that the perceived impact of cognitive impairment manifested later in recovery post-stroke. This endorses Pollock et al[4] priority-setting exercise highlighting the need for research to improve chronic cognitive difficulties. It also supports the use of a PROM that has been specifically developed with chronically-impaired service users to ensure it reflects that priorities and values of such individuals. Interventions for chronic conditions may require different outcomes than acute interventions, as they should aim to improve social, psychological and emotional health; issues that users are best placed to comment on[33].

Participants also acknowledged the impact that their cognitive problems had on informal carers and the National clinical guideline for stroke[3] for England and Wales recognises the important role that carers play in supporting chronic conditions. In fact, participants in this study were very concerned by carer impact; such that reduction of perceived carer impact could theoretically reduce impact for stroke survivors themselves. As such, a PROM would usefully include items that explore perceived carer impact.

Strengths and limitations of the study

Cognitively-impaired participants are often excluded from qualitative research, given their potential difficulties processing, understanding and/or expressing their experiences. However, it is important to gather views of relevant populations when developing tools for those populations. Interviewing these participants - some of whom might otherwise have been excluded - was achievable by referring to existing guides [25-26], utilising PPI for developing materials and training researchers, and the use of supportive communication techniques with aids.

Data were collected until the research team were satisfied that data saturation had been reached. Participants had a variety of time post-stroke, education levels, current employment status and cognitive impairment. However, despite age being a driver for purposive sampling, participants in this study were relatively young. A broad age range of people was invited to participate. However, it is possible that younger stroke survivors are more bothered by their cognitive impairments - for example, if they are leading cognitively demanding lives that include work - and thus, they may have been more driven to participate in this study. This may not be a limitation as these are the very individuals who seek cognition rehabilitation.

Participants were also almost exclusively of 'White British' ethnic origin and those who were not fluent in English pre-morbidly were not included. This has implications for generalisability. Future work could include ethnicity as a target variable for purposive sampling to achieve more variability and include non-English speakers with translators or appropriate interviewers.

Carers were interviewed separately (where available) by KWN as part of a wider study. It is possible that interviewing stroke survivors and carers as dyadic pairs could have given us different results. However, the goal here was to explore stroke survivors' views for the purposes of designing a PROM specifically for stroke survivors and PPI endorsed the use of individual one-to-one interviews. The results of carer interviews will be reported in a separate paper but preliminary analysis suggests that carer views broadly corroborate patient views on impact.

For reporting of this study, we have followed consolidated criteria for reporting qualitative research (COREQ)[34].

Summary: implications for outcome measure design

These findings inform desirable qualities of a patient-centred PROM for cognitive rehabilitation trials. It should:

- Include items relating to perceived impact of a comprehensive range of cognitive skills rather than limitations in activities thought to be of importance;
- Address the specific impact of a broad range of cognitive problems on mood, self-identity and social participation;
- Be accessible: including wording and items that respondents endorse and understand;
- Measure impact rather than impairment: this might involve a shift away from reporting frequency of a problem and towards looking at aspects of 'bothered' or 'frustration;
- Include items that explore perceived impact on carers.

Preliminary reviews suggested that no PROM exists that meets all of the above criteria. A subsequent systematic review of existing PROMs (paper in preparation) supported this and a new PROM for cognition has been developed and is undergoing psychometric evaluation (NIHR portfolio entry: http://public.ukcrn.org.uk/Search/StudyDetail.aspx?StudyID=15113). This new tool meets the above criteria and satisfies the criteria highlighted by Lawrence and Kinn[12] for patient-centred outcome measures in that it has been informed primarily by service users and it measures outcomes that have been identified and prioritised as valued (for example, it asks specific questions about social participation and emotion as impacted by cognition). The findings of this study endorse the use of a patient-reported (as well as patient-centred) outcome measure that asks directly about perceived impact of these 'hidden' problems in terms of amount of associated "bother" rather than "frequency," as this appears to be how service users articulate impact of problems.

In addition, we set out to develop a PROM for evaluation of trials to rehabilitate chronic cognitive difficulties after stroke. We have begun to address the criteria related to using tools

at “appropriate times and points” and for specific goals[12] – in this case, evaluation of a rehabilitation trial. It is important to clearly articulate the purpose of any new measure and the context in which it has been developed. Once psychometric testing has been completed, data will be available on whether the PROM appears to be useful in terms of reliability, validity and responsiveness. If appropriate, it will then be freely disseminated with investigators’ brochure to encourage further validation; it is possible that the PROM could be useful for everyday clinical use, as well as trial evaluation.

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Study 2: Systematic Review of PROMs

A systematic review of patient-centred, patient-reported outcome measures (PROMs) for cognitive rehabilitation after stroke

Presented in a format suitable for publication. This article has been submitted to Clinical Rehabilitation and has received feedback from the editor. It will shortly be resubmitted.

A systematic review of patient-centred, patient-reported outcome measures (PROMs) for cognitive rehabilitation after stroke

Abstract

Objective: To identify available patient-centred PROMs for use in trials assessing rehabilitation for post-stroke cognitive difficulties and assess against service user-defined criteria.

Data sources: We performed searches to end of March 2014 in six reference databases; four instrument databases; hand-searched books reviewing outcome measures; and sought expert knowledge.

Review methods: Eligible tools were PROMs relevant to impact of multiple cognitive domains and developed in English. Assessment was on seven criteria (met/unmet) derived from service user interviews related to: patient-centredness; assessing a range of stroke-related cognitive issues; assessing skills; exploring impact of problems as opposed to frequency; exploring mood, participation and self; being accessible for users; measuring perceived carer impact.

Results: Of 167 tools identified, 20 met inclusion criteria with only one – the Burden of Stroke Scale (BOSS) - developed in stroke. None of the identified tools performed positively against more than four of the seven assessment criteria. Most tools assessed frequency of difficulties, and perceived carer impact was rarely explored. The effect of cognition on participation and sense of self was rarely measured and accessible formatting was seldom considered.

Conclusion: There are no patient-centred PROMS addressing the impact of cognitive problems that have been developed for – and are appropriate for use in – trials of comprehensive cognitive rehabilitation after stroke. To address this gap, we developed the Patient Reported Evaluation of Cognitive State (PRECiS) scale and will report psychometric properties presently.

KEYWORDS: patient-centred; patient reported outcome measures (PROMS); cognition; impact; stroke; trials; rehabilitation.

Introduction

Many stroke survivors have enduring issues with cognition that have functional consequences independent of those caused by physical impairment[1]; impacting quality of life and long-term recovery[2, 3]. Improving cognition is the number one research priority for life after stroke as identified by stroke survivors, carers and health professionals[4]. Although it is possible to have difficulties in one cognitive domain, stroke survivors typically have deficits across multiple domains[5] and the National Clinical Guideline for Stroke[6] for England and Wales recommends treating cognitive problems comprehensively.

Recent reviews[7-9] conclude that interventions for cognition rarely assess outcomes beyond impairment that are of 'real life' importance to patients or take their perspective on outcome using patient reported outcome measures (PROMS). Cognitively-impaired individuals are routinely and systematically excluded from the development and use of PROMs[10, 11]. As such, PROMs used for cognition may not meet criteria for being *patient-centred*[12].

A recent review of just the stroke literature identified only three patient-centred outcome measures for stroke[13]. None are suitable for trials of multi-domain cognitive rehabilitation as they: focus on physical and social function[14]; ask about cognitive (and physical) function but not specific impact[15]; or are domain-specific for communication[16].

There is a need for a patient-centred PROM that specifically addresses the impact of a broad range of cognitive problems after stroke and that is developed in collaboration with cognitively-impaired stroke survivors. We worked with this demographic to explore patient perspectives on the important, measureable impacts of cognitive difficulties that should be priorities for inclusion in an outcome measure[17]. As well as the requirement for patient-centredness, we identified six additional desirable qualities for a PROM[17]:

1) Assessing a range of cognitive domains;
2) Assessing cognitive function (e.g. concentration), rather than specific activities (e.g. reading)[i];

[i] Because activities are specific to – and vary between – individuals e.g. .reading, bird-watching, card-playing.

3) Measuring impact in terms of 'bother' or similar, rather than frequency or amount;
4) Including effect of cognitive problems on mood, self-identity and social participation;
5) Accessibility to respondents;
6) Exploring perceived impact on informal carers.

We undertook this review of a broad set of relevant literature to identify whether any existing tools, perhaps from other neurological conditions, met these seven criteria and might be used in stroke.

Method

Four methods were used to identify available instruments up to end of March 2014:

1. Electronic searches of Psychinfo, Medline, EMBASE, AMED, CINAHL and the British Nursing Index. Searching used a combination of terms and keywords relating to adult cognition and cognitive impairments; PROMs; and psychometric validation. No terms restricted the search to stroke so that tools developed in other conditions would also be identified. Results were imported into a reference manager (EndNote) and duplicates were removed. Appendix 3 (page 164) gives an example of the PsychInfo search strategy.
2. The following databases of instruments were also searched for terms related to cognition and PROMs: registry of outcome measures (ROM: http://www.researchrom.com/); Patient-reported outcome and quality of life instrument database (PROQOLID: http://www.proqolid.org/); Oxford patient-reported outcomes measurement group (http://phi.uhce.ox.ac.uk/); Canadian Interdisciplinary Network for Complementary and Alternative Medical Research outcomes database (IN-CAM: http://www.outcomesdatabase.org/).
3. Books that reviewed outcome measures were also searched [18-20].
4. Healthcare professionals and researchers involved in the Organisation for Psychological Research in Stroke (OPSYRIS) were consulted via email to ask about relevant tools that should be included.

Instruments were eligible for assessment if they were PROMS for adults, developed in the English language and not limited to a single domain of cognition. Eligible PROMs were subsequently critically assessed in two stages.

The first stage assessed how far PROMs met the aforementioned seven desirable criteria from our qualitative study[17]. A copy of the PROM - as delivered to participants - was sought as was information on the developmental processes and original purpose of the tool. It was outside of the funding abilities of this research to acquire full copies of tools if they were not freely available either online or directly via the author. The seven criteria largely related to face validity and conceptual underpinning of the tool. Each criterion was broken down into descriptors that could subsequently be assessed as either met or not, as follows:

1) *Patient-centred.* To meet this criterion, service users had to be involved in development of the tool;
2) *Assessing a range of post-stroke cognitive domains*. The range of domains of interest was: executive function; memory; attention; spatial neglect; perceptual abilities; aphasia; and motor apraxia. Each was assessed as covered if conceptualised in tool development or if included content/items used relevant wording as found in Cochrane review topics[21-27] and a review of post-stroke subjective cognitive complaints[28];
3) *Assessing cognitive skills.* This criterion was met if at least 2 items used appropriate wording, assessing a broad cognitive skill(s) that was not activity-specific;
4) *Measuring impact.* PROMs met this criterion if the purpose of the tool or the wording of the items / rating scales were approached in a way to assess impact of a problem as opposed frequency / amount of difficulty. A cautious assessment was used: if the rating scale related to frequency / amount but the item wording was geared towards impact (such as bother, worry, hindering daily life and synonyms), it would score as met;
5) *Mood, self-identity and social participation.* At least one question must be included related to each of these three broad dimensions and must ask directly about the effect of cognitive issues. It would not be sufficient for a tool to ask questions about changes in these dimensions unrelated to cognitive issues.

6) *Accessibility for respondents*. Broken down into four components that all needed to be met: the average (mean) Flesch-Kincaid readability score across all items should be ≥70[29, 30]; less than 10% of items should achieve a score of <60 (50-59 becomes 'fairly difficult' and lower scores relate to 'confusing' sentence structures[30]); formatted for a cognitively-impaired respondent (e.g. large / bolded / coloured text); simple response scales, changing only as necessary with item wording;
7) *Perceived impact on carers*. This criterion was met if at least one addressing question was included. For example asking about patient perceptions of carer worry, sadness or burden.

If PROMs met all stage one assessment criteria, stage two of assessment considered other psychometric properties of the tool such as reliability and construct validity. A critical appraisal checklist was adapted from several sources[31-33] for these purposes (see Appendix 4 from page 167).

Results

The search strategy generated a list of 167 tools. 147 measures were ineligible for stage one assessment. The top three reasons for exclusions were: only dealing with one domain of cognition (N=38); quality of life or activities of daily living tools that did not include items exploring the impact of cognitive limitations (N=33 and 25 respectively); tools that dealt with non-related issues such as back pain (N=25). A full breakdown of reasons for exclusion is shown in Figure 1 overleaf. 20 PROMS were assessed and Table 1 shows the performance of all tools against criteria.

Figure 1. Overview of search results and reasons for exclusions:

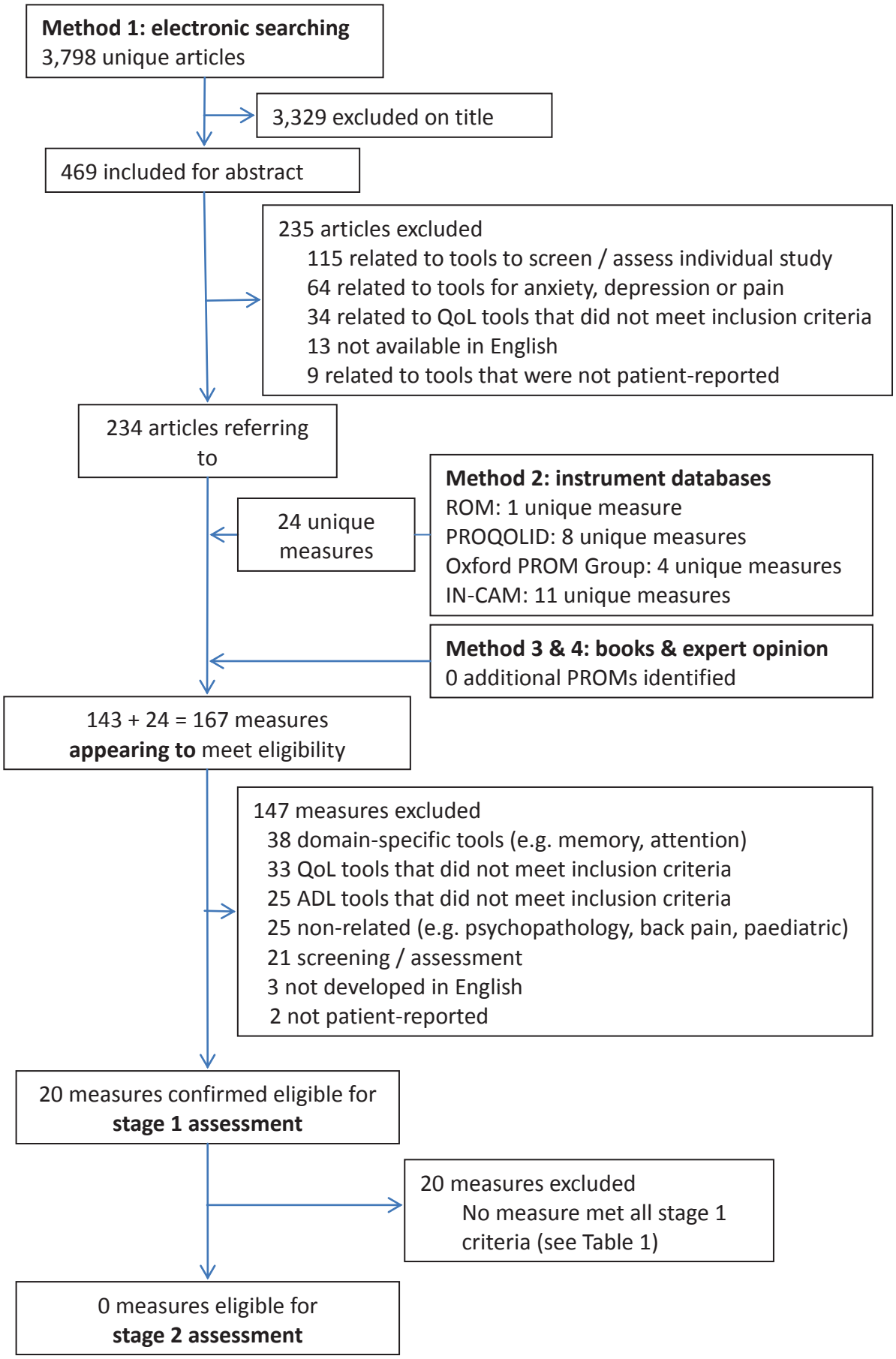

Table 1. Details of the performance of 20 PROMs (presented alphabetically) against the seven stage one assessment criteria.

Assessment criteria codes.

1 = Patient Centred.

2 = Range of cognitive domains covered. If not met, lettered list of issues covered: Executive (a); Memory (b); Attention (c); Neglect (d); Perception (e); Aphasia (f); Motor apraxia (g).

3 = Assessing cognitive skills.

4 = Measuring impact not frequency/amount assessed.

5 = Effect of cognition on dimensions of life. If not met, lettered list of dimensions covered: Mood (a); Self (b); Participation (c).

6 = Accessibility to users. If not met, lettered list of sub-criteria met: Mean Score ≥70 (a); <10% items scoring <60 (b); Formatting considerations (c); Response Scale simple (d).

7 = Perceived impact on informal carer.

Tool name*	Description	1	2	3	4	5	6	7
3CL[34]	28-item checklist to assess cognitive decline [full copy unavailable]	×	× (abcf)	×	×	×	× (d)	×
ABNAS[35]	24 items to assess effects of anti-epileptic drug treatment.	✓	✓	✓	×	×	× (ad)	×
AM-PAC[36]	19 items for applied cognitive activity limitations in post-acute settings.	✓	× (abcf)	×	×	×	× (d)	×
BOSS[37]	64 items to assess health status following stroke.	✓	× (abcf)	✓	×	✓	×	×
CDS[38]	39 items to assess the cognitive side effects of anti-depressants.	×	✓	×	×	×	× (a)	×
CFSS[39]	18 items to assess cognitive function in general population	×	× (abcefh)	×	×	×	× (d)	×
CFQ[40]	25 items to measure frequency of cognitive errors.	×	✓	×	×	×	× (ad)	×
EBIQ[41]	63 items to assess difficulties following acquired brain injury.	×	× (abcfg)	×	×	×	× (d)	✓
ECog[42]	39 items to diagnose mild cognitive impairment onset.	×	✓	×	×	×	×	×
EFQ[43]	41 items to explore coping following brain surgery.	×	× (abcf)	✓	×	×	× (d)	×
FACT-Cog[44]	37 items to assess changes in cognition due to chemotherapy.	✓	✓	✓	×	× (ac)	× (ad)	×
MCQ[45]	13 items to assess quality of life in mild cognitive impairment	✓	× (abf)	✓	✓	× (a)	× (d)	✓
MANS[46]	87 items to aid diagnosis of neurodegenerative conditions.	×	✓	×	×	×	×	×
MASQ[47]	38 items to assess self-awareness of cognitive skills.	×	✓	×	×	×	× (d)	×

PAF[48]	48 items to assess cognitive difficulty for neuropsychological assessment	✓	× (abcdef)	✓	×	×	×	×
PDQ[49]	20 items to assess perceived cognitive function in Multiple Sclerosis.	✓	× (abc)	✓	×	×	× (ad)	×
PPFS[50]	6 items to assess pharmacologic intervention in psychotic disorders.	×	× (abcf)	✓	×	×	× (d)	×
PROMIS[51]	34 item bank to assess applied cognition.	✓	× (abcef)	✓	×	✓	× (ad)	×
PROCOG[52]	55 item bank to assess impact of mild to moderate cognitive impairment.	✓	× (abcef)	×	✓	✓	× (d)	✓
SASCI-Q[53]	29 items to assess cognitive impairment for diagnostic utility	✓	× (abcf)	×	×	× (a)	×	×

*Tool Names in full: 3CL = Cognitive Change Checklist; ABNAS = A-B neuropsychological assessment schedule; AM-PAC = Activity measure for post-acute care; BOSS = Burden of Stroke Scale; CDS = Cognitive Difficulties Scale; CFSS = Cognitive functioning self-assessment scale; CFQ =Cognitive Failures Questionnaire; EBIQ = European Brain Injury Questionnaire; ECog = Everyday Cognition questionnaire; EFQ = Everyday Functioning Questionnaire; FACT-Cog = Functional Assessment of Cancer Therapy Cognitive Scale; MCQ = Mild Cognitive Impairment Questionnaire; MANS = Multi-dimensional Assessment of Neuro-degenerative Symptoms; MASQ = Multiple Abilities Self-Report Questionnaire; PAF = Patient's Assessment of Own Functioning; PDQ = Perceived Deficits Questionnaire; PPFS =Patient Perception of Functioning Scale; PROMIS = Patient Reported Outcomes Measurement Information System; PROCOG = Patient-Reported Outcomes in Cognitive Impairment; SASCI-Q = Sahlgrenska academy self-reported cognitive impairment questionnaire

Patient-centred

Ten out of 20 measures included some user involvement in the development phases of the measure [35-37, 44, 45, 48, 49, 51-53]. Of those, only the Burden of Stroke Scale[37] was developed specifically with stroke survivors. Those tools that involved patients in their development typically used interviews or focus groups to identify or refine items.

Assessing a range of cognitive domains

All 20 measures included items related to more than one cognitive domain. Aspects of memory function were assessed in all tools whereas stroke-specific domains such as apraxia and unilateral neglect were only assessed as covered in five[35, 38, 39, 41, 46] and eight tools[35, 38, 40, 42, 44, 46-48] respectively.

Assessing cognitive skills

There was a focus on assessing a variety of different activities (as many as 41 in the Multi-dimensional Assessment of Neuro-degenerative Symptoms (MANS)[46]) as opposed to focusing on the cognitive skills that underpin the activities.

Measuring impact

Almost all tools asked respondents to rate the frequency or amount of a problem presumed to be of importance. Only two PROMS[45, 52] asked respondents to rate aspects of bother, worry, anxiety, sadness or frustration that were specifically related to cognitive function.

Mood, self-identity and social participation

Only three of the tools had items that explicitly asked about the effect of cognitive problems on these broader dimensions[37, 51, 52]. Other tools asked questions related to these dimensions but they were not linked to cognitive function and therefore not capturing intended outcomes for our purposes.

Accessibility for respondents

No tools were deemed to meet this criterion by fulfilling all four of the defined sub-criteria. No tools appeared to have considered accessible formatting such as large text or user-friendly layout. Included measures required between 6 and 87 responses (mean = 36).

Perceived informal carer impact

Only three tools[41, 45, 52] included item(s) that could arguably be described as exploring carer impact. For example, the Mild Cognitive Impairment Questionnaire (MCQ)[45] asks whether respondents have to rely on partners or other people to "help you remember things" and whether respondents worry that they have "upset other people because of (your) memory problems."

Overall: tools and summary

The two best-performing tools (both meeting four criteria) were developed to look at impact and/or quality of life in mild cognitive impairment: Patient-Reported Outcomes

in Cognitive Impairment (PRO-COG)[52] and Mild Cognitive Impairment Questionnaire (MCQ)[45].

The 'applied cognition' bank of items developed by the Patient Reported Outcomes Measurement Information System (PROMIS)[51] to assess perceived cognitive functioning, met three criteria.

The Burden of Stroke Scale (BOSS)[37] (three criteria met) and European Brain Injury Questionnaire (EBIQ)[41] (one criteria met) were the only PROMs developed for acquired brain injury. The former was designed to evaluate overall health status after stroke and the latter to assess experience of difficulties following traumatic brain injury.

Other tools were originally designed to assess the impact of different medical interventions on cognitive function: anti-epileptics[35]; anti-depressants[38]; anti-psychotics[50]; chemotherapy[44]; brain surgery[43] and all these tools met between one and three assessment criteria. Seven were designed to collect reported changes in cognitive function but with the primary purpose of aiding diagnosis of neurodegenerative disease or mild cognitive impairment (MCI)[34, 39, 40, 42, 46, 48, 53] and none of these tools met more than two criteria. The Multiple Abilities Self-Report Questionnaire (MASQ)[47] was designed to assess awareness of cognitive abilities and met one assessment criterion. The Perceived Deficits Questionnaire (PDQ)[49] was developed to assess cognitive difficulties in multiple sclerosis (two criteria met). The Activity Measure for Post-Acute Care (AM-PAC)[36] met one criterion.

Overall, the 20 tools represented a wide range of measures. Copies of tool with full item wording as presented to patients were available for all but one of the tools[34]. None of the identified tools performed positively against more than four of the stage one assessment criteria. As such, none were included in stage two assessment.

Discussion

We identified 167 unique tools and assessed 20 PROMS. Ultimately, no PROM was identified that met all stage one assessment criteria (that had been generated through

qualitative interviews with stroke survivors[17]) for a PROM to use in trials of comprehensive cognitive rehabilitation in life after stroke. As such, other psychometric properties relevant to stage two assessment were not considered further.

Included tools were not developed for the purpose of evaluating comprehensive cognitive rehabilitation after stroke. Their failure to meet our assessment criteria has no implications for their validity in their intended uses. Only one tool was developed in stroke[37] and this was not designed as a cognitive-specific impact measure. PROMs could theoretically be adapted to meet the criteria we assessed. For example, changing wording to be more accessible; including items for perceived carer impact; or asking about 'bother' as opposed to 'frequency'. However, any published psychometric properties of a tool are specific to the items and content of the tool[54] so alterations would require re-validation.

Most tools were heavily loaded towards memory issues and almost all tools explored frequency or amount of difficulty with cognitive-related activities assumed to be of importance, rather than asking directly about the impact of cognitive problems. Some did have sections that included items related to mood, social participation and self-identity but these items were rarely linked to cognition and thus would not be sufficient for our needs.

The Mild Cognitive Impairment Questionnaire (MCQ)[45] and the Patient-Reported Outcomes in Cognitive Impairment (PROCOG)[52] were the best performing tools included in stage one assessment. The range of issues covered was limited and accessibility was of concern, meaning that neither tool would be suitable for post-stroke comprehensive cognitive rehabilitation. However, they are theoretically underpinned to explore quality of life specifically related to mild cognitive impairment. Tools that do not ask about the impact of cognition in this way may not be sensitive enough to assess the effect of interventions for this issue. Research in other chronic conditions affecting cognition such as cancer[55] and Parkinson's disease[56] have endorsed the viewpoint that cognitive issues impact dimensions of life that should be explored explicitly in measurement tools.

Accessibility of tools was often a concern, given that they were specifically targeted towards a demographic that might have issues processing information. Interestingly, none of the tools appeared to have considered issues of formatting (text size / layout)

when presented to participants. Accessibility and formatting would be an important point of consideration for any newly developed tool with stroke patients who may have specific issues with language processing and comprehension. It is likely that aphasia-friendly versions of tools would need to be designed and validated.

Strengths and Weaknesses of the Review Process

This review took a structured approach to tool assessment with a two-stage process to critically appraise psychometric properties only of tools that were assessed as appropriate for our target population. Our previous work with stroke survivors[17] highlighted the importance of patient-centredness plus additional core criteria related to content validity and conceptual approach. A tool that does not meet these criteria cannot be fit for purpose with this population, regardless of psychometric properties.

This review utilised a multi-faceted, broad search strategy to identify PROMs related to cognition, regardless of aetiology. This was to find tools that may be suitable for use in stroke trials, subject to some potential adaptation and validation. Only one eligible tool was developed for stroke and this may have contributed to the fact that no tools were judged to fully meet the assessment criteria. However, the broadness of the search strategy could be justified since the best performing tools were the MCQ[45] and PROCOG[52], developed outside of stroke.

Assessment of the criteria was necessarily subjective in parts and it therefore erred on the side of inclusion (classifying as 'met'). For simplicity we dichotomised assessments as met or not. This approach risked losing some detail, for example on extent of accessibility, but was sufficient to meet our aims.

Clinical Messages

- Whilst there are PROMs whose strength should be utilised, none fully meet the seven criteria (defined by users) for use in trials of comprehensive cognitive rehabilitation in life after stroke.
- Our newly-developed Patient-Reported Evaluation of Cognitive State (PRECiS) fills this gap. Dissemination of PRECiS is underway.

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3 Tool development

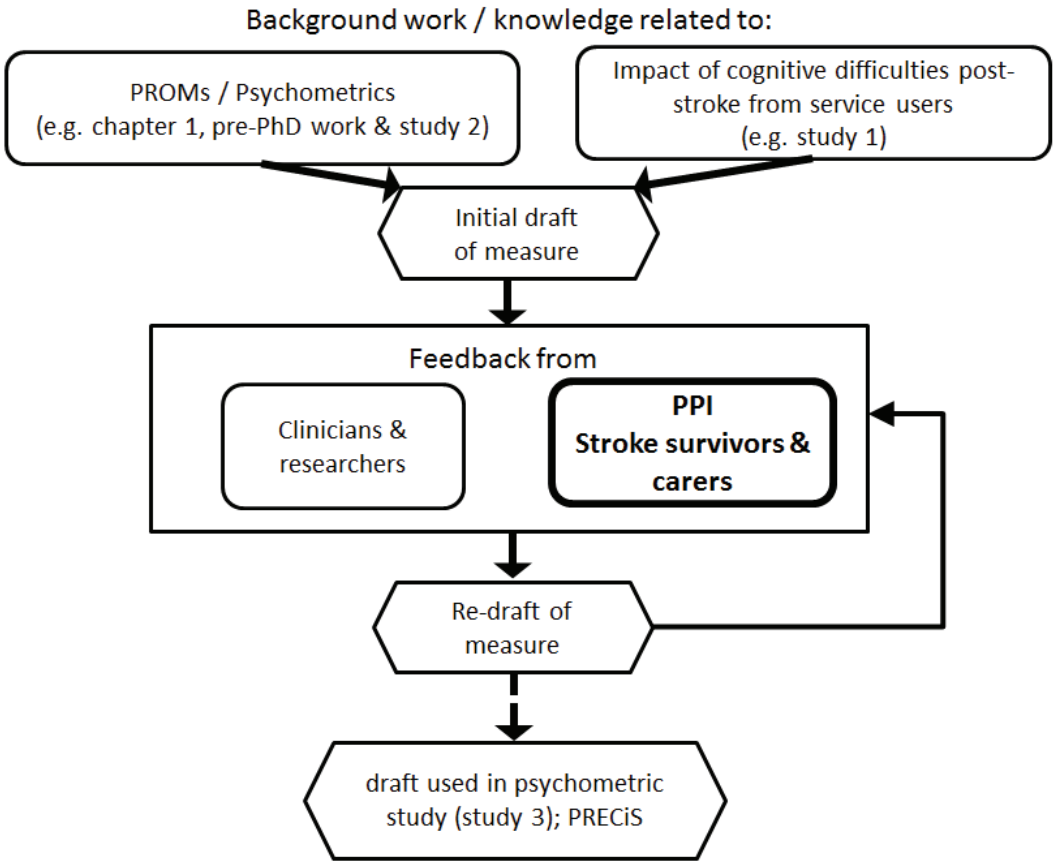
The aim of this pilot test stage was to develop and refine a new PROM for use in trials of comprehensive cognitive rehabilitation in life after stroke: the Patient-Reported Evaluation of Cognitive State (PRECiS).

This chapter is not intended for publication but gives information on the process used to achieve this aim and produce the version of PRECiS used in the psychometric study (study 3). The iterative process that involved synthesising information from different sources to make informed decisions is described (see 3.1) and the decisions taken are highlighted with reference to different aspects of PRECiS including: content and number of items (see 3.3 and 3.4); formatting (see 3.6); and acceptability and face validity (see 3.9). The version of PRECiS used in the psychometric study is described in section 3.11and included in appendix 5 (from page 168). The discussion chapter (see 4.3.4) considers strengths and weaknesses of this process.

3.1 The process

Figure 3.1 below shows the process contributing to the decision-making for developing the version of PRECiS used in the psychometric study.

Figure 3.1 Integrating different sources of knowledge to inform development of PRECiS



The literature on cognitive difficulties, impact of health conditions and PROM development (see chapter 1) was one means of developing contextual knowledge about the subject matter to help inform decisions about PRECiS e.g. see section 3.4 Number of items. In addition, pre-PhD work on the ACT NoW study (Assessing Communication Therapy in the North West (Bowen, et al., 2012)) had included developing another patient-centred PROM; the Communication Outcomes after Stroke (COAST) scale (Long, et al., 2008). COAST was developed in collaboration with researchers, healthcare professionals and the ACT NoW Research User Group; a dedicated group of service users including stroke survivors with communication problems and their carers. Knowledge gained through working closely with the ACT NoW user group and the wider ACT NoW team also informed the decision-making process for developing PRECiS (e.g. see 3.6 Formatting).

The qualitative study (study 1) (Patchick, et al., 2014) led to recommendations about the content of a PROM for post-stroke cognitive rehabilitation (e.g. see 3.2 A starting point for content and concept) and also provided data on how stroke survivors describe their issues that informed specific wording and content of items (e.g. see 3.3 Content of items). Patient and Public Involvement (PPI) with service users had informed the methods and materials used in the qualitative phase and a dissemination event to feedback and validate the conclusions drawn from the interviews supported the credibility of the recommendations made. The interview schedule developed through PPI included lay definitions of cognition and this was carried through into eventual drafts of PRECiS (see 3.7 Including an introduction).

The systematic review (study 2) was primarily concerned with identifying and appraising tools against the user-defined criteria that emerged from the qualitative study (Patchick, et al., 2014). The PROMs identified in the review did not met criteria and were not deemed suitable for our purposes, but they had strengths to be learnt from. Four reviewed PROMs in particular were useful: the two best-performing tools (both meeting four of seven criteria) were the Patient-Reported Outcomes in Cognitive Impairment (PRO-COG) (Frank et al., 2006) and Mild Cognitive Impairment Questionnaire (MCQ) (Dean et al., 2014); the Burden of Stroke Scale (BOSS) (Doyle, 2002) met three criteria and it was developed with people with stroke; The 'applied cognition' bank of items developed by the Patient Reported Outcomes Measurement Information System (PROMIS) (Becker, Stuifbergen, & Morrison, 2012) also met three criteria and some stroke survivors were involved in early developmental stages. Promising items and features from these tools were extracted into a spreadsheet database to view commonalities and help inform PRECiS (e.g. see 3.5 Response scales).

All these preliminary aspects together helped inform the first draft of PRECiS that was then subject to consultation and feedback from service users and healthcare professionals. There

were five different community stroke groups that provided consultation on drafts of the measure and they were:

1. Speakeasy in Bury – a charity set up for stroke survivors with aphasia and their carers;
2. Different Strokes group in Kendall – a community group for younger stroke survivors;
3. Bolton West Stroke Group – a Stroke Association community support group;
4. Macclesfield and District Young Stroke Society (MADYSS) – a community group that, despite the name, had members with a wide range of ages.
5. The Brain and Spinal Cord Injury Center (BASIC) - offering rehabilitation services in Greater Manchester for individuals with acquired brain injury (including stroke) and their families.

In addition two individual stroke survivors, former members of a research user group for the ACT NoW study (Bowen, et al., 2012), also provided one-to-one feedback including role-play / practice testing of early drafts. Consultation with service users typically involved a brief explanation of what the tool was aiming to do and then going through it page-by-page for feedback on content, coverage, layout, and other aspects of format. The feedback from service users informed several refinements before healthcare professionals and researchers were consulted. Feedback from healthcare professionals and researchers was sought by email using the mailing list from the Organisation for Psychological Research into Stroke (OPSYRIS) that had almost 100 recipients. Email feedback was received from 32 individuals (including research trial managers, NHS occupational therapists and clinical psychologists (e.g. see 3.9 Acceptability and face validity)). Service user consultation was carried out again after OPSYRIS-provided-feedback had been considered. Consultation with service users, healthcare professionals and researchers informed re-drafts whilst attempting to remain true to the background work that had been carried out.

There were numerous sources of information to influence the content of the tool. The variation in feedback received from multiple sources meant that decision-making for refinements was challenging. Refinements were made based on informed decision-making, although the opinions of stroke survivors with cognitive issues were weighted most heavily given that they were potential future users of PRECiS and therefore best placed to comment on its accessibility (Fitzpatrick, et al., 1998; Gibbons & Fitzpatrick, 2012).

The collective result of this work is the version of PRECiS that was taken forward for psychometric field testing and is described below (section 3.11 with a copy of PRECiS plus administrator guide included in Appendix 5 from page 168)

3.2 A starting point for content and concept

The recommendations from the qualitative study informed underlying qualities of a patient-centred PROM for trials of cognitive rehabilitation (Patchick, et al., 2014). It should:

- Include items relating to perceived impact of a comprehensive range of cognitive skills rather than limitations in specific activities;
- Address the direct effect of cognitive problems on mood, self-identity and social participation;
- Measure impact rather than impairment: involving a shift away from reporting frequency of a problem and towards looking at aspects of 'bother' or 'frustration';
- Include items that explore impact on informal carers, as perceived by respondents;
- Be accessible: including wording and items that respondents endorse and understand.

These recommendations provided a conceptual foundation for PRECiS.

3.3 Content of items

Some of the item content was influenced by direct quotations from the qualitative study. For example, “capable” was wording used by stroke survivors and it seemed to capture notions of independence and views of the self:

> “If I could do all of what I wanted to do, go out, drive, do all that, I’d do it, and **I’d be capable of doing it if that was right**. But it’s not, so I can’t.” [P16]
>
> “I am quite useless, because I can’t remember, you know, being…being what I’m…**what I want…err, capable**, you know.” [P13 - stroke survivor with aphasia]

In subsequent feedback with service users, “capable” was a well-received term for ability and independence. The qualitative study also endorsed the inclusion of "ordering" as a broad term that captured a cognitive skill most closely related to apraxia; in early schedule-development work when establishing how to define 'cognition' as well as in the interviews themselves. Two participants with known apraxia used similar wording when describing some of their difficulties:

> “I’m just sorry I can’t get it **done in the right order**.” [P16]
>
> “I can’t sequence and I really still have difficulties with sequencing. So anything that requires a **sequence of actions** …. I actually have to get somebody else to help me do it and it’s stupid because it’s so annoying, it’s such a basic thing.” [P11]

The qualitative study recommendation to measure impact rather than frequency in language that users understood led to the use of the word "bothered" to capture impact. This was well-

used in the qualitative interviews (Patchick, et al., 2014) but was also validated in subsequent feedback sessions when stroke survivors felt that it was an endorseable word; more so than 'stronger' words such as stressed or frustrated.

Early drafts of PRECiS used "non-physical problems" and "thinking and memory" instead of "cognition" as it was felt they may be more accessible. From user feedback received, “non-physical problems” was deemed too ambiguous, whilst “thinking and memory” meant that a) each item sentence was too long; and b) the full spectrum of cognition might be overlooked. The decision was taken to use the term ‘cognition’ with a definition supplied in an introductory section (see 3.7) that could be referred to throughout the session i.e. a separate page with the definition that was kept in view.

At a late stage in the feedback process, two clinical psychologists gave interesting feedback about the problem-based content of the measure and felt there should be a move away from this in measurement tools. They suggested that a solution-based approach that was more positive would be preferable for picking up the effect of rehabilitation e.g. items that ask for agreement with a statement such as: “I feel I can cope with memory problems.” This feedback was received in the final stages in tool development and whilst appealing, it had not been articulated by any of the service users or carers in feedback. Also, the process did not permit returning back to the original qualitative interview respondents, but all interviewees had expressed impact of cognition in terms of loss and negative connotations. The decision was to continue with the tool that had already had a significant amount of development and consultation and to prioritise service user feedback over that from professionals.

3.4 Number of items

The four strongest tools from the PROMs review in study 2 (described in section 3.1) contained between 13 and 64 items (mean = 41). Across all 20 tools reviewed there was an even wider range of number of items: between six and 87 responses (mean = 36). Recommendations in the literature for PROMs development suggest that a measurement tool containing a maximum of 30 items is preferred: any more is difficult to explore well in test phases and a large number of items is also likely to artificially inflate estimates of internal consistency (Johnson et al., 2011; Streiner & Norman, 2008). Furthermore, it is desirable to reduce burden on respondents.

The first draft of PRECiS that was taken to a formal user consultation meeting (at Speakeasy group) had 30 items: 19 related to specific cognitive skills and functions; 2 items related to perceived self; 2 that related to perceived carer impact; 4 relating to social roles and participation e.g. family and work and 3 exploring emotions. Feedback from Speakeasy and other groups gradually shaped refinements such that, whilst the overall number of items only

reduced by three (to N=27), the balance within sections changed. The number of questions about cognitive skills was reduced. Originally there were several items relating to different aspects of communicating e.g. finding words; communicating with people you know well; communicating with strangers. User consultees tired of this many questions and suggested that the important point was the bigger picture that you were able to “get your point across and communicate.” The suggestion was to pare down questions to just get to the root of the problem i.e. ‘communicating’ rather than all the specific ways in which one communicates and in which one might have problems. Conversely, the number of questions pertaining to mood and participation was increased. As in qualitative interviews, service users felt that these were very important impacts and warranted more questions; particularly more on different types of emotional impacts.

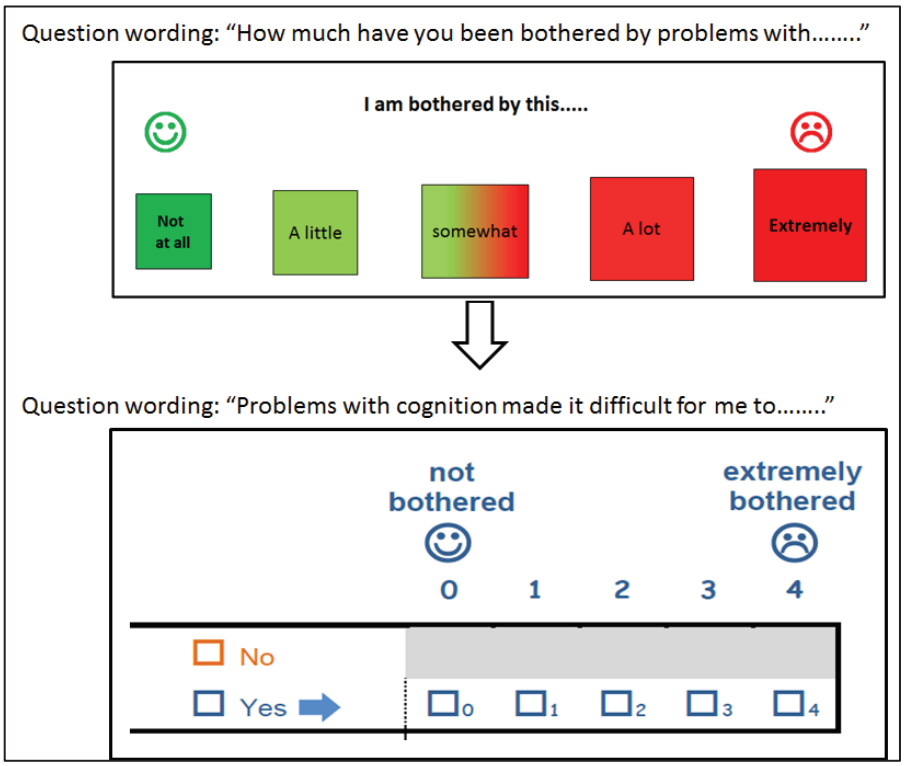
3.5 Response scales

Cox (1992) and Streiner & Norman (2008) advocate the use of either Likert-scaled items or visual analogue scales (VAS) to provide richer data than binary response scales. There is evidence that Likert-scales are more acceptable than VAS to respondents in terms of ease-of-use (Fitzpatrick, et al., 1998). For the four best performing tools in the PROMS review (described in 3.1), all used Likert scales with five response categories that had verbal anchors for each response. The acceptability of Likert-scaling was also endorsed by the ACT NoW research user group when developing the COAST scale (Long, et al., 2008). Anchoring end points with emoticons (smiley and sad faces) was used to provide further clarity of the meaning of response scale end points.

In the early versions of the scale, there was one question per A4 page and the item wording combined the notion of problems and bother such that the response rating scale asked respondents only to rate their bother. For example: Question: "In the past week, how much have you been bothered by problems with….." Answer: "I have been bothered by this…. " service user consultation led to a pivotal change in this design; they wanted to be able to show clearly that they could have a problem or difficulty but had learnt to cope with it so that they were not bothered by it. This eventually led to altering the layout of questions such that respondents first answered yes/no to indicate whether there was a difficulty and then to rate bother associated with that difficulty, if applicable (see figure 3.2 overleaf). Alongside this change in the rating scale, there were other changes to the formatting overall (see section 3.6) that led to the overall size of the questionnaire being reduced and having more than one question per page. A knock-on of this was the decision to remove the interim descriptors from each of the response boxes (e.g. "a little", "a lot") in order to keep the size of the font used in

the questionnaire large and the layout uncluttered. There was not consistent feedback regarding the discussion to remove these descriptors: some service users and healthcare professionals liked the use of descriptors for each box; others felt that it led to more burden since it required more reading and interpretation. The latter consultees endorsed the use of anchoring the end points with descriptors and using numbers in between.

Figure 3.2 how the response scale changed: the image at the top is the first draft and the image at the bottom shows the final version used in PRECiS for the psychometric study



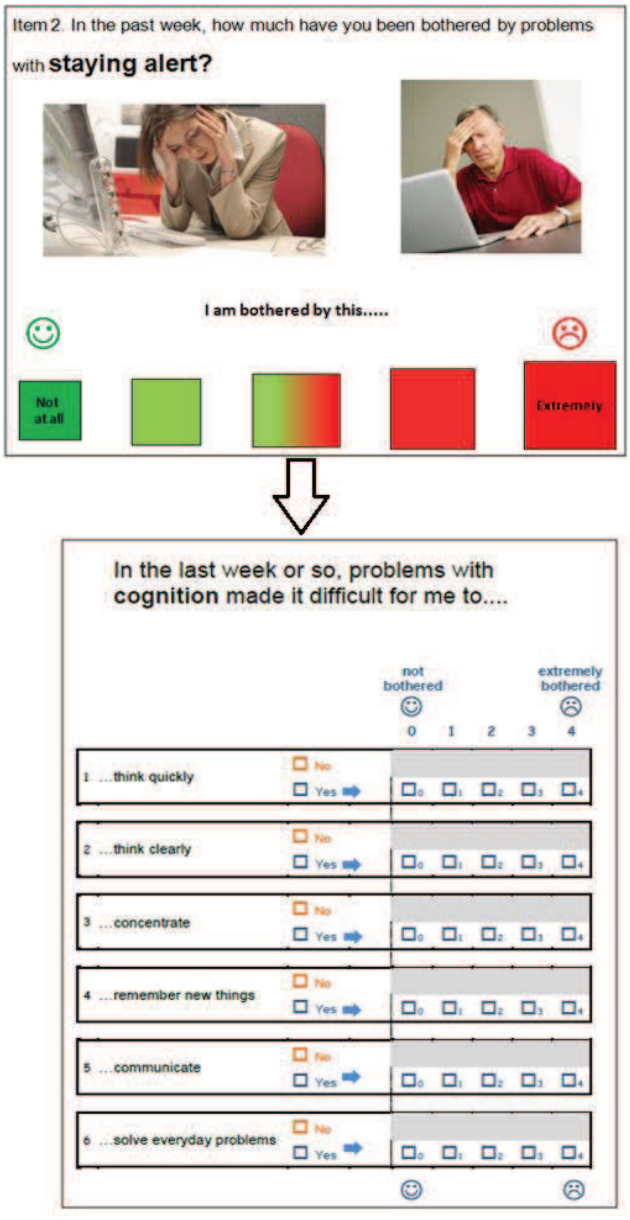
3.6 Formatting

Consideration of formatting was overlooked in all of the 20 PROMS reviewed in study 2 but is important to factor in (Cox, et al., 1992; Gibbons & Fitzpatrick, 2012). The (COAST) Scale (Long, et al., 2008) had considered overall formatting and had worked with users to optimise understanding through the use of: large, coloured, bolded text for emphasis; smiley and sad faces to anchor response scales; well-spaced layouts; having one page per question and spiral binding to facilitate perusal for stroke patients with hemiplegia; and pictures to improve understanding.

As stated in section 3.5, early drafts of PRECiS originally used one question per page. The layout was modelled on the COAST and pictures were included for each question. The pictures used were a cause of much discussion; with disagreements about the clarity of pictures used to appropriately capture something like "solving problems" or "family life" without ambiguity. Feedback was mixed regarding the decision taken to remove the pictures overall but making

this change did have the knock-on benefit – that had been suggested and subsequently approved by service users – of significantly reducing the physical size of the questionnaire since more than one question could be fitted per page. Professionals' email feedback on the later versions with pictures removed, suggested that the addition of photos would be beneficial but they did recognise the difficulty of including photos that would capture concepts well enough to support understanding. The use of pictures to aid understanding is generally recommended although it does not always support comprehension and some people with communication difficulties do prefer text-only versions of information (Brennan, Worrall, & McKenna, 2005; Rose, Worrall, & McKenna, 2003). Figure 3.3 shows the transition from the early stage of the questionnaire - with one question per page and pictures that were ultimately deemed to be ambiguous - to the final version used in the psychometric study.

Figure 3.3: how the formatting changed: the image at the top is the first draft and the image at the bottom shows the final version used in PRECiS for the psychometric study



Despite the difficulty selecting photos, service users endorsed having a version of PRECiS that could be used with people who might have difficulty processing a page with more than one question / concept. As such, an alternative version with just one question per page with very large font sizes was made available to be utilised with individuals who struggled with the primary paper format. The number of people who required this alternative version was recorded in the psychometric study (study 3).

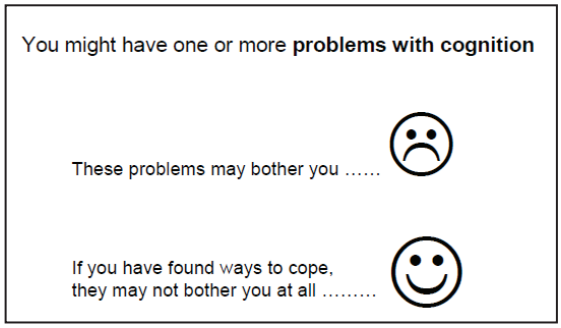
3.7 Including an introduction

Stroke survivors felt that it was useful to have some orientation to a questionnaire and suggested an introduction to give the rationale behind the questions and more definitions of wording. The first two pages of the PRECiS questionnaire booklet were subsequently designed as introductory sections and can be viewed in appendix 5 (from page 168).

The description of cognition evolved from early in the qualitative study when service users helped inform the interview schedule. This was built on during the consultation stages carried out until the feedback received about the definition of cognition was well-received by most users. Healthcare professionals and researchers also were positive about the introductory sections in their email feedback. However they sometimes suggested adding more to the definition of cognition; generally referring to their clinical knowledge and understanding of the complexities of cognition that they felt might not be fully captured.

The smiley / frowny faces were also included early in this introduction to support understanding of what was meant by the term 'bother' in PRECiS (see figure 3.4).

Figure 3.4: extract from introductory section including orientation to the term bother



3.8 Including exploratory 'moderator' questions

The qualitative work theme, 'impairment doesn't equal impact' (Patchick, et al., 2014) echoed the WHO ICF (WHO, 2001) model of health and functioning, and it led to the inclusion of five additional exploratory questions in PRECiS. These questions were not designed to be added to a total impact score of PRECiS. They were a means of exploring variables such as acceptance of

cognitive issues and perceptions of support, that might influence how highly a person rated the impact of their cognitive difficulties. The suggestion to include these questions came from the background research and qualitative work, as opposed to a suggestion from service users themselves. However, when the rationale for potential inclusion was given, stroke survivors understood why these questions were relevant to ask and endorsed their inclusion; they also gave feedback on wording for clarity. In the version of PRECiS used in the psychometric study, respondents rate their level of agreement (from 'not at all' to 'completely') with the following:

1. "I try to hide my problems with cognition from others." This was included since attempting to hide problems would potentially exacerbate them and effect perceived impact; others cannot provide practical or emotional support for a problem when it is hidden from them;
2. "I feel unsupported for my problems with cognition." Included since support networks are important in moderating the impact of any disease. Individuals who agreed with this statement might be more likely to rate bother more highly;
3. "I find it hard to accept my problems with cognition." Included as adjustment and acceptance are also important for recovery. Individuals who agreed with this statement might rate bother more highly;
4. "Other people say I have problems with cognition that I don't see." This was included as a means of exploring patient self-awareness; if individuals are being told that they have problems that they themselves are unaware of, it may be indicative of insight issues that could influence how ratings are interpreted.
5. "Other people don't understand the effects of my problems with cognition." This is similar to the second additional question (feel unsupported) in that it might help explain rated bother on the 27 core PRECiS items.

3.9 Acceptability and face validity

As described in section 3.1, the purpose of consulting with service users and prioritising their feedback throughout was due to the need for accessibility and acceptability in a measure (Fitzpatrick, et al., 1998; Gibbons & Fitzpatrick, 2012). The early stages of feedback changed the content of PRECiS considerably and by the end stages, service users did not suggest changes to improve accessibility nor addition of new items for added coverage. Overall, based on the feedback received, the measure appeared acceptable to users and to achieve the goal of exploring impact of cognitive difficulties.

Healthcare professional feedback was also broadly positive about the tool and its coverage although there were some concerns about whether respondents would be able to disentangle

the cognitive issues from the physical. However, many commented that PRECiS was filling a gap in outcomes available and that they would be keen to use it in their clinical practice.

In reducing the number of items to get to the 'bigger picture' (see section 3.4), the questions became broad. For example "family life" was subsumed in one item plus some explanatory text. Service user consultation and role play with former ACT NoW user group members supported this approach. The discussions around the interpretability of items informed development of a user guide to offer a structured approach to describing and defining items in more detail if required by respondents (see 3.10).

PRECiS was attempting to reach a balance between comprehensive and brief but this is a challenging mix. Whilst the final version of PRECiS had support from service user consultees, the acceptability, interpretability and coverage were all topics that needed to be explored in a larger sample of service users who had not been involved in its development (see Study 3: Psychometric Study).

3.10 Administrator guide

The feedback received from users suggested that, at least in the first instance, PRECiS would need to be tested more widely and facilitated by face-to-face meeting with a researcher to a) provide support where required and b) enable more feedback to be collected.

The consultation process also informed an associated guide to be used by researchers when supporting PROM completion. This included examples of how items could be further explained if participants were unclear (see appendix 5 from page 168).

3.11 The Result: PRECiS for field testing

A final draft of PRECiS with associated user guide was generated for field testing in the cross-sectional psychometric study. Appendix 5 (from page 168) shows the tool in full and it is described here:

PRECiS includes two introductory pages that first define the term cognition, then state the purpose of the questionnaire: to ask respondents to think about how cognition has effected them in the last week or so. It introduces the term ‘bothered’ with an unhappy face and advises that “if you have found ways to cope….” cognition may not bother at all (with a happy face shown).

27 items are then spread over six pages (using large font and spacing), asking respondents about the impact of cognition on four dimensions: skills (12 items); family and life participation (six items); mood (six items); and sense of self (three items). There is one question within the

'self' section that relates to perceived impact on their carer and asks whether respondents feel like they are a "burden." For each item, respondents first indicate whether they experience a problem or effect at all (yes/no) and if so, they rate the bother associated with that problem on a Likert scale from 0 (not bothered) to 4 (extremely bothered). A total score out of 108 is calculated based on the answers to these 27 questions. As per the requirements for accessibility in study 2 (systematic review) a Flesch-Kincaid Readability Analysis (performed at https://readability-score.com/) showed one item with a readability score less than 60 (57). Overall the readability analysis showed a mean score across all items of 75.3, which had been recommended in study 2 based on background literature (Agarwal et al., 2013; Sullivan & O'Conor, 2001).

Five additional questions are included on the back page of PRECiS booklet, designed to explore beliefs about cognition in general that might influence perceived impact and overall PRECiS score (see section 3.8).

PRECiS used large, bolded, coloured text and was professionally printed on heavy A4 paper and stitch bound in the centre to make the pages easy to turn for stroke survivors who may have limited dexterity. An alternative version was available if needed that used much larger text with one question per page and was spiral bound to facilitate perusal.

Study 3: Psychometric Study

PRECiS (Patient-Reported Evaluation of Cognitive State): a new measure for post-stroke cognitive rehabilitation

Presented in a format suitable for publication. This article was not of sufficient interest to the journal Stroke. It is currently undergoing revisions for submission to an alternative journal.

PRECiS (Patient-Reported Evaluation of Cognitive State): a new measure for post-stroke cognitive rehabilitation

Abstract

Background and purpose*:* PRECiS was developed through qualitative work, systematic review and service user consultation. It is a 27-item patient-centred, patient reported outcome measure (PROM) assessing perceived impact of cognitive problems. We sought to psychometrically assess PRECiS.

Methods: A cross-sectional, community-based psychometric study exploring acceptability, internal consistency, construct validity and reliability (including inter-rater reliability with informal carers as proxy respondents). A sub-sample was visited twice for test-retest reliability and opportunistic qualitative data on sensitivity to change were collected.

Results: 159 (visit 1) and 66 (visit 2) stroke survivors with a range of cognitive difficulties were analysed. PRECiS showed good acceptability (no missing values or floor/ceiling effects and minimal skewness); high internal consistency ($\alpha = 0.94$, indicative of potential redundancy); with moderate to strong construct correlations in the directions hypothesised (0.40 to 0.74). An ICC of 0.85 indicated good test-retest reliability. Where self-reported change had occurred from visit 1 to 2, PRECiS appeared sensitive: ANOVA to compare across 3 groups (self-reported change = none; positive; or negative) was significant ($p=0.001$). Using carers as proxy respondents is not supported by this analysis (inter-rater ICC = 0.43).

Conclusions: PRECiS is a patient-centred, practical and reliable measure to assess impact of cognitive problems from the unique perspective of stroke survivors. It may be useful for informing rehabilitation approaches and assessing their effectiveness. Further testing is required to assess removal of potentially redundant items and explore sensitivity to change.

Key Words: Psychometric; Outcome measures; Patient Centred; Cognitive disorders; Rehabilitation; Reliability and Validity.

Introduction

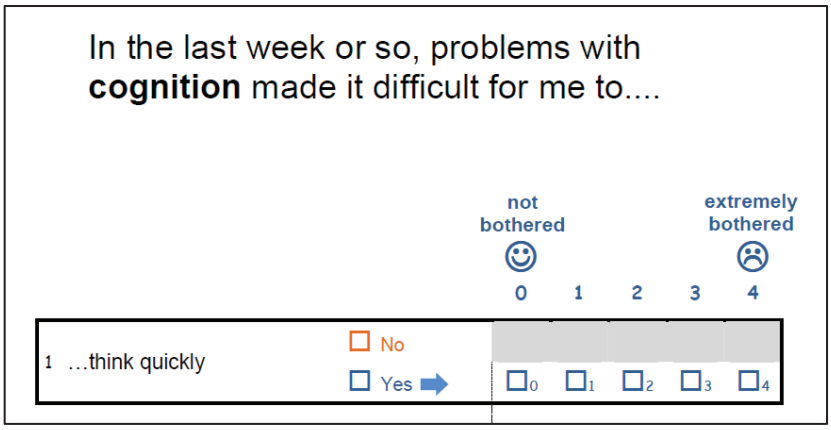
Stroke survivors are commonly left with a variety of cognitive issues that lead to adverse impacts on confidence, mood and long-term functional recovery[1-3]. Improving cognition is the number one priority for life after stroke research according to health professionals, carers and stroke survivors themselves[4]. Recent reviews conclude that - as well as significant gaps in the evidence base regarding how best to rehabilitate these issues - interventions rarely assess outcomes that are of 'real life' importance to patients or take their perspective on outcome; typically achieved through patient-reported outcome measures (PROMs)[5-6].

Individuals with cognitive issues are often systematically excluded from the development and use of PROMs due to their issues with comprehension and communication[7-8]. One of the more commonly used PROMs for cognition in stroke trials is the Cognitive Failures Questionnaire[9] but this is geared towards memory issues and was developed without input from service users. Overall, there is a lack of patient-centred outcome measures available for stroke trials[10].

To address this gap, we set out to develop a patient-centred PROM that would be suitable for cognitive rehabilitation trials in stroke. Qualitative work[11], systematic review of existing PROMs (paper in preparation) and pilot testing including patient and public involvement (PPI) with service users and carers, led to the development of the Patient-Reported Evaluation of Cognitive State (PRECiS).

PRECiS measures the perceived impact of persisting problems with cognition. Impact is conceptualised as 'bother' as opposed to frequency or amount of problem; a concept recommended by service users[11] and used elsewhere in stroke[12]. Its format and content are designed to facilitate its use with cognitively-impaired stroke survivors. It includes large-print text that is bolded and coloured for emphasis, as well as lay-friendly definitions of concepts such as 'cognition'. PRECiS includes 27 core items asking respondents about the impact of cognition on four dimensions: skills (12 items); family and life (six items); mood (six items); and sense of self (three items). For each core item, respondents first indicate whether they experience a problem at all and if so, they rate the bother associated with that problem on a Likert scale from 0 (not bothered) to 4 (extremely bothered). Five additional PRECiS questions ask respondents to rate agreement with statements that we hypothesised would influence rated bother. These statements include: whether respondents try to hide their problems with cognition; how supported they feel; how far they have accepted their problems with cognition; whether others tell them they have problems that they don't see themselves; and if they feel that others understand the effects of their problems with cognition. An example of a PRECiS core item is shown in Figure 1 overleaf.

Figure 1. Example item from PRECiS



Aim

The aim was to quantitatively and qualitatively assess psychometric properties of PRECiS including: acceptability to respondents; internal and external validity. Reliability was explored including test-retest and inter-rater (using informal carers as proxy respondents).

Methods

Design and participants

A cross-sectional, interview-based psychometric study. Participants were adults living in the community across two sites in England. Eligibility also comprised: being at least six months post-stroke (no upper limit) with self-reported ongoing difficulty due to cognition; an ability to provide informed consent and communicate in English through any communicative medium. Aphasia-friendly versions of the information and consent materials were available and the use of communication aids was encouraged. People were excluded if they had a known diagnosis of comorbidities leading to cognitive decline e.g. dementia. Adult English-speaking informal carers of recruited stroke survivors were also invited to participate in the study.

Participants were recruited through four routes between November 2013 and August 2014:

1. Primary care physicians sent letters of invitation to potentially eligible stroke register participants;
2. Healthcare professionals invited eligible stroke survivors;
3. The study was advertised using posters at participant identification centres;
4. Community stroke support groups were visited.

Appropriate ethics and governance approvals were obtained from the National Research Ethics Service, participating hospitals and primary care practices.

Procedures and measures

Participants were visited by one of two researchers in their homes to take part: one researcher was based in the North, one in the South of England. Demographic details and an indicator of disability (a modified Barthel Index[13]), were collected for stroke survivors and available carers. Stroke survivors also completed cognitive screening (the Montreal Cognitive Assessment (MoCA)[14], the Apraxia Screen of TULIA (AST)[15], the Star Cancellation Test[16], and the Frenchay Aphasia Screening Test (FAST)[17]). Stroke survivors then completed PRECiS with as much support from the researcher as they required. Support included introducing the questionnaire and its purpose; providing examples to support interpretation; using an alternative version of the questionnaire with one question per page. An administrator guide maximised consistency in PRECiS delivery and administrators were asked to record concerns about respondents' insight or comprehension. Carers completed PRECiS as proxy respondents independently, although support was provided if required. Participants gave their views on the coverage and acceptability of PRECiS following completion.

Comparison with other measures explored aspects of construct validity. To reduce burden, not all participants were asked to complete all comparison measures; we sought a minimum of N=50 respondents on each of the scales to be compared[18-19]. Respondents either completed measures of mood (the PHQ9 for depression[20] and the GAD7 for anxiety[21]) plus the Nottingham Extended Activities of Daily Living (NEADL)[22]; or they completed a stroke-specific quality of life tool, the Stroke Impact Scale (SIS)[23]. If participants were unable to complete the full 59-item SIS (typically due to fatigue or understanding) a short form composite version was available[24].

A sub-sample completed PRECiS a second time within two weeks. The sub-sample was purposively selected to include participants achieving a diverse range of scores at visit one to maximise variability. Qualitative data were collected to explore the assumption of stability across visits: asking participants about activities since visit one and whether they felt their mood and/or cognition were the same, better or worse.

If the visit(s) revealed significant distress and/or unmet needs, researchers provided information about local services available. Patients could be advised to make an appointment with their General Practitioner or referred to community services, such as those provided by the Stroke Association (www.stroke.org.uk).

Data analysis

Total and dimension PRECiS scores were computed by adding the ratings from relevant items i.e. total score was all 27 items, 'family and life' dimension was relevant six items. Each item was scored from 0 to 4, with a 'No' response (indicating no perceived difficulty) transformed to a 0; equivalent to a 'Yes+0' response (indicating that a perceived difficulty did not bother the participant at all). Maximum possible score on the 27 item scale was 108, indicating highest impact. Standard psychometric methods[25] were used to evaluate PRECiS, as follows:

Acceptability and practicality. Missing data for each item should be <10%, with remaining missing data imputed as the mean of non-missing responses. Skewness values should be <±1 for at least 75% of items. For floor/ceiling effects, no items should have >80% endorsement at the top/bottom extreme. Time to complete and qualitative feedback were also analysed.

Internal consistency. The extent to which items measure the same construct was explored using Cronbach's alpha (criteria of > 0.8) and item total correlations (criteria ≥ 0.2) across scale as a whole and dimensions.

Construct validity – internal. To explore validity of the four dimensions of PRECiS (skills; life; mood; and self), item convergence and item discrimination was assessed using Pearson product moment correlations between each item and the dimension total score. Item correlation with its proposed dimension should be 2 standard errors ($2/\sqrt{n}$) greater than correlations between the item and non-proposed dimensions[26]. Factor analysis with varimax rotation was performed.

Construct validity – external. Correlations between PRECiS and comparator measures were calculated. Hypotheses were: PRECiS would correlate most strongly with mood measures (PHQ9, GAD7 and SIS emotion subscale), given the well-documented relationship between cognition and mood[27-29]. Consistent with the World Health Organisation's framework for understanding the differential impact of disease[30], we predicted that perceived impact of cognitive issues would not necessarily map to measurable cognitive impairment given the many variables that moderate this relationship, including pre-morbid levels of cognition, and external support. As such, we did not specify a priori that a relationship would exist between the cognitive screen data and PRECiS data. To examine the value of the five additional PRECiS

questions described above, a linear regression used them as predictor variables for PRECiS score. We also included age, sex and time post-stroke as possible predictors.

Carer as proxy respondent (inter-rater reliability). The reliability of this approach was explored using intraclass correlation coefficients (criteria of >0.8). A linear regression explored whether "trying to hide cognitive issues" (PRECiS additional question) was a predictor for discrepancy between patient and carer scores.

Test-retest reliability and sensitivity to change. Intraclass correlation coefficients were computed (criteria of >0.8). Bland-Altman plots[19] highlighted participants with scores varying by ≥ 10% (> 10 points on the 108-point scale). Qualitative data collected at visit two categorised participants into groups reporting positive, negative or no change in cognition and/or mood. Change scores across groups were compared using ANOVA.

Results

Participants

Of the 235 stroke survivors referred to the research team, 164 (70%) were eligible and agreed to participate.

159 (97%) stroke survivor participants provided usable data for psychometric analysis. The five excluded from analysis chose to withdraw after being unable to complete PRECiS due to severe receptive aphasia and/or cognitive impairment. The 159 included had a wide range of scores on cognitive screens and measures of stroke severity (see Table 1 for characteristics). Despite all stroke survivors reporting cognitive difficulties that were often observable, some did perform at a level sufficient to pass screening tests. Many were employing strategies to do so, for example mnemonic strategies or deliberate scanning in star cancellation. We recruited stroke survivors with a range of ages (from 34 to 93 years) and times post stroke (from 6 months to 25 years).

Table 1. Characteristics of stroke survivors with usable PRECiS data (N=159)

Variable[¥]	N (%) / Mean (SD)
MALE	89 (56%)
Age (years)	65 (12)
Years since stroke	3.1 (3.7)
Ethnicity: White British	156 (98%)
Asian	1 (0.6%)
Black	2 (1%)
Employment: Retired	93 (59%)
Full/part/self/voluntary	28 (18%)
Unemployed / sick leave	28 (18%)
Other	10 (6%)
At least 12 years education	82 (52%)
Live with others	110 (69%)
Modified Barthel	16.8 (4.4).
MoCA	19.6 (5.5).
Cognitive Impairment Indicated (<26)	138 (87%)
FAST*	24.6 (5.4).
Aphasia indicated (score & age dependent)	67 (43%)
AST TULIA*	11.3 (1.3).
Apraxia indicated (<9)	7 (4%)
Star Cancellation †	51.1 (7.5).
Neglect indicated (≤44)	14 (9%)

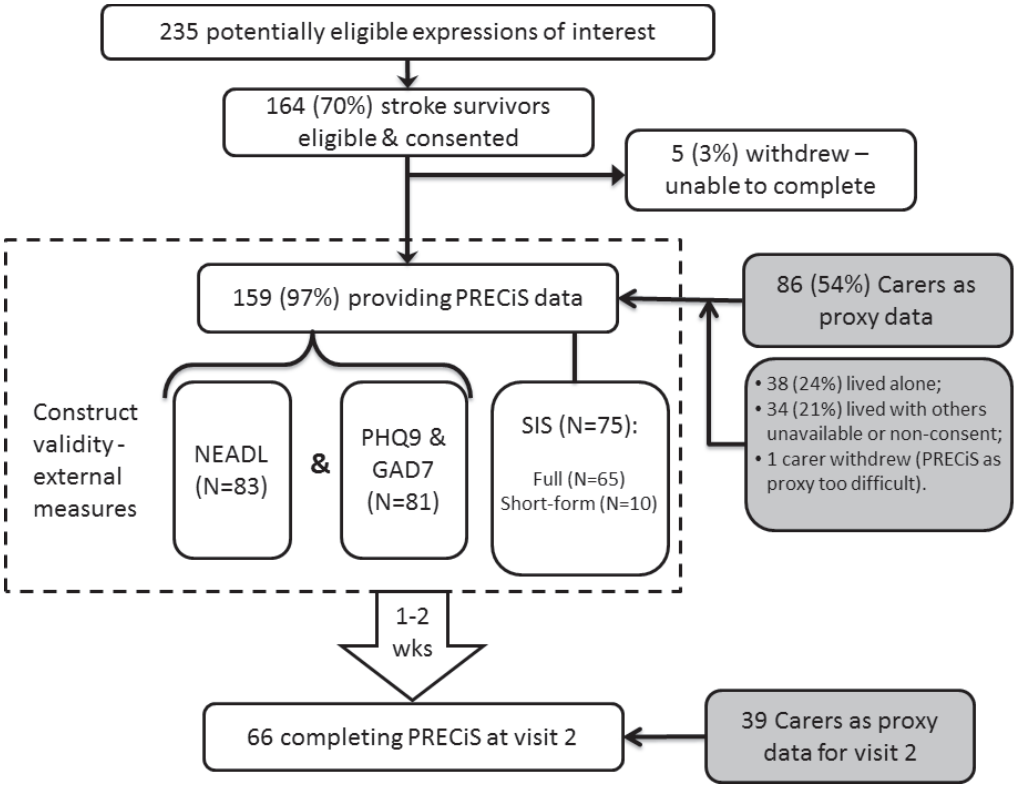
*1 participant refused to complete this screen

†2 participants could not complete this screen due to visual processing issues

¥Acronyms used: MoCA = Montreal Cognitive Assessment; FAST = Frenchay Aphasia Screening Test; AST TULIA = Apraxia Screen of TULIA (Test for Upper Limb Apraxia)

Carer-as-proxy data were available for 86 of the 159 stroke survivors participants (54%). For the 73 without carer-as-proxy data, 38 (24%) lived alone, 34 (21%) lived with others who were either unavailable during the visit (e.g. during working hours) or chose not to take part (e.g. using the visit as an opportunity to do independent errands) and one carer agreed to take part but found it too difficult to answer PRECiS as a proxy. The 86 participating carers were mostly female (n=59, 67%) and aged between 33 and 84 years old (mean = 61). The majority (n=70, 81%) were partners of the participating stroke survivor and 35 (41%) were working. Carers mostly had good functional abilities, with all carers achieving scores of at least 15 out of 20 on the modified Barthel. The flowchart in figure 2 summarises the number of participants and the measures they completed.

Figure 2. Flowchart showing number of participants and the measures they completed



Acceptability and Practicality

Table 2 summarises the psychometric properties of PRECiS For the 159 (97%) completing PRECiS, there was good acceptability with very few missing items and a median time to complete of 13 minutes. Adapted versions of material were used with 15 (9%) stroke survivors. Adapted materials were not sufficiently accessible for the five participants who were unable to complete PRECiS and withdrew. Whilst all other stroke survivor participants, including those with severe issues, completed PRECiS, researchers did note concerns about comprehension or insight for 31 (19%). Concerns about comprehension were primarily raised when participants appeared to contradict themselves throughout the visit. For example: expressing that they had difficulties with their memory in the early stages of the visit, then selecting that they had 'no problems' with memory in PRECiS (item 3). The administrator guide encouraged researchers to prompt participants to reflect on their answers in these cases but ultimately, answers were recorded as given by participants.

Missing data were minimal and there were no floor/ceiling effects; respondents made use of the full range of responses, showing minimal end aversion (see Figure 3). Skewness was an issue for 15% of items (N=4): three of the skills items (finding new places; using objects; and ordering actions) and item 16 (friendships) from the life dimension. All were positively skewed indicating that, they were highly endorsed and highly bothersome.

Table 2. Psychometric properties of 27-item PRECiS

Sample scores (max score possible): Mean (SD)

Total PRECiS 27 item (108): 40.2 (25.2)

Skills. 12 items (48): 16.6 (11.2) Life. 6 items (24): 9.1 (6.2)

Mood. 6 items (24): 9.7 (6.9) Self. 3 items (12): 4.6 (4.2)

Acceptability

Missing data (> 10% missing items): 0 Skewness (> ±1. n of items effected): 4 (15%)

Floor / Ceiling effects: 0 Minutes to complete. Mean: (SD): 14.9 (7.4)

Internal consistency. Cronbach's α: 0.94

Skills: 0.87 Life: 0.83 Mood: 0.71 Self: 0.79

Item-total correlation range. Pearson r: 0.33 to 0.75

Skills: 0.35 to 0.72 Life: 0.29 to 0.58

Mood: 0.49 to 0.72 Self: 0.55 to 0.71

Construct validity. Pearson r with PRECiS total score*

PHQ9 (N=81): 0.72 NEADL (N=83): -0.40 GAD7 (N=81): 0.47

SIS Physical (N=65): -0.48 SIS Communication (N=65): -0.56

SIS Emotion (N=65): -0.74 SIS Memory (N=65): -0.66 SIS Social (N=65): -0.67

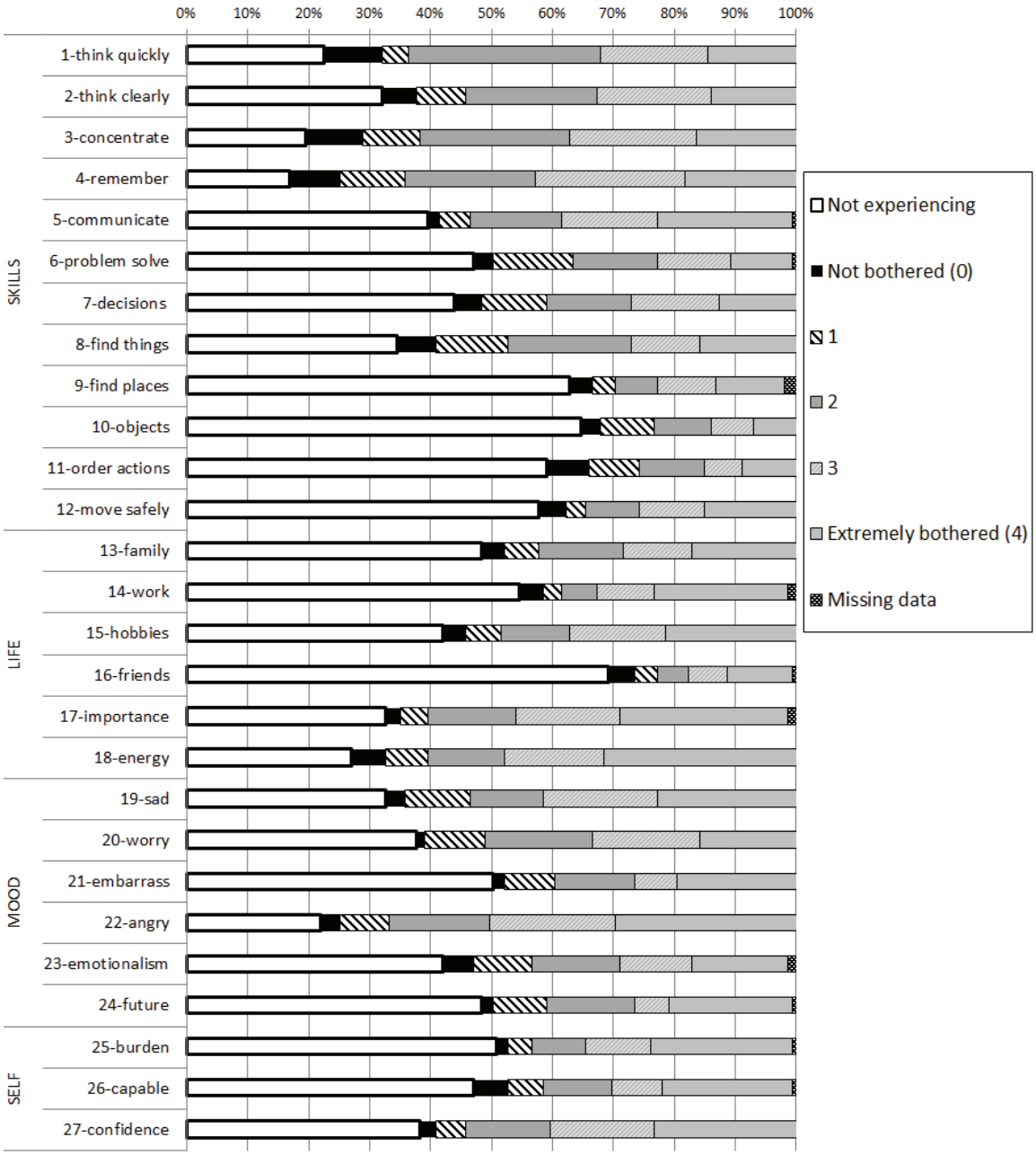
Short Form SIS Composite(N=10): -0.24

mBI (N=159): -0.29 MoCA (N=159): -0.18 FAST (N=158): -0.21

AST TULIA (N=158): -0.20 Star (N=157): -0.11

*Acronyms: PHQ9 = Patient Health Questionnaire-9; NEADL = Nottingham Extended Activities of Daily Living; GAD7 = Generalized Anxiety Disorder-7; SIS = Stroke Impact Scale; mBI = modified Barthel Index; MoCA = Montreal Cognitive Assessment; FAST = Frenchay Aphasia Screening Test; AST TULIA = Apraxia Screen of TULIA (Test for Upper Limb Apraxia); Star = Star Cancellation

Figure 3. Frequency of endorsement of ratings across each item



Qualitative feedback from the 159 participants who completed PRECiS was broadly positive. Participants mostly found the questionnaire content clear and easy to understand and respond to. They felt it was a good length and format, with good coverage of the topic; enjoying rating 'bother' as a relatable and endorsable concept. There was some critique of these dimensions as well. 34 participants (21%) commented on difficulties with the clarity of PRECiS; reporting that it was challenging to consider cognition outside of physical function and/or understand the term cognition, despite the pictorial memory aid with definition. Some found it difficult to consider the impact of chronic cognitive problems over the “last week or so” given how they had adapted to the condition but still felt bothered if considering a pre-stroke self. A small number

(N=15, 9%) felt that "bothered" wasn't an appropriate word as it alluded to caring; suggesting that "stressed" or "frustrated" might better capture impact.

Internal consistency

Cronbach's alpha was high at 0.94, with dimension α ranging from 0.71 to 0.87. The high α is suggestive of possible redundant items. Candidate items were items 2 (thinking clearly), 24 (feeling negative about the future) and 26 (feeling capable) due to high correlations (>0.6) with other conceptually similar items (respectively: concentration; feeling sad; feeling like a burden). Item-total correlations were all > 0.2 (ranging from 0.29 to 0.75).

Construct validity – internal

108 item/dimension correlations were explored for item convergence and discrimination relating to the four proposed dimensions (skills; life; mood; and self). Our items did not meet pre-specified criteria so the structure of the scale does not statistically map the four dimensions. The format and content of the measure was supported by participants in qualitative feedback; they found the different 'sections' intuitive.

Factor analysis demonstrated substantial loading (>0.4) of all items except items 9 (finding places, 0.38) and 14 (work, 0.35) on the first unrotated factor that explained 38.6% of variance. A 6-factor solution explained 61% of the variance but the content of factors on the rotated varimax solution did not map onto four conceptual dimensions. However, the content of these factors was moderately stable over the two visits, particularly for two factors, reflecting 'skills' and 'mood and self' dimensions. Items 13 to 18, representing 'impact on life' did not demonstrate stable factor loadings across the two visits.

Construct validity - external

The target for a minimum of N=50 for each tool to be compared was exceeded (see table 2). However, ten of the 75 participants (13%) selected to complete the Stroke Impact Scale (SIS) had difficulties completing the full version.

PRECiS correlated strongly with measures of depression PHQ9 (r= 0.72) and the emotion subscale of the SIS (r=0.74). Correlations were less strong with GAD7 measure of anxiety (r=0.47) and as hypothesised, lower still with NEADL (r=0.4). Correlations with SIS varied but, of all the subscales, the strongest correlation was emotion (-.74) and the lowest was physical (-.48). Relationships between PRECiS and baseline severity / cognitive screen data were weakest (r=0.11 to 0.29).

A regression model using age, sex, time post stroke and the five additional exploratory PRECiS questions explained 55% of the variation in total PRECiS score The strongest predictors in the model were acceptance of cognitive issues and the perception that others understood the effects of problems with cognition ($p \leq .01$). Younger stroke survivors rated more bother ($p=0.01$).

Carer as proxy respondent (inter-rater reliability)

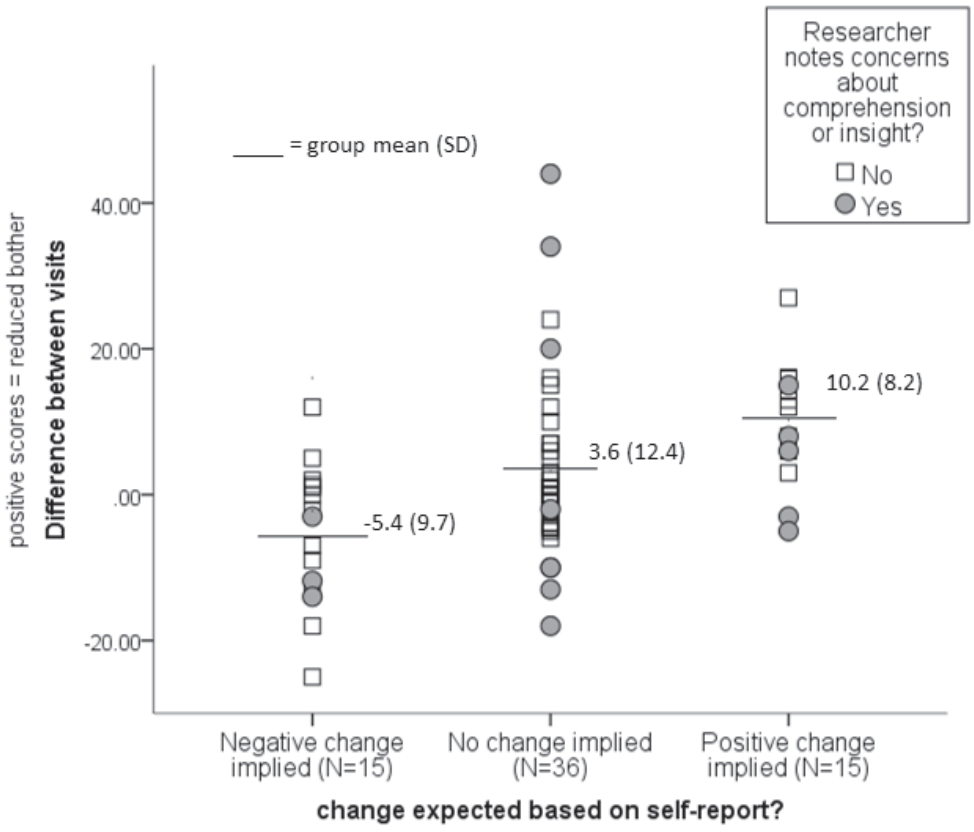
86 informal carers completed PRECiS as proxy respondents. There was low-to-moderate agreement between stroke survivors and carers (ICC = 0.43, 95%CI = 0.24 to 0.59). The responses to the PRECiS additional question asking for agreement with the statement "I try to hide my problems with cognition" predicted 58% of the variance in difference between stroke survivor and carer scores; higher agreement on this question meant larger discrepancy. Qualitative carer feedback suggested that whilst many completed the measure as a proxy with ease, they were not confident rating 'bother' as it was not directly observable.

Test-retest reliability and sensitivity to change

66 participants completed PRECiS a second time, within two weeks of visit one. PRECiS had high test-retest reliability (ICC = 0.85, 95%CI = 0.76 to 0.9). The mean difference in scores from visits one to two was 3 (SD=12.1). Bland-Altman plots revealed that, whilst the majority of participants (N=40, 61%), were within acceptable limits of change (maximum of 10 points difference on the 108 point scale), changes varied from -25 to +44 points on PRECiS across the sample as a whole.

Qualitative data collected at visit two related to changes in perceived impact of cognitive issues and/or mood. Participants were categorised into three groups for implying positive change (N=15), negative change (N=15), or no change (N=36). For example, P001 reported that visit one had prompted the re-uptake of taught strategies to overcome her cognitive issues and she had been pleased with the results (positive change). Major life changes had occurred for some participants between visits e.g. P002 workplace mentoring (positive change), P005 return to work (positive change), P057 dealing with house repossession (negative change). Figure4 shows change scores according to these three groups, including means and standard deviations. There was a statistically significant difference between the groups ($F(2,63)=7.65$, $p=0.001$), consistent with the subjective classification.

Figure 4. Scatterplot of raw changes in score against 'perceived changes' categories. On the Y-axis a positive number = a positive change / reduced bother ratings



Discussion

PRECiS is a new patient-centred PROM developed to measure perceived impact of cognitive difficulties. It has been evaluated in a population of community-dwelling adults in the chronic phase post-stroke with a wide variety of measureable cognitive impairment and self-reported cognitive difficulties, including those with and without aphasia. The analysis demonstrates that PRECiS is acceptable to potential users, is practical to use (interviewer-administered), has good reliability and construct validity, with some supportive evidence of sensitivity to change. PRECiS provides an important insight into the stroke survivor perspective; one that may help guide rehabilitation approaches as well as evaluate their effectiveness. This perspective may not be available to others, including carers with a close relationship; given the finding that carers are not reliable proxy respondents.

The analysis has proposed three items (number 2, 24, and 26) that may be candidates for exclusion. However, removal of these items only minimally effects α coefficient and participants reported positive face validity for all items. Thus the items have been retained and future work will include service user consultation around removal of these items as well as testing with a new sample to explore impact on psychometric properties.

We included people with a wide range of cognitive and communication difficulties who may otherwise be excluded from PROM development and completion[7]. The mode of administration – including administrator guide and alternative format to support understanding – allowed flexible delivery reflecting the varying needs of participants. The aim was to maximise motivation and ability to respond without influencing reliability. A more standardised mode of administration, such as by phone or mail, may not have been appropriate for this patient group. Future work will be required to generate guidelines for the minimum levels of cognition and insight necessary to complete PRECiS, since researchers reported concerns about understanding that may have influenced the reliability of scores for 31 (19%) included participants. In addition, five of the 164 (3%) initially recruited were excluded due to difficulties completing PRECiS. However, this compares favourably with 10 of 75 (13%) participants who were unable to complete the full SIS; a well-used scale in stroke.

The notion of insight and comprehension is a potential issue for any PROM that seeks the perspective of those with cognitive issues. It is a potential strength of this study that we collected qualitative feedback from stroke survivors, carers and researchers to support analysis of the data as a whole and we would recommend this approach for future use. Whilst the majority fed back that they found PRECiS simple to understand and use, a minority of participants highlighted potential ambiguities or difficulties. Some found it difficult to consider the impact of cognition outside of physical function. For example, severely physically impaired / house-bound participants might be bothered by difficulty maintaining relationships with friends, regardless of the effect of cognitive abilities on their friendships. However, the majority of respondents were positive about the approach and research in the field of cancer[31] and Parkinson's disease[32] have endorsed the viewpoint that cognitive issues impact dimensions of life that should be explored explicitly in measurement tools as well as rehabilitation approaches.

Our cross-sectional study was not designed to detect change. However, we wished to test the assumption that perceived impact would remain stable between two closely-timed visits. This might be particularly relevant in our population that were in the chronic stages post-stroke; rarely receiving care packages in the community for their issues. For these individuals, visit one could have negative implications by focusing attention on negative impacts of cognition. Conversely the process of measurement and discussion could be therapeutic in itself. Or, by chance, important life events could occur between visits that could legitimately have an effect on perceived impact. By asking simple questions at the beginning of visit two, we were able to opportunistically explore this in more detail and our findings suggest that PRECiS may be sensitive to change. However, changes were not always in the direction expected and participants would sometimes focus heavily on the timescale implications (rating "in the last

week or so") such that an incident in the short time between visits could be prominent in the memory and influence levels of bother. Future work must test sensitivity to change.

Previous work highlights the limitations of rating frequency of a problem; particularly for cognitively impaired individuals for whom accurate recall might be an issue[11, 25]. PRECiS uses 'bother' as an alternative, which may be susceptible to fluctuations due to variable emotional state. However, emotional state has been shown to significantly contribute to perceived recovery[33] and it would arguably effect any self-report measurement tool, given the level of psychological engagement required to complete a questionnaire. Improving emotional state and outlook would therefore be an important aspect of any rehabilitation intervention and PRECiS is equipped to detect change in these cases.

The sample was almost exclusively of white British ethnicity. This has implications for generalisability and future work would usefully include testing within a more ethnically diverse population. The mean age of 65 years old is relatively young for a stroke population[34]. However, a broad age range of individuals was invited to participate. Age was a significant factor influencing PRECiS scores (with younger stroke survivors reporting more bother) and it is possible that younger stroke survivors overall are more bothered by their cognitive difficulties – for example, if still of working age and unable to work. Younger stroke survivors may therefore have been more driven to participate in this study since they are the individuals who seek cognitive rehabilitation and, by extension, on whom PRECiS would eventually be used.

Summary

PRECiS is a practical and reliable measure of perceived impact of cognitive problems on aspects of ability / skills; life and family; mood and self. It is designed for interviewer administration, with associated guide available to allow flexibility of approach and support completion. Pending more data collection on responsiveness to change, it may be particularly useful for pragmatic trials of cognitive rehabilitation and have clinical utility.

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4 Discussion

This chapter provides a discussion of work as a whole to synthesise findings and suggest directions for future research.

The purpose of the research described in this thesis was to identify a patient-centred, patient reported outcome measure (PROM) for trials of comprehensive cognitive rehabilitation after stroke. Specifically, the work was carried out in stages including: qualitative study (study 1); review of PROMs (study 2); developmental / pilot work (chapter 3); and psychometric field study (study 3).

The aims, methods and findings from each of these stages are summarised below. The summaries here include details that were not included previously due to word limitations, given the need to make studies suitable for publication. Each study write up has necessarily contained some discussion due to the alternative format of this thesis. Discussions are expanded in this chapter; strengths and limitations of each part of the work are considered, including consideration of PRECiS as a tool. These inform possible avenues for future research (see section 4.6).

4.1 Qualitative study (study 1)

4.1.1 Aim

The first step was to understand service user perspectives on the important and measureable impacts of persisting cognitive problems in order to generate requirements for a PROM.

4.1.2 Method

Semi-structured interviews with stroke survivors experiencing cognitive difficulties. Recruitment methods are described in study 1 (published in Health Expectations (Patchick, et al., 2014)), and essentially involved self-referral to the research team. Thematic framework analysis derived themes to describe the data overall. These themes then informed recommendations for a PROM.

4.1.3 Findings & Discussion

16 stroke survivors were interviewed. Table 1 from study 1 shows participant characteristics.

With the help of the interview schedule, communication aids and training facilitated by PPI - stroke survivors articulated in-depth narratives about the impact of their cognitive problems. Seven emergent themes were identified. They are discussed below, and the ways in which they informed recommendations for the PROM are highlighted in **bold text**.

4.1.3.1 Theme 1: Hidden problems

"If people can see you're disabled, they understand" [P08].

The 'invisible' nature of cognitive problems would often lead to attempted masking of the problems and withdrawal from social situations. This could make the lived experience of them quite different to physical problems that were sometimes seen as better understood by others. Participants also implied that, on balance, cognitive problems are worse than physical difficulties due to their invisibility. Other qualitative work with stroke survivors has suggested the opposite; that physical impairments cause the greatest limitations to function and recovery (Ellis, Focht, & Grubaugh, 2013; Pound, et al., 1998). Impairments post-stroke will differentially effect individual stroke survivors, according to their unique life situation. With this in mind, the **recommendation for a patient-reported outcome for cognition that seeks subjective viewpoints** was endorsed.

4.1.3.2 Theme 2: Focus on underlying cognitive skills, not specific activities

Participants described personalised activities that were limited by their cognitive issues; from bird naming to cooking. Whilst the activities varied, participants recognised and articulated that a limited number of cognitive abilities underpinned a variety of activities. This informed the **recommendation for a PROM to include items that ask about a broad range of underlying cognitive abilities, not specific activities.**

This recommendation is potentially a contentious one; outcome measures used in rehabilitation typically include information on specific activities (Wade, 2003) and clinically, the finding that basic cognitive capacity underlies multiple activities is nothing new. But participants themselves articulate their impacts in terms of cognitive capacity; using specific activity limitations as *examples* of how these manifest in everyday life. If a PROM is to be applied to a heterogeneous sample in a research trial, it would be desirable to use language that is common to – and relatable for - multiple participants. For example, person A never reads but likes watching films. Person B is the opposite. Asking about limitations in reading and/or film watching is relevant for cognition generally but might only be relevant to one patient or the other; whilst asking about concentration would be pertinent for both. In addition, the ability to watch films or read is likely to be effected by abilities less related to cognition e.g. visual problems.

4.1.3.3 Theme 3: Damaged sense of self and limits to social participation

Damage to cognitive capacity often led to negative outcomes in terms of how people think about themselves and how they engage socially. As such, **a recommendation for a PROM is that**

it should include items that ask about the specific impact of cognition on self-identity and social participation.

It could be argued that this finding would not apply to all stroke survivors. For others, it may be physical abilities that contribute more to their perceptions of themselves. This echoes the discussion above from theme 1 (see 4.1.3.1). However, participants in this study were recruited because of their difficulties with cognition that they perceived were disrupting their lives. All interviewees reported that they would take up cognitive rehabilitation if offered. They are candidates for cognitive rehabilitation and, by definition, potential users of the developed PRECiS scale.

4.1.3.4 Theme 4: Emotional issues

Cognitive difficulties and negative emotion could exacerbate one another in a negative loop. The recommendation here was that **a measurement tool should address the specific impact of cognitive problems on mood**. Cognition was perceived to have an important relationship with mood that is observed in existing literature (Barker-Collo, 2007; Kauhanen, et al., 2000) and thus the sensitivity of a PROM might be maximised if questions asked directly about the impact of cognitive issues on these dimensions.

4.1.3.5 Theme 5: Impairment does not equal impact

The perceived impact of cognitive problems could be ameliorated by support networks, attitudes and environmental aids such that the relationship between levels of impairment / function and perceived impact was complex and individual. Impact was not about the frequency or severity of a problem but dependent on life situations and was typically discussed in terms of how much negative emotion it caused; how much “bother”, “upset” or “frustration” it led to.

The recommendation here was that a PROM should capitalise on this finding and measure impact directly by moving away from asking about frequency or amount of a problem and exploring aspects of 'bother' or 'frustration.'

4.1.3.6 Theme 6: Awareness of cognitive difficulties takes time

Stroke survivors tended to report that the perceived impact of cognitive issues became prominent later in stroke recovery. It would be important to offer cognitive rehabilitation at this stage, and to include a PROM to help evaluate it. This finding does not inform content of a PROM directly. It supports the original objective to focus on life after stroke within the PhD and, by extension to recruit participants who were classified as being in this stage (see methodology section 2.2). In addition, it underlines the importance of developing interventions and appropriate outcome measures that reflect the priorities and values of participating individuals. This can be achieved at least in part by involving those who could conceivably be end users of

the developed interventions or outcomes; an approach used in these qualitative interviews and the PhD work as a whole.

4.1.3.7 Theme 7: Perceived level of impact on carers:

Stroke survivors often reported how concerned they were about the effects of their cognitive difficulties on informal carers and family members; they worried about being a burden. When stroke survivors perceived this negative effect on others, it could feed back into their own negative feelings about themselves. **The recommendation here was that a PROM should include item(s) that explore patient perceptions of impact on carers, so that any changes in this area are not missed.**

4.1.3.8 Concluding remarks about the qualitative study

Qualitative work for instrument development is most readily used to obtain specific descriptive words and phrases that then become actual items (Rothman et al., 2009; Rowan & Wulff, 2007; Smith et al., 2005). The work described in this thesis did utilise the qualitative work for these ends, with a **recommendation that a tool should be accessible; including wording and items that respondents endorse and understand.** This is linked with patient-centredness and is an important factor in tool development generally.

In addition, the qualitative work was also used to generate recommendations in bolded text above that underpinned multiple aspects of face validity and conceptual underpinning of a required tool. The findings and recommendations from the qualitative work were fed back to participants during a dissemination event, where participants confirmed that they felt the findings were trustworthy and credible. The recommendations for a PROM were used to shape critical appraisal criteria for the review of PROMs (study 2) that is discussed in section 4.2.

4.1.4 Strengths & limitations of the qualitative study

The use of semi-structured interviews was agreed through PPI and allowed for in-depth exploration of topics that arose to ensure that, as recommended by Williams et al (2013), the derived measure captured information most relevant to patients in accessible terms. The semi-structured approach to interviews was designed to ensure that all points of exploration are addressed and, whilst it allowed room for discussion of emergent points, ultimately the questions asked will of course influence the answers given. Had the questions explicitly explored mechanisms for recovery or coping, it is likely that PRECiS would have ended up as a very different tool; perhaps one that took a more positive solution-based approach. Ultimately though, the interview schedule was derived through collaboration with service users and the findings deemed credible by interviewees at the feedback event.

The challenge of including stroke survivors with cognitive & communication problems in research means that they have been excluded from previous qualitative work (O'Connell et al., 2001; Pound, et al., 1998) and work developing PROMs (Dawson, et al., 2010; Kroll et al., 2012). One potential strength of this work is that relevant stakeholders have been included in the developmental work of PRECiS so that the end product is appropriate for its intended population (Scientific Advisory Committee of the Medical Outcomes Trust, 2002; Streiner & Norman, 2008).

Participants self-referred to the research team after being given initial study information either during their attendance at stroke community groups or through contact with healthcare professionals. Healthcare professionals were asked to keep a note of the number of individuals they had told about the study and the number who agreed to more information / their details being passed to the research team. However, these records were rarely kept up-to-date and leaflets were often left with individuals or stroke groups so that potentially interested parties could collect leaflets in their own time and decide whether to self-refer. This method had drawbacks as it meant that there were no data on uptake percentages e.g. the number who self-referred against the number who received information. Neither are there means of comparing the sample to non-sample on characteristics of interest. However, it was a pragmatic solution to identifying hard-to-reach community-based stroke survivors whilst giving them autonomy and time to decide whether to participate.

This route to recruitment also meant that participants were effectively self-selected for recruitment by virtue of them having concerns about their cognition. This limits generalisability to stroke survivors who may not have such concerns. However, those included are potentially individuals who would be more likely to seek cognitive rehabilitation and, by extension, be end-users of PRECiS.

The finding that participants included had limited ethnic diversity was highlighted and discussed previously in Study 1: Qualitative Study. This finding has implications for judging the validity of the conclusions drawn when considering other ethnicities and, by extension, the applicability of PRECiS as a tool. Future work would usefully explore whether PRECiS is acceptable to individuals of other ethnicities (see future research section 4.6.3).

The sample was also relatively young for a stroke population (Lee, Shafe, & Cowie, 2011), despite age being a driver for purposive sampling. A broad age range of people was invited to participate. However, it is possible that younger stroke survivors are more bothered by their cognitive impairments and thus, they may have been more driven to participate in this study. Again, a potential strength is that these may be the very individuals who seek cognition rehabilitation.

Theme seven in this study related to how stroke survivors perceive their cognitive difficulties as having a negative impact on informal carers and how this could feed back into their negative feelings about themselves. This led to the recommendation to include item(s) exploring perceived burden on carers in a PROM. However, given the important role that carers can play in rehabilitation efforts, another consideration is the importance of measuring carer impact from the perspective of carers themselves. Measurement tools do exist to explore carer impact e.g. the Carers of Older People in Europe (COPE) Index (McKee et al., 2003) and the Carer Strain Index (Robinson, 1983). However, it may be relevant to develop a measure for carers that, like PRECiS, explores the specific impact of cognitive difficulties. This is discussed below as an avenue for future research (see future research section 4.6.7).

4.2 Systematic review of available PROMs (study 2)

4.2.1 Aim

The aim was to identify whether any existing tools, perhaps from other neurological conditions, satisfied the recommendations for a PROM derived from the qualitative interviews, and might be used in stroke.

4.2.2 Method

Relevant sections in the methods chapter (see section 2.5) and study 2 detail the methods used. No terms restricted the search to stroke so that tools developed in other conditions would also be identified.

Eligible PROMs were reviewed in two stages. The first stage appraisal criteria were based on the user-derived recommendations from the qualitative study. Stage two considered other psychometric properties such as reliability and construct validity.

4.2.3 Findings & Discussion

No PROM was identified that met more than four out of the seven stage one assessment criteria, generated through qualitative interviews with stroke survivors (Patchick, et al., 2014). Their failure to meet these criteria has no implications for their validity in their intended uses (only one was developed in stroke and it was not cognitive-focused). However, it meant that other psychometric properties relevant to stage two assessment were not considered further.

Guidance for critical appraisal of tools often gives specific recommendations when considering quantitative aspects of psychometric properties e.g. reliability coefficients between 0.7 and 0.9 (Scientific Advisory Committee of the Medical Outcomes Trust, 2002); (Fitzpatrick, et al., 2006). Recommendations are less structured for assessing appropriateness and acceptability of a tool for a trial as these qualities tend to be more qualitative in nature and should be derived from

patient priorities and specific to the trial (Fitzpatrick, et al., 1998; Guyatt & Cook, 1994). Arguably the most important feature of a PROM is acceptability; if target service users find the PROM unacceptable, there may be issues in completion of the measure (Gibbons & Fitzpatrick, 2012). As Fitzpatrick (1998) states: "The selection of a patient-based measure for a trial therefore remains to some extent a matter of judgement and as much an art as a science."

The recommendations that were generated from the qualitative work were central for meaningful critical appraisal of appropriateness, acceptability, and conceptual underpinning of existing tools in this review. They allowed more confidence in the appraisal process of attributes that, as described, are more nebulous than their quantitative counterparts.

4.2.4 Strengths & limitations of the review

This review utilised a multi-faceted, broad search strategy to identify PROMs related to cognition, regardless of aetiology. Chapter 2 (section 2.5.1) highlights an alternative search strategy that was implemented early in the PhD – using trials as a proxy for identifying measurement tools – but that was found not fit for purpose. The rationale for the approach used in the study was to broaden the net and find tools that *may* be suitable for use in stroke, subject to some potential adaptation and validation. Only one eligible tool was developed for stroke and this may have contributed to the fact that no tools were judged to fully meet the assessment criteria. However, it did identify tools that might otherwise have been missed and indeed, that performed best against review criteria.

The search strategy only accessed published research or tools that were known by healthcare professionals and researchers in the field. It is possible that a tool has been developed but remains unpublished or unknown and thus was not identified. However, throughout the course of my PhD I have disseminated widely at local and national conferences dedicated to stroke (e.g. the UK Stroke Forum) and to outcomes development (e.g. COMET (Core Outcome Measures in Effectiveness Trials) meeting) and have often received positive feedback about the need for a tool like PRECiS.

Supervisors gave feedback on the approach, but ultimately, I was the only researcher that was involved in every aspect of the review. Guidelines for conducting systematic reviews describe the recommendation for a minimum of two reviewers (Liberati et al., 2009) to ensure rigour.

4.3 Development work (chapter 3)

4.3.1 Aim

To develop and refine a version of PRECiS to be used in the psychometric study.

4.3.2 Method

Chapter 3 provides information on the methodology used and describes how different aspects of PRECiS were influenced by synthesising many different sources of information.

4.3.3 The 'finding' – PRECiS

The 'result' of this work is the version of PRECiS that was used in the psychometric study (study 3). This is described in section 3.11, page 88 and included in Appendix 5 (from page 168). Consideration of the strengths and limitations of PRECiS as a tool is provided below in section 4.5; after consideration of the developmental work and the psychometric study.

4.3.4 Strengths & limitations of development work

The process of development was sequential and used discrete stages to develop a first draft of the tool (including qualitative study) and then used feedback from different groups (stroke survivors via stroke groups and healthcare professionals / researchers via email) to make refinements. The process limited the ability to return to previous stages and gain feedback from individuals who have given their opinion on other things / earlier versions. For example, individuals who took part in the qualitative interviews did validate the conclusions drawn from the qualitative research during a feedback event but they did not provide feedback on actual versions of PRECiS. A consensus process such as the Delphi method might have tackled this by allowing all participants to have multiple opportunities for providing feedback on different versions of the tool. It was felt that a Delphi process would not be suitable – particularly with stroke survivors with cognitive difficulties – for encouraging discussions on potential areas of improvements (as opposed to drilling down to options to be chosen / voted on). In addition, a Delphi process typically gives equal weighting to all respondents but the biggest decisions on refinements to PRECiS were made as a result of PPI with user groups. It could be critiqued that service users' opinions were weighted more heavily than healthcare professionals when making refinements. This was due to their position as potential future users of PRECiS and therefore it was felt that they were best placed to comment on its accessibility and acceptability; arguably the most important criteria for a PROM (see Chapter 1, section 1.8.1).

Guidance on involving the public in health suggests that it is beneficial to have a dedicated PPI group with pre-defined expectations and role descriptions (INVOLVE, 2012). Whilst a dedicated PPI group may have been useful in this research, every new community stroke group or organisation that was visited for feedback on the measure brought up different aspects of PRECiS that might have been improved. This made decision-making challenging but could be seen as a strength of the research. Feedback was elicited from multiple service users with different points of view about improvements to be made; and all relevant as potential users of

PRECiS due to their cognitive difficulties. That said, these stroke survivors were not formally screened for cognitive difficulties as that would not have been appropriate within the context of their involvement. Anecdotally though, it would be reasonable to say that those who were able to provide detailed feedback were less severely affected by cognitive impairment than some of those included in the subsequent psychometric study. The consultation work was driven by identifying community groups that agreed to take part in the process. On arrival at a meeting, some attendees did not wish to be involved in the process of providing feedback and they tended to be those who were more severely impaired and might have difficulty engaging in a group discussion. Feedback on PRECiS was sought in the psychometric study on a one-to-one basis; including with individuals who were more severely impaired.

One of the limitations of testing a measurement tool within a pilot setting that is highlighted by Streiner and Norman (2008) is that people may attend to items and consider questions in more depth than they would when completing as actual questionnaire respondents. This is a kind of Hawthorne effect that is hard to avoid when seeking detailed feedback on a tool to inform development. Feedback was additionally sought from actual respondents when PRECiS was field tested in the psychometric study. Data collectors also recorded observations on how individuals completed PRECiS and any areas that they appeared to find particularly easy or difficult and these are highlighted below when considering the strengths and limitations of PRECiS as a tool (see section 4.5).

Email feedback was received from 32 healthcare professionals and researchers out of around 100 recipients on the OPSYRIS mailing list. Whilst reminders were sent and responses encouraged, it was not possible to gain any opinions from those who did not respond. Focus groups with healthcare professionals would have been an alternative means of gaining feedback where all participants would have been encouraged to give their opinions; positive or negative. However, as per above, more effort was focused on seeking feedback from stroke survivors as end users of the tool.

4.4 Psychometric study (study 3)

4.4.1 Aim

To field test PRECiS in a large sample in order to gain quantitative and qualitative data on its psychometric properties.

4.4.2 Method

A cross-sectional, community-based psychometric study. Recruitment mainly involved self-referral to the research team. Study 3 gives detail about the procedures and measures involved.

The study was adopted onto the National Institute for Health Research (NIHR) Clinical Research Network Portfolio and Service Support Costs were secured from the Primary Care Research Network (PCRN). This helped secure Participant Identification Centre (PIC) sites in Greater Manchester to support recruitment. I was the sole researcher recruiting, carrying out home visits and collecting data in Greater Manchester for this study. In early 2014, recruitment was under-target and, after several sites had shown an interest in the study, I setup a second site based at North East London Foundation Trust (NELFT) that began recruiting in April 2014; again with one local researcher (an occupational therapist by background) collecting all study data.

4.4.3 Findings & Discussion

164 stroke survivors were recruited; (see Table 1 from Study 3 for participant characteristics). Study 3 contains detail on results and this section gives more discussion, followed by consideration of strengths and limitations of the study and of PRECiS as a tool in general.

As in the qualitative study, participants were almost exclusively white British. As such, all developmental work for PRECiS has been carried out within a limited ethnic group. Future work would be required to validate PRECiS within an ethnically diverse population (see future work section 4.6.3). Also similar to the qualitative study, participants in the psychometric study were relatively young for a stroke population. It was posited in the qualitative study discussion that younger stroke survivors may have been more driven to participate in this study (and potentially, by extension in cognitive rehabilitation) if they are more bothered by their cognitive difficulties. This argument is supported by the finding that age was an influential variable in a regression model to predict PRECiS scores; with younger stroke survivors reporting more bother.

A small proportion of recruited stroke survivors (5/164 (3%)) were unable to complete PRECiS due to comprehension issues. This compares favourably to completion rates for the Stroke Impact Scale (10/75 (13%) could not complete). Whilst study 3 data suggest good acceptability of PRECiS overall, future work would be useful to develop a potentially easier-access version with accompanying guidelines for minimum capacity required to complete (see future work section 4.6.2).

Three candidate items were identified as potentially redundant based on statistical analysis. In papers that describe validation of tools (Duncan, et al., 1999; Long, et al., 2008), items that are identified as redundant are often removed based on statistical analyses alone. Making changes to tool content based purely on statistical findings can lead to removal of items that are considered highly relevant by patients in terms of clinical impact (Juniper et al., 1997). As such,

a priority for future work is to carry out user consultation before deciding which items to remove (see future work section 4.6.1).

Unidimensionality was not necessarily a pre-requisite of PRECiS, since it was split into 'dimensions' assessing impact of cognition on skills (12 items); family and life participation (six items); mood (six items); and sense of self (three items; including one related to burden / perceived impact on carers). However, PRECiS does satisfy some threshold indices for unidimensionality (Streiner & Norman, 2008, p.317), which is an interesting finding and one that arguably supports the whole premise of PRECiS: that it is important that measurement tools explore the specific impact of cognition on important dimensions of life and mood. This premise came from the findings of the qualitative study (Patchick, et al., 2014) and is supported by research in other chronic conditions impacting cognition such as cancer (Ah et al., 2013) and Parkinson's disease (Lawson et al., 2014). It may be that 'impact of cognition' is a sufficiently cohesive construct that, even when it is explored across multiple dimensions, statistically it demonstrates unidimensionality.

This cross-sectional study was not designed to detect sensitivity to change since no substantive intervention took place between visits. However, the qualitative data collected at visit 2 provided a means of opportunistically testing the assumption of stability between two closely-timed visits (a requirement for test-retest reliability). This might be particularly relevant for a population that are several months or years post-stroke, rarely receiving care packages in the community for their issues. For these individuals, visit one could have negative implications by focusing attention on negative impacts of cognition. Conversely the process of measurement and discussion could be therapeutic in itself and encourage the re-uptake of compensation strategies. Or, by chance, important life events could occur between visits that could legitimately have an effect on perceived impact. The findings, as described in study 3, suggest that change scores in PRECiS were consistent with qualitative reports of change e.g. participants who reported negative changes in their mood or cognition at visit 2 tended to get higher scores on PRECiS at visit 2, indicating more perceived impact. Participants reported that they could not remember the ratings they had given for PRECiS in visit 1; suggesting this was not a deliberate change of rating to reflect their verbal self-report. The findings are indicative that PRECiS is sensitive to change and future work would explore this psychometric property (see future work section 4.6.6).

There was a lack of agreement between PRECiS scores for stroke survivors and carers-as-proxy respondents. This is a reflection of the subjective nature of PRECiS; providing a unique perspective of the stroke survivor. This highlights the importance of developing easier-access versions of PRECiS, that was suggested earlier in this section and is a topic for future research

(see future work section 4.6.2): if it is only patients themselves that can legitimately complete PRECiS, it must be acceptable to them and useable by them. This reiterates another point for future work highlighted earlier: that it may also be useful to develop a tool that is like PRECiS but for carer perspectives (see future work section 4.6.7).

4.4.4 Strengths and Limitations of the psychometric study

Some strengths and limitations have been addressed in course of discussing the findings e.g. the ethnically limited sample that suggests an avenue for future research (see future work section 4.6.3). This section addresses other strengths and limitations that have not yet been discussed.

As with the qualitative study, participants in the psychometric study self-referred to the research team. GPs and healthcare teams sent mailouts to potentially eligible participants inviting them to self-refer by returning a reply slip if interested. Due to research governance issues and data protection, no data were available to the research team on the characteristics of those who did not respond to mailouts. This means there are no data on the uptake percentages, or characteristics of the sample versus non-sample. However, as with the qualitative study it was a pragmatic solution to identifying hard-to-reach community-based stroke survivors.

The pragmatic approach to recruitment was also reflected in the decision to limit the number of comparison measures that each participant completed. Whilst this was a decision taken to reduce the burden of participation for stroke survivors who may fatigue easily, it did lead to a reduction in the precision of statistical comparisons between measures. The purpose of this study was to gain preliminary data on aspects of acceptability, reliability and validity; it could not reasonably have explored all possible constructs of interest in the one study. Exploring construct validity with higher numbers and different measures is an area for future research (see future work section 4.6.4).

The mode of administration that has been evaluated in this study involved face to face completion with support to maximise motivation and ability to respond. It may have contributed to the acceptability of the measure and high completion rates but it tends to be more resource-heavy than standardised methods such as postal or phone completion. However, a more standardised mode of administration may not be appropriate for this patient group. This needs to be explored further. Resource use is always an important consideration for trialists and this may limit how readily researchers designing a trial would select PRECiS as an outcome measure with its current validation. Future work would usefully explore for which participant groups other modes of administration for PRECiS would be appropriate (see future work section 4.6.5).

4.5 Strengths & Limitations of PRECiS as a tool

The quantitative and qualitative data presented in Study 3 shows that PRECiS was well-accepted by participants who provided useable data. However, as has been highlighted more work is required to develop aphasia-friendly versions of PRECiS to support completion with very severe difficulties and/or generate guidelines for minimum levels of capacity required to complete PRECiS (see future work section 4.6.2).

Whilst PRECiS was well-accepted and received positive feedback, there were some aspects pointed out by both participants and data collectors that relate to its interpretability. Some users did point out the difficulty of disentangling the cognitive impacts from other difficulties (see study 3 for feedback data). In addition, the nebulous concepts within some questions e.g. "family life," made interpretability difficult for some participants. As discussed in the development work (see chapter 3, section 3.9) the number of items were reduced to get to the 'bigger picture' and reach a balance between PRECiS being comprehensive yet brief. Whilst all questionnaires require cognitive effort to complete, the questions in PRECiS that require interpretation e.g. "what is family life to you?" may be particularly challenging for people with cognitive difficulties to complete. If so, PRECiS as it stands may well be best administered face-to-face with the administrator's guide, so that respondents can have standardised support to give their answers.

PRECiS asks users to rate "bother" to capture perceived impact. The term "bother" is utilised in other PROMs (Dean et al., 2013; Frank, et al., 2006) and was derived from stroke survivors themselves (Patchick, et al., 2014). It was considered a relatable concept by most of the 159 stroke survivors in the psychometric study (that were from both North England and South England). A small number of participants (N=15, 9%) did not like the term "bother" for capturing impact overall. In addition, "bother" may not maintain conceptual equivalence across different languages and cultures (Gawlicki et al., 2014). This may limit the cross-cultural utility of PRECiS; an avenue for future research (see future work section 4.6.3).

PRECiS did not demonstrate inter-rater reliability when carers were used as proxy respondents and did not have strong correlations with measured impairment. This reflects the subjective nature of PRECiS; providing a unique insight into stroke survivors perceptions of impact. Whilst this is not a limitation per se, it demonstrates that PRECiS scores may not be objectively 'verifiable' and, that being the case, it is more challenging to generate hard assumptions about how PRECiS scores would correlate to other measures. This has implications for validity testing (see future work section 4.6.4). It may also effect the clinical utility of PRECiS outside of a trial. In a trial, the random allocation ensures that all known and unknown factors influencing

outcome – in this example, PRECiS scores - are balanced across groups (Sibbald & Roland, 1998). For clinical purposes it is useful for a measurement tool to have hard data on relationships with other constructs and how scores can be interpreted clinically. That said, PRECiS could be a useful clinical tool when interviewer-administered; completion of the tool provides a means of assessing insight and raising awareness of cognitive issues. It also provides ramps for discussion to identify the influences of rated bother in order to address them in cognitive rehabilitation. Cognitive rehabilitation could address multiple underlying influences of impact (e.g. compensatory strategies; environmental aids; familial support) and reduce impact. PRECiS would potentially be sensitive to change, regardless of the mechanisms by which change occurs.

PRECiS development considered the look and formatting of the tool when presented to respondents and this led to the decision to print the questionnaire using a professional printing company using coloured ink in places and heavy paper to facilitate perusal. This has cost implications for producing PRECiS. Future work would usefully consider whether this level of professional printing is required for users or whether there is a cheaper and quicker alternative (see future work section 4.6.2).

4.6 Future research

4.6.1 Redundant items

Before any items are removed based on statistical analysis, qualitative feedback from service users will be sought about the clinical relevance of the items. Currently, PRECiS has 27 items giving a total score out of 108. 25 items and a total score of 100 would be a round number to aim for.

The psychometric study identified three items (number 2: thinking clearly; number 24: feeling negative about the future; and number 26: feeling capable) that may be candidates for exclusion due to redundancy, based on quantitative data (high correlations with other items). Personally, I feel that item 2 could be removed without loss as it is conceptually similar to 'concentration'. In addition, given the high inter-correlations of feeling 'negative about the future' with 'feeling sad', I believe that item 24 could also be dropped. The item for feeling 'capable' does have inter-correlations with feeling 'like a burden' but it was wording that came directly from the qualitative study and therefore may be worth retaining depending on user opinions.

A priority for future work would be to carry out user consultation to discuss removal of two out of these three items. The PPI links built throughout this research mean it is relatively straight-forward to arrange some user consultation to make these decisions. It would be important for

this issue of redundant items to be resolved before any further testing is carried out with PRECiS.

4.6.2 Developing alternative versions of PRECiS

A small number of psychometric study participants could not complete PRECiS in its current format and there were concerns about comprehensions for others. There was a version of PRECiS available that used large writing and had one question per page but more work would usefully be put into a full aphasia-friendly version of the tool that might lead to better uptake and fewer comprehension issues.

Future work will utilise PPI links developed throughout the study (particularly with groups that include people with aphasia such as Speakeasy) to generate an aphasia-friendly version of PRECiS. This could be carried out concurrently with consultation about the redundant items, since developing appropriate alternative versions of PRECiS is a priority before future validation testing can be carried out. These sessions could also be used to gain feedback on whether PRECiS could be formatted and printed in a way that did not necessitate professional printing at costly prices. For example, showing images in black and white and printing on standard A4 pages that are stapled on one side rather than stich-bound (in a format suitable for standard photocopying).

I envisage that developing these versions will take around five dedicated sessions of consultation that typically last half a day with lunch provided. It would involve going through the questionnaire item by item to gain feedback on interpretability and suggestions whether pictures are considered worthwhile to add (copyright of images would need to be carefully considered if so). User consultation sessions can be challenging to arrange in terms of finding premises and dates for attendance so this work may take several weeks to complete with different groups.

It may be possible to develop concurrent guidelines about levels of cognitive ability required for PRECiS completion. However, the purpose of PRECiS is to provide the patient perspective so arguably, it should be attempted with everyone, whatever their level of cognitive ability. As such, it may be more important to develop a structured reporting tool for researchers to document any concerns related to comprehension (assuming face to face completion). This could then be used to aid interpretation of PRECiS scores when analysed.

4.6.3 Generalisability across ethnicities and cultures

Primary data collection with research participants in both the qualitative and psychometric study included a sample that was almost exclusively White British. In addition, there is evidence that “bother” may not maintain conceptual equivalence across different cultures (Gawlicki, et

al., 2014). PRECiS would usefully be tested within a more ethnically and culturally diverse population to explore interpretability and accessibility.

Recruiting stroke survivors in the community was a challenge given that they were often not receiving any treatment and thus 'off the books' of health services. As such, narrowing inclusion criteria for recruitment to target particular ethnic or cultural groups is particularly challenging but could be achieved using targeted mailouts.

4.6.4 More data on construct validity

Whilst there are good data on face and content validity (within the tested samples) there are limited data related to aspects of construct validity. As discussed, the subjective nature of PRECiS and the multi-faceted understanding of impact present challenges for generating hard, testable assumptions about how PRECiS scores will correlate with other measures. PRECiS was primarily developed 'bottom-up' – from stroke survivors who were involved as research participants and consultees. It was less informed by 'top down' theory-driven approaches to measurement tool construction and did not hypothesise strict relationships between constructs.

The exploratory PRECiS questions provided *some* additional data on variables that might influence the perception and rating of impact (e.g. exploring acceptance and perceived support / understanding). These questions were influential in a regression model that explained 55% of the variance in PRECiS scores such that those who reported more acceptance of their issues and more social support, had lower PRECiS scores. It is feasible that these constructs could be explored systematically in validation testing for PRECiS; for example self-efficacy may relate to acceptance and could be explored using the Stroke Self-Efficacy Questionnaire (Jones, Partridge, & Reid, 2008) and the quality of support networks, could be explored by the Stroke Social Network Scale (Northcott & Hilari, 2013).

Further validation of PRECiS, including concurrent completion of these comparison measures could potentially be carried out within the context of a Masters research programme or perhaps be one study within a new PhD. A minimum of N=50 respondents would be desired to statistically explore correlations. There are possible avenues for setting up PRECiS validation studies as a Masters project within Nottingham University, following conversations with stroke researchers there. This option will also be explored within the University of Manchester.

4.6.5 Modes of administration

PRECiS has been validated as an interview-administered tool, taking a median time of 13 minutes to complete. It may be desirable if other forms of administration were validated that might reduce resource and time implications of collecting data face-to-face. Future work could test response rates and gain feedback on using PRECiS as a postal or telephone questionnaire in

stroke survivors with different characteristics e.g. varying levels of cognitive ability. However, it is possible that the face-to-face administration of PRECiS contributed to its high acceptability and good response rates with few missing data. Stroke survivors with cognitive difficulties may well need support to complete questionnaires and face to face support allows flexible administration reflecting the varying needs of participants.

As such, it would be worthwhile seeking anecdotal opinions from users about the suitability of using PRECiS as a postal or phone questionnaire whilst carrying out consultation described in section 4.6.1 to explore redundant items.

4.6.6 Sensitivity to change

Data are required on PRECiS' sensitivity to change over time. It would usefully be utilised within a trial of cognitive rehabilitation designed to reduce patient impact, in order to test this robustly. That is, to get responses on PRECiS both before and after an intervention to see if changes in scores occur in line with expectations. This would likely involve the use of other validated scores to help determine whether change has occurred.

There has been discussion with stroke researchers at Oxford University who are hoping to run a stroke rehabilitation trial in the near future. They are interested in PRECiS as a tool and there is some potential for PRECiS to be an 'add-on' to their trial for the purposes of gaining more data on validity. Researchers at the University of Manchester are also interested in PRECiS as a tool and may consider using it within a trial as an 'add-on' for further validation data (not as a primary outcome).

4.6.7 Tool exploring carer impact

PRECiS was designed as a tool to explore patient impact and data have shown that it is not a suitable tool for proxy completion by carers. However, the importance of carer impact was raised by patients themselves. There is scope for development of a tool like PRECiS but for carers of people with cognitive difficulties.

The qualitative study (study 1) was carried out as part of a larger research project where a separate researcher carried out similar interviews exploring impact with carers (Woodward-Nutt, et al., 2013). These carer interview transcripts can be analysed in a similar way to patient interview transcripts to develop a carer tool. This may take a dedicated study, in the same way as PRECiS required dedicated time to generate. A potential alternative route would be to explore whether PRECiS might be adapted to make it suitable for exploring carer's own viewpoints. This could potentially be achieved through PPI consultation specifically set up with carers of stroke survivors to see if adapting PRECiS for them appears credible for exploring impact. PPI consultation in a more focus-group format for seeking feedback can be achieved

more quickly than qualitative analysis of the carer interview transcripts and thus it might be the preferred route for developing a carer tool.

4.7 Concluding remarks

This PhD has used mixed methods and PPI and resulted in a new patient-centred, patient-reported outcome measure with intended eventual use in trials of cognitive rehabilitation in life after stroke. PRECiS brings together multi-faceted aspects of life that users defined as being impacted by cognition. The common underlying construct across the measure is essentially 'impact of cognition' and this is explored across a variety of dimensions via a manageable number of items; including them in one rating scale that has good acceptability to users and evidence of reliability and validity.

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Appendix 1: Bowen & Patchick co-authored book chapter

25 pages in total
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Chapter 16
Cognitive Rehabilitation and Recovery After Stroke

Audrey Bowen and Emma Patchick

Introduction

As the previous chapters have described in detail, many of those fortunate to survive their stroke do so with detrimental alterations to their cognitive and psychological well-being. These impairments impact the affected individual's ability to participate in, and benefit from, multidisciplinary stroke rehabilitation, to safely and independently carry out activities of everyday living, and to resume pre-morbid personal, social, and vocational roles [1–4]. Previously automatic and effortless tasks require exhausting levels of concentration and, despite the efforts invested, often end in perplexing and de-motivating failure. Uncertainty in one's own abilities and reliance on others makes people with cognitive problems vulnerable to frustration, humiliation, worry, and feelings of hopelessness. These topics are covered elsewhere in this book. The current chapter focuses on cognitive rehabilitation by exploring the evidence base from the perspective of informing clinical service improvements and strives to root cognitive recovery firmly within a broader psychological context.

> I couldn't understand why things were so much harder…I couldn't follow things. I worked before my stroke and was…am…an intelligent man, but didn't feel that way anymore. The tests were interesting for me…some bits were so easy, other bits just made me unravel… things I knew I should be able to do. It really helped me and my wife that the girls explained why this was happening…that it was the stroke, not me. I guess I felt it gave me some control to understand it…. Quote from person with stroke. Reprinted with permission from NHS Improvement -Stroke [31].

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DOI 10.1007/978-1-4614-7672-6_16,

Consensus on Prioritizing Psychological Problems

Stroke survivors often seek to express that they feel like a different person, their essence has changed, and their self-identity as well as esteem has been threatened, not necessarily by their hemiplegia or their hemianopia but by changes to the cognitive functions underlying their capacity for language, attention, spatial awareness, memory, and so on [5]. Families notice a difference too, although as they anecdotally report, it is the dysexecutive impairments altering social behavior that cause the greatest concern about having "lost" the person they knew. It is therefore not surprising that there is a consensus amongst people with stroke, their health service providers, and stroke rehabilitation researchers regarding the importance of the behavioral consequences of stroke.

Research into psychological problems was raised as a priority area by the National Stroke Strategy for England [6] despite, or perhaps because of, uncertainties regarding the most effective rehabilitation interventions. When stroke survivors were recently asked about their unmet needs following stroke, almost half of the 799 respondents reported problems with their mood and cognition [7]. Of those, a high proportion felt that issues such as memory and concentration had not been addressed appropriately, especially when compared with other issues such as mobility and pain. Similarly, the James Lind Alliance took a comprehensive and rigorous approach to identifying research priorities relating to life after stroke by consulting with stroke survivors, caregivers, and health professionals as well as searching relevant literature. They concluded that the number one research priority was investigating the best ways to improve cognition after stroke [8].

Quality of the Evidence Base for Cognitive Rehabilitation

One conclusion that might be drawn from the above is that there is very little existing research in cognitive rehabilitation. However, there is in fact an abundance of literature on the topic, and cognitive rehabilitation research is now well established with contributions from several fields including neuropsychology, cognitive psychology, clinical psychology, neurorehabilitation, occupational and speech and language therapy, and acquired brain injury. The full gamut of research designs are employed from qualitative methods exploring survivors' perspectives and priorities through the whole range of quantitative methodologies. The latter consist of single case designs and case series, cohort and case–control observational studies, experimental group designs (within and between subject controls) up to and including randomized controlled trials, and the recent emergence of health economic evaluations. Readers interested in the topic of research design for the evaluation of complex interventions such as cognitive rehabilitation are referred to the framework proposed by the Medical Research Council [9, 10].

Perhaps this abundance of evidence is the problem. How do those charged with improving national and local clinical services extract the most relevant and reliable

research, especially where it appears contradictory? The two most internationally accepted methods of evidence synthesis for clinical service development are the Cochrane Collaboration's established systematic review and meta-analysis, disseminated widely throughout the world via the web-based Cochrane Library [11]; and the national clinical guidelines/recommendations for stroke now produced and regularly updated by a growing number of countries, e.g., Australia [12], Canada [13], the UK (except Scotland) [14], and a separate guideline for Scotland [15]. Cochrane reviews employ a tried and tested formula for systematic searching to extract and include published and unpublished data that meet agreed quality standards, thereby reducing the risk of bias. This usually restricts the review to evidence collected from well-conducted randomized controlled trials.

From Cochrane Reviews to National Clinical Stroke Guidelines

Cochrane reviews of cognitive rehabilitation that focus on dysfunctions such as neglect, apraxia, memory, perception, and attention problems exist, and others—such as those concerned with executive dysfunction—are close to publication. The Cooksey review of UK healthcare research highlighted two problematic "gaps" that hold back clinical service development in healthcare generally [16]. One of the gaps is relevant to cognitive rehabilitation and is specifically concerned with how we transfer research evidence into clinical knowledge or clinical practice.

Assumptions that data/evidence and knowledge are one and the same are naïve, as is the expectation that clinicians can and will automatically implement published evidence and evidence syntheses into practice. National clinical guidelines seek to address this gap [12–15]. They perform the essential translator role, producing recommendations for implementation into clinical practice based on high quality searching, evidence appraisal, and consensus level agreement. Where evidence is missing, recommendations are formulated around expert opinion and good practice points. Often they also complete the loop by conducting national audits of adherence to the recommendations [17, 18]. This can help by highlighting areas of practice in need of greatest improvement such as the area of psychological needs, including cognitive rehabilitation, in England [7]. The Canadian guideline (i.e., their Stroke Strategy: Best Practice Recommendations) explicitly includes helpful links to "Implementation Resources and Knowledge Transfer Tools" for each topic within stroke care [13, 19].

Aims of This Chapter

There are now several excellent textbooks [20, 21], journal review papers [22, 23], and Cochrane reviews on post-stroke cognitive rehabilitation that can be referred to for detailed descriptions of both the interventions and the studies that evaluate their

efficacy [24–30]. The current chapter describes and compares the recommendations for cognitive rehabilitation currently advocated in various National guideline, which themselves are heavily influenced by the Cochrane reviews and randomized controlled trials. We review each cognitive area and conclude with what has been termed "comprehensive holistic neuropsychological rehabilitation." The evidence base for this borrows heavily from the traumatic brain injury literature but suggests a pragmatic way forward for stroke rehabilitation services. The final issues considered will be service organization and the workforce needed to deliver effective cognitive rehabilitation, with reference to the recent National Health Service (NHS) Improvement Program's useful stepped care model of improving stroke services for people with cognitive and mood problems in England [31].

Cognitive Rehabilitation: Screening and Assessment

The most striking common feature of the clinical guidelines is their emphasis on screening and assessment to elicit underlying cognitive impairments and determine the likely functional and personal impact for each individual with stroke. In some guidelines a larger proportion of the recommendations focus on assessment compared to restorative or compensatory interventions (e.g., Scottish). Providing explanations to demystify patients and caregivers is often a core recommendation and the rationale for this is illustrated in the previous quote from a person with stroke [31]. The following definition of cognitive rehabilitation from the Scottish guideline places this message up front. It also highlights the current paucity of evidence for the benefits of assessment [15]. Although the Scottish guideline writers raise a valid methodological concern with the one existing study, the practical and cost implications of using qualified psychologists rather than assistants would need careful consideration.

> Cognitive rehabilitation concerns efforts to help patients understand their impairment and to restore function or to compensate for lost function (e.g., by teaching strategies) in order to assist adaptation and facilitate independence….When cognitive problems are suspected and relatives report personality change, the patient can be referred to a clinical psychologist to provide assessment and where appropriate, psychological intervention which may include career education and support. One [randomized controlled trial] found a trend only toward reduced [caregiver] strain when this service was provided. Assistant psychologists, not fully trained clinical psychologists, were used in this study. Reprinted with permission from Scottish Intercollegiate Guidelines Network [15]

Key recommendations on the topic of screening and assessment have been extracted and presented in Table 16.1. These include the reminder that assessment should determine a person's cognitive strengths and not just their impairments. The stroke team needs to be informed regarding the person's learning potential and how best to maximize that, not just for the rehabilitation of their cognitive difficulties, but as an "integral part of the [multidisciplinary] rehabilitation plan" [15]. Other recommendations common amongst guidelines concern balancing the utility of

Table 16.1 Recommendations from National Clinical Guidelines: screening and assessment for cognitive problems (selected extracts)

Australia	a) All patients should be screened for cognitive and perceptual deficits using validated and reliable screening tools. b) Patients identified during screening as having cognitive deficits should be referred for comprehensive clinical neuropsychological investigations.
UK[a]	A. Interventions or patient management should be organised so that people with cognitive difficulties can participate in the treatments and regularly reviewed and evaluated. B. Every patient seen after a stroke should be considered to have at least some cognitive losses in the early phase. Routine screening should be undertaken to identify the patient's broad level of functioning, using simple standardised measures (e.g. Montreal Cognitive Assessment MOCA). C. Any patient not progressing as expected in rehabilitation should have a more detailed cognitive assessment to determine whether cognitive losses are causing specific problems or hindering progress. D. Care should be taken when assessing patients who have a communication impairment. The advice from a speech and language therapist should be sought where there is any uncertainty about these individuals… E. The patient's cognitive status should be taken into account by all members of the multidisciplinary team when planning and delivering treatment. F. Planning for discharge from hospital should include an assessment of any safety risks from persisting cognitive impairments. G. People returning to cognitively demanding activities (e.g. some work, driving) should have their cognition assessed formally beforehand.
Scotland	A full understanding of the patient's cognitive strengths and weaknesses should be an integral part of the rehabilitation plan. *Screening* Short, standardised cognitive screening measures can be used by a health professional with knowledge and experience of the presentations of cognitive functioning and factors influencing it. They can be used as a broad screen to reduce the possibility that problems will be missed and as a measure of progress. It is important for staff to understand that these screening measures will miss some of the cognitive problems which can be most important for rehabilitation and eventual functioning. These are varied but can include such issues as poor awareness of deficits or their implications, slowing of information processing, and the ability to cope with distraction. Care needs to be taken in selecting measures for use with people who have communication difficulties and, ideally, the selection should be made in collaboration with a speech and language therapist. *Assessment* Screening measures do not provide information about the depth and nature of the patient's problems or strengths and therefore do not constitute an assessment sufficient for rehabilitation planning or for establishing suitability for a particular work role (e.g. operating machinery). Administering and interpreting full assessment results requires specialist training and should be carried out in the context of clinical interviews with access to background information. Stroke patients should have a full assessment of their cognitive strengths and weaknesses when undergoing rehabilitation or when returning to cognitively demanding activities such as driving or work. Cognitive assessment may be carried out by occupational therapists with expertise in neurological care, although some patients with more complex needs will require access to specialist neuropsychological expertise.

(continued)

Table 16.1 (continued)

Canada	1. All high-risk patients should be screened for cognitive impairment using a validated screening tool. 2. Screening to investigate a person's cognitive status should address arousal, alertness, attention, orientation, memory, language, agnosia, visuospatial/perceptual function, praxis and executive functions such as insight, judgment, social cognition, problem solving, abstract reasoning, initiation, planning and organization. 3. The Montreal Cognitive Assessment is considered more sensitive to cognitive impairment than the Mini Mental Status Exam in patients with vascular cognitive impairment. Its use is recommended when vascular cognitive impairment is suspected. Additional validation is needed for the Montreal Cognitive Assessment as well as other potential screening instruments such as the 5-min protocol from the Vascular Cognitive Impairment Harmonization recommendations. 4. Post-stroke patients should also be screened for depression, since depression has been found to contribute to cognitive impairment in stroke patients. A validated screening tool for depression should be used. 5. Post-stroke patients who have cognitive impairment detected on a screening test should receive additional cognitive and/or neuropsychologic assessments as appropriate to further guide management.

[a]Covers all of the UK except Scotland, which has a separate guideline

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- National Stroke Foundation. Clinical Guidelines for Stroke Management; 2010. Melbourne, Australia [12]
- Intercollegiate Stroke Working Party. National clinical guideline for stroke, 4th edition. London: Royal College of Physicians, 2012 [14]
- Scottish Intercollegiate Guidelines Network (SIGN). Management of patients with stroke: Rehabilitation, prevention and management of complications and discharge planning. A national clinical guideline. Edinburgh: SIGN, 2010 [15]
- Lindsay MP, Gubitz G, Bayley M, Hill MD, Davies-Schinkel C, Singh S, and Phillips S. Canadian Best Practice Recommendations for Stroke Care (Update 2010). Prepared by the Canadian Stroke Strategy Best Practices and Standards Writing Group, on behalf of the Canadian Stroke Strategy (a joint initiative of the Canadian Stroke Network and the Heart and Stroke Foundation of Canada). 2010; Ottawa, Ontario, Canada: Canadian Stroke Network [13]

brief screening tools against consideration of their limitations, when to refer for more detailed assessment and by whom. Examples of useful tools are given in some guidelines. The Canadian and the recent update of the UK (except Scotland) guidelines suggest the Montreal Cognitive Assessment as a simple, standardized screening tool. The latter suggests more detailed assessments within later sections covering specific cognitive impairments (Table 16.1).

Timing and Workforce Mobilization: Cognitive Screening and Assessment

Workforce competencies for cognitive screening and assessment require careful planning as does the timing of these activities, which should influence clinical decision-making and outcomes for people with stroke, without using valuable

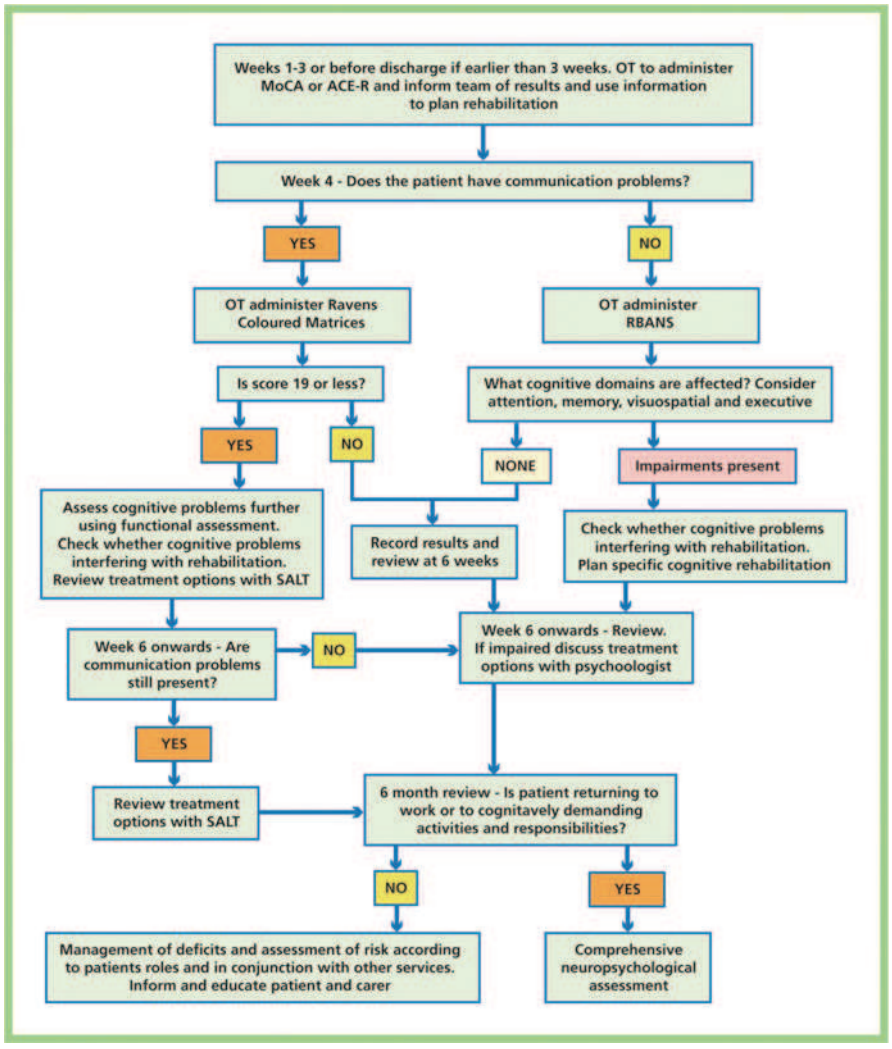

Fig. 16.1 Pathway for assessing cognitive problems. Reprinted with permission from Gillham S, Clark L. Psychological care after stroke—improving stroke services for people with cognitive and mood disorders. NHS Improvement—Stroke, 2011. http://www.improvement.nhs.uk/stroke/Psychologicalcareafterstroke/tabid/177/Default.aspx

resources to simply confirm the obvious (i.e., most acute stroke patients will have some cognitive impairment). Investigations should provide more information than a simple "cognitive impairment absent/present" tick box. Guidelines emphasize the roles of occupational therapists and psychologists. A recent document from the NHS Improvement Stroke program for England [31] suggests a pathway for assessing cognitive problems by way of the first step towards cognitive rehabilitation (Fig. 16.1). As shown, key time points in the UK model are: pre-transfer of care

from hospital to community at 6 weeks and 6 months. The latter review is recommended for identifying long-term problems persisting beyond the period when much spontaneous recovery has occurred. For some people with stroke, this can also be a significant time during which they appreciate the extent of their residual cognitive difficulties and the need to adjust and accept compensatory rehabilitation strategies and aids. Canada recommends the following more frequent cognitive screening/assessment regime (and extends this to those who have had a transient ischemic attack) "at various transition points throughout the continuum of stroke care [13]":

1. During presentation to emergency when cognitive, perceptual, or functional concerns are noted.
2. Upon admission to acute care, particularly if any evidence of delirium is noted.
3. Upon discharge home from acute care or during early rehabilitation if transferred to inpatient rehabilitation setting.
4. Periodically during inpatient rehabilitation stage according to client progress and to assist with discharge planning.
5. Periodically following discharge to the community by the most appropriate community healthcare provider according to client's needs, progress, and current goals.

Beyond Assessment: General Cognitive Rehabilitation

The National guideline differ slightly in how they treat the management of cognitive problems after assessment. Rather than covering general cognitive rehabilitation most (e.g., Australia, Scotland, and UK except Scotland) go straight to domain-specific advice (e.g., interventions for memory and neglect). These often include recommendations of assessment tools specific to that impairment but the point here is that they also cover restorative and compensatory techniques. The Canadian guideline includes recommendations for the rehabilitation of cognitive problems as a single collective (see Table 16.2). This includes the broadest range of interventions including psychopharmacology (not reprinted here, see full report [13]) since this guideline covers "vascular cognitive impairment and dementia."

Domain-Specific Recommendations

The Australian, Scottish, and UK (except Scotland) guidelines take the approach of dividing cognition into specific impairments. Recommendations for attention, memory, neglect, and aphasia are covered by all. Apraxia and executive functions are included in the UK (except Scotland) and Australian guidelines. Agnosia is specifically covered by the Australian guideline whilst the most recent guideline (UK with the exception of Scotland) makes recommendations more broadly on perception. Space does not permit detailed coverage of all eight domains. The approach taken

Table 16.2 Canadian recommendations: interventions for general cognitive problems (extracts)

Patients who demonstrate cognitive impairments in the screening process should be referred to a healthcare professional with specific expertise in this area for additional cognitive, perceptual and/or functional assessments. • Additional assessments should be undertaken to determine the severity of impairment and impact of deficits on function and safety in activities of daily living and instrumental activities of daily living, and to implement appropriate remedial, compensatory and/or adaptive intervention strategies. • A team approach is recommended, and healthcare professionals may include an occupational therapist, neuropsychologist, psychiatrist, neurologist, geriatrician, speech–language pathologist or social worker.
An individualized, patient-centered approach should be considered to facilitate resumption of desired activities such as return to work, leisure, driving, volunteer participation, financial management, home management and other instrumental activities of daily living.
Intervention strategies including rehabilitation should be tailored according to the cognitive impairments and functional limitations as well as remaining cognitive abilities, as identified through in-depth assessment and developed in relation to patients' and caregivers' needs and goals.
Strategy training provides individuals who have limitations in activities of daily living with compensatory strategies to promote independence and should be offered to patients with cognitive challenges. The evidence for the effectiveness of specific interventions for cognitive impairment in stroke is limited and requires more research. • Attention training may have a positive effect on specific, targeted outcomes and should be implemented with appropriate patients.
Compensatory strategies can be used to improve memory outcomes.

Extracts reprinted with permission from Lindsay MP, Gubitz G, Bayley M, Hill MD, Davies-Schinkel C, Singh S, and Phillips S. Canadian Best Practice Recommendations for Stroke Care (Update 2010). Prepared by the Canadian Stroke Strategy Best Practices and Standards Writing Group, on behalf of the Canadian Stroke Strategy (a joint initiative of the Canadian Stroke Network and the Heart and Stroke Foundation of Canada). 2010; Ottawa, Ontario, Canada: Canadian Stroke Network [13]

has been to extract the relevant information into tables to enable comparisons between guidelines. The reader is referred to the original documents for specifics on the studies on which these recommendations were made.

Although this modularized approach to cognitive rehabilitation is an oversimplification intended to aid clarity, it is also a true reflection of the design of the majority of the rehabilitation studies, which focus on a single impairment (e.g., neglect). In clinical practice, rehabilitation acknowledges that each cognitive domain, such as perception, attention, and memory, cannot be considered in isolation, as most everyday activities draw on a range and interaction of cognitive abilities.

Attention/Concentration

Each of the four guidelines mentions the pivotal role played by attention and the impact of attentional impairments. The ability to select and concentrate on relevant information or events is fundamental to everyday life. When this ability is impaired,

other cognitive skills will be affected. Attention can therefore be considered a "mediator" or starting point for many aspects of cognition. Attentional deficits have an acute negative impact on functional ability [32–34].

Trials of rehabilitation of attention involve a number of different approaches. Computerized rehabilitation has been used; this allows repetition of tasks that draw on attention [35–37]. Approaches also focus on practice and development of specific strategies for time pressure management (TPM) [38, 39]. TPM is an intervention directly aimed at behavioral and cognitive change in treatment situations that are designed to mirror real-life situations. The goal is to develop alternative cognitive strategies to compensate for mental slowness. Attention process training (APT) has also been used [40, 41]. APT is "a theoretically based, hierarchical, multilevel treatment, including sustained, selective, alternating, and divided attention" [40].

A Cochrane systematic review of attention [24] concluded that there was no evidence to refute or support the use of specific rehabilitation techniques for attentional impairments that improve functional independence after stroke. An update to this review is in progress. The latest update to Cicerone's review of cognitive rehabilitation for attention impairments [23] made practice standard recommendations for interventions for traumatic brain injury but this may well be applicable to stroke. The UK (except Scotland) guidelines, the most recently updated of all the guidelines, make recommendations based mainly on consensus opinion and a recent underpowered randomized controlled trial [39] of TPM (see Table 16.3). Although inconclusive, the latter trial suggests that TPM shows promise with younger, more physically independent stroke survivors and that it is feasible to train staff to deliver TPM in hospital or community stroke services.

Overall, there is a lack of high quality trials to inform selection of specific interventions and much of the evidence is at consensus level. Adequately powered randomized controlled trials of TPM and other interventions (e.g., APT) would greatly improve the evidence base for these commonly disabling impairments (Table 16.3).

Memory

Memory impairments (see Chap. 8) are related to a general reduction in functional ability for everyday tasks, even after factors such as age and stroke severity are taken into consideration [42]. Memory impairments also are upsetting for family members who cope with the consequences of forgetfulness; caregiver well-being correlates negatively with a patient's memory problems [43]. The following simple three-step model has been advocated as useful for explaining and offering interventions to rehabilitate the effects of memory impairments:

1. Encoding—organizing and processing information for later recall. Encoding may happen consciously or unconsciously.
2. Consolidation—the process by which a piece of information becomes stored in memory in a more permanent way.
3. Retrieval and recognition—recalling previously encoded and consolidated information in a meaningful way [44].

Table 16.3 Recommendations from National Clinical Guidelines: Attention (extracts)

Australia	Cognitive rehabilitation can be used in stroke survivors with attention and concentration deficits
Canada	The evidence for the effectiveness of specific interventions for cognitive impairment in stroke is limited and requires more research • Attention training may have a positive effect on specific, targeted outcomes and should be implemented with appropriate patients
Scotland	There is not yet sufficient evidence to support or refute the benefits of cognitive rehabilitation for patients with problems of attention
UK[a]	A. Any person after stroke who appears easily distracted or unable to concentrate should have their attentional abilities (e.g. focused, sustained and divided) formally assessed B. Any person with impaired attention should have cognitive demands reduced through: – having shorter treatment sessions – taking planned rests – reducing background distractions – avoiding work when tired. C. Any person with impaired attention should: – be offered an attentional intervention (e.g. Time Pressure Management, Attention Process Training, environmental manipulation), ideally in the context of a clinical trial – receive repeated practice of activities they are learning.

[a]Covers all of the UK except Scotland, which has a separate guideline
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- National Stroke Foundation. Clinical Guidelines for Stroke Management; 2010. Melbourne, Australia [12]
- Intercollegiate Stroke Working Party. National clinical guideline for stroke, 4th edition. London: Royal College of Physicians, 2012 [14]
- Scottish Intercollegiate Guidelines Network (SIGN). Management of patients with stroke: Rehabilitation, prevention and management of complications and discharge planning. A national clinical guideline. Edinburgh: SIGN, 2010 [15]
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As suggested in Table 16.4, there are two main methods used in memory rehabilitation: (1) approaches to help encode, store, and retrieve new information (e.g., deep [semantic] encoding of material); and (2) teaching compensatory techniques to reduce disabilities (e.g., diaries, electronic organizers, and audio alarms). The Cochrane review for memory impairments post-stroke [26] concluded that there was "no evidence to support or refute the effectiveness of memory rehabilitation on functional outcomes, and objective, subjective, and observer-rated memory measures." The more recent guidelines' conclusions regarding the effectiveness of memory rehabilitation note there are serious limitations in the evidence base. The Australian and UK (except Scotland) recommendations are the most detailed and are very similar. There is widespread agreement between Cochrane reviewers and guideline writers that research is needed to establish both the clinical effectiveness

Table 16.4 Recommendations from National Clinical Guidelines: Memory (extracts)

Scotland	There is not yet sufficient evidence to support or refute the benefits of cognitive rehabilitation for patients with problems of attention or memory.
Canada	The evidence for the effectiveness of specific interventions for cognitive impairment in stroke is limited and requires more research. • compensatory strategies can be used to improve memory outcomes
Australia	Any patient found to have memory impairment causing difficulties in rehabilitation or adaptive functioning should: • be referred for a more comprehensive assessment of their memory abilities • have their nursing and therapy sessions tailored to use techniques which capitalise on preserved memory abilities • be assessed to see if compensatory techniques to reduce their disabilities, such as notebooks, diaries, audiotapes, electronic organisers and audio alarms, are useful • be taught approaches aimed at directly improving their memory • have therapy delivered in an environment as like the patient's usual environment as possible to encourage generalisation.
UK[a]	A. Patients who complain of memory impairment and those clinically considered to have difficulty in learning and remembering should have their memory assessed using a standardised measure such as the Rivermead Behavioural Memory Test (RBMT). B. Any patient found to have memory impairment causing difficulties in rehabilitation or undertaking activities should: • be assessed medically to check that there is not another treatable cause or contributing factor (e.g. hypothyroidism) • have their profile of impaired and preserved memory abilities determined (as well as the impact of any other cognitive deficits on memory performance for example, attentional impairment) • have nursing and therapy sessions altered to capitalise on preserved abilities • be taught approaches that help them to encode, store and retrieve new information for example, spaced retrieval (increasing time intervals between review of information) or deep encoding of material (emphasizing semantic features) • be taught compensatory techniques to reduce their prospective memory problems, such as using notebooks, diaries, electronic organisers, pager systems and audio alarms • have therapy delivered in an environment that is as similar to the usual environment for that patient as possible.

[a]Covers all of the UK except Scotland, which has a separate guideline

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- Intercollegiate Stroke Working Party. National clinical guideline for stroke, 4th edition. London: Royal College of Physicians, 2012 [14]
- Scottish Intercollegiate Guidelines Network (SIGN). Management of patients with stroke: Rehabilitation, prevention and management of complications and discharge planning. A national clinical guideline. Edinburgh: SIGN, 2010 [15]
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(particularly at an activity rather than impairment level of outcome measurement) and the patient acceptability of different memory rehabilitation approaches, recruiting larger, more representative, groups of stroke patients (Table 16.4).

Neglect

Unilateral spatial neglect was originally classified as a perceptual impairment, before being widely accepted as an attentional disorder. It tends to stand alone these days perhaps because neglect is the most frequently researched topic within cognitive rehabilitation for stroke. The disabling effects of neglect have been well documented [45] (see Chap. 4). Although severe neglect is rather easily recognized, diagnosing milder neglect can be less obvious and only become apparent when observing higher-level activities such as driving, preparing a meal, and interacting in real-world social situations [46]. These difficulties obviously impact patient function and safety on transfer of care from hospital to community.

There is a relative wealth of research evidence in this field. Twelve randomized controlled trials were included in the Cochrane review of the cognitive rehabilitation of neglect [25]. A recent update of this review (in press) has included a further 11 trials [47–57]. Providing visual scanning training remains a popular intervention in neglect trials, as is the use of prisms. The latter is sometimes prescribed as an aid to be routinely worn on glasses but recent pilot trials have succeeded in determining the feasibility (but not yet the effectiveness) of prism adaptation training, a short therapist-led intervention using prisms during a specific computerized training activity [54].

The original review [25] concluded that cognitive rehabilitation can improve performance on impairment level tests but there is insufficient evidence to support or refute its effectiveness at reducing disability, one of the main aims of rehabilitation. This gap in the evidence base is due to limitations in the quality of the research studies, especially around the reduction of bias and the choice of appropriate outcome measures. The updated review will provide a systematic determination of whether the evidence base has been strengthened recently but for now the National guideline recommendations remain mostly at the consensus level and stress the need to invite people with neglect to participate in clinical trials (Table 16.5).

Aphasia

Aphasia (see Chap. 6) rehabilitation is a topic that has generated considerable research interest for decades and yet controversies regarding the quality of the evidence base remain. Clinical uncertainty persists around the most clinically and cost-effective method of supporting people with aphasia. Several major trials [55–58] and an update to the existing Cochrane review [28] that are likely to impact on National guideline were recently published. The new trials primarily concern impairment-focused intervention delivered at varying rates of intensity in the acute phase of the stroke pathway. Overall, the recent evidence does not support this

Table 16.5 Recommendations from National Clinical Guidelines: Neglect (extracts)

Canada	No specific recommendation beyond assessment
Scotland	Patients with visuospatial neglect should be assessed and taught compensatory strategies.
Australia	a) Any patient with suspected or actual neglect or impairment of spatial awareness should have a full assessment using validated assessment tools. b) Patients with unilateral neglect can be trialled with one or more of the following interventions: • simple cues to draw attention to the affected side • visual scanning training in addition to sensory stimulation • prism adaptation • eye patching • mental imagery training or structured feedback.
UK[a]	A. Any patient with a stroke affecting the right cerebral hemisphere should be considered at risk of reduced awareness on the left side and should be tested formally if this is suspected clinically. B. Due to the fluctuating presentation of neglect a standardised test battery such as the Behavioural Inattention Test should be used in preference to a single subtest, and the effect on functional tasks such as dressing and mobility should be determined. C. Any patient shown to have impaired attention to one side should be: – given a clear explanation of the impairment – taught compensatory strategies to help reduce impact on functional activities such as reading – given cues to draw attention to the affected side during therapy and nursing procedures – monitored to ensure that they do not eat too little through missing food on one side of the plate – offered interventions aimed at reducing the functional impact of the neglect (eg visual scanning training, limb activation, sensory stimulation, eye patching, prism wearing, prism adaptation training), ideally within the context of a clinical trial.

[a]Covers all of the UK except Scotland, which has a separate guideline

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- Intercollegiate Stroke Working Party. National clinical guideline for stroke, 4th edition. London: Royal College of Physicians, 2012 [14]
- Scottish Intercollegiate Guidelines Network (SIGN). Management of patients with stroke: Rehabilitation, prevention and management of complications and discharge planning. A national clinical guideline. Edinburgh: SIGN, 2010 [15]
- Lindsay MP, Gubitz G, Bayley M, Hill MD, Davies-Schinkel C, Singh S, and Phillips S. Canadian Best Practice Recommendations for Stroke Care (Update 2010). Prepared by the Canadian Stroke Strategy Best Practices and Standards Writing Group, on behalf of the Canadian Stroke Strategy (a joint initiative of the Canadian Stroke Network and the Heart and Stroke Foundation of Canada). 2010; Ottawa, Ontario, Canada: Canadian Stroke Network [13]

approach at this time point. A qualitative study of patients' perspectives, nested within our own trial, suggested service reorganization to provide a more psychosocial approach to early aphasia rehabilitation, perhaps shifting the cognitive neuropsychological model approach to later [59–63]. In addition to rehabilitation directed

at the language impairment, emerging evidence supports the effectiveness of structured behavioral interventions in reducing low mood in people with aphasia [64] right across the pathway.

This flurry of recent research interest in aphasia is welcome news for people with aphasia and their caregivers but makes it difficult to compare the latest recommendations from guidelines as several have yet to be updated (see Table 16.6). Interested readers are referred directly to the studies referenced previously and to the recent Cochrane review and UK (except Scotland) guideline. There remains a striking need for research into interventions for people with chronic aphasia and to supporting caregivers and other communication partners.

Table 16.6 Recommendations from National Clinical Guidelines: Aphasia (extracts)

UK[a]	A. All patients with communication problems following stroke should have an initial assessment by a speech and language therapist to diagnose the communication problem and to explain the nature and implications to the patient, family and multidisciplinary team. Routine reassessment of the impairment or diagnosis in the early stages of stroke (immediate and up to four months) should not be performed unless there is a specific purpose eg to assess mental capacity. B. In the early stages of stroke (immediate and up to four months) patients identified as having aphasia as the cause of the impairment should be given the opportunity to practise their language and communication skills as tolerated by the patient. C. Beyond the early stages of stroke (immediate and up to four months), patients with communications problems caused by aphasia should be reassessed to determine if they are more suitable for more intensive treatment with the aim of developing greater participation in social activities. This may include a range of approaches such as using an assistant or volunteer, family member or communication partner guided by the speech and language therapist, computer-based practice programmes and other functional methods. D. Patients with impaired communication should be considered for assistive technology and communication aids by an appropriately trained clinician. E. Patients with aphasia whose first language is not English should be offered assessment and communication practice in their preferred language. F. Education and training of health/social care staff, carers and relatives regarding the stroke patient's communication impairments should be provided by a speech and language therapist. Any education and training should enable communication partners to use appropriate communication strategies to optimise patient engagement and choice, and the delivery of other rehabilitation programmes. G. Any person with stroke at home who has continuing communication difficulty due to aphasia and whose social interactions are limited by it should be provided with information about any local or national groups for people with long-term aphasia, and referred to the group as appropriate.
Canada	Patients with aphasia should be taught supportive conversation techniques. Access to training for care providers in programs that facilitate communication with stroke survivors with aphasia.
Scotland	Aphasic stroke patients should be referred for speech and language therapy. Where the patient is sufficiently well and motivated, a minimum of two hours per week should be provided. Where appropriate, treatments for aphasia may require a minimum period of six months to be fully effective. Referral to the volunteer stroke service …. should be considered as an adjunct.

(continued)

Table 16.6 (continued)

Australia	a) All patients should be screened for communication deficits using a screening tool that is valid and reliable. b) Those patients with suspected communication difficulties should receive formal, comprehensive assessment by a specialist clinician. c) Where a patient is found to have aphasia, the clinician should: • document the provisional diagnosis • explain and discuss the nature of the impairment with the patient, family/carers and treating team, and discuss and teach strategies or techniques which may enhance communication • in collaboration with the patient and family/carer, identify goals for therapy and develop and initiate a tailored intervention plan. The goals and plans should be reassessed at appropriate intervals over time. d) All written information on health, aphasia, social and community supports (such as that available from the Australian Aphasia Association or local agencies) should be available in an aphasia-friendly format. e) Alternative means of communication (such as gesture, drawing, writing, use of augmentative and alternative communication devices) should be used as appropriate. f) Interventions should be individually tailored but can include: • treatment of aspects of language (including phonological and semantic deficits, sentence level processing, reading and writing) following models derived from cognitive neuropsychology • constraint-induced language therapy • the use of gesture • supported conversation techniques • delivery of therapy programs via computer. g) The routine use of piracetam is NOT recommended. h) Group therapy and conversation groups can be used for people with aphasia and should be available in the longer term for those with chronic and persisting aphasia. i) People with chronic and persisting aphasia should have their mood monitored. j) Environmental barriers facing people with aphasia should be addressed through training communication partners, raising awareness of and educating about aphasia in order to reduce negative attitudes, and promoting access and inclusion by providing aphasia-friendly formats or other environmental adaptations. People with aphasia from culturally and linguistically diverse backgrounds may need special attention, for example, from trained healthcare interpreters. k) The impact of aphasia on functional activities, participation and quality of life, including the impact upon relationships, vocation and leisure, should be assessed and addressed as appropriate from early post-onset and over time for those chronically affected.

[a]Covers all of the UK except Scotland, which has a separate guideline

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- National Stroke Foundation. Clinical Guidelines for Stroke Management; 2010. Melbourne, Australia [12]
- Intercollegiate Stroke Working Party. National clinical guideline for stroke, 4th edition. London: Royal College of Physicians, 2012 [14]
- Scottish Intercollegiate Guidelines Network (SIGN). Management of patients with stroke: Rehabilitation, prevention and management of complications and discharge planning. A national clinical guideline. Edinburgh: SIGN, 2010 [15]
- Lindsay MP, Gubitz G, Bayley M, Hill MD, Davies-Schinkel C, Singh S, and Phillips S. Canadian Best Practice Recommendations for Stroke Care (Update 2010). Prepared by the Canadian Stroke Strategy Best Practices and Standards Writing Group, on behalf of the Canadian Stroke Strategy (a joint initiative of the Canadian Stroke Network and the Heart and Stroke Foundation of Canada). 2010; Ottawa, Ontario, Canada: Canadian Stroke Network [13]

Other Cognitive Domains: Apraxia, Perception, Agnosia, and Executive Functions

As mentioned previously, not all the guidelines address each of these topics so, where available, they are simply listed in a single table (see Table 16.7). Cochrane reviews exist for apraxia [27] and perception [29] and one on executive function has been submitted for publication [30]. The apraxia review is now out of date but relevant rehabilitation trials published since that review are included in the recent UK (except Scotland) guideline (see the guideline's evidence tables). Generally these topics lack a clear evidence base (in the case of apraxia of speech [65] there are no trials at all) and implications for future research are discussed in the reviews. The Australian guideline selects the management of agnosia as a research priority, although they are alone in this (Table 16.7).

Table 16.7 Recommendations from National Clinical Guidelines: other cognitive domains

Apraxia: Australia	a) People with suspected difficulties executing tasks but who have adequate limb movement should be screened for apraxia and, if indicated, complete a comprehensive assessment. b) For people with confirmed apraxia, tailored interventions (e.g. strategy training) can be used to improve ADL.
Apraxia: UK[a]	A. Any person who has difficulties in executing tasks despite apparently adequate limb movement should be assessed formally for the presence of apraxia. B. Any person found to have apraxia should: – have their profile of impaired and preserved action abilities determined using a standardised approach (e.g. Test of Upper Limb Apraxia TULIA) – have the impairment and the impact on function explained to them, their family, and their treating team. – be given therapies and/or taught compensatory strategies specific to the deficits identified ideally in the context of a trial
Executive functions: Australia	a) Patients considered to have problems associated with executive functioning deficits should be formally assessed using reliable and valid tools that include measures of behavioural symptoms. b) External cues, such as a pager, can be used to initiate everyday activities in stroke survivors with impaired executive functioning. c) In stroke survivors with impaired executive functioning, the way in which information is provided should be considered.
Executive functions: UK[a]	A. Any person who appears to have adequate skills to perform complex activities but who fails to organise the tasks needed should be formally assessed for the dysexecutive syndrome, for example using the Behavioural Assessment of the Dysexecutive Syndrome (BADS). B. Any person with an executive disorder and activity limitation should be taught compensatory techniques. This may include internal strategies (eg self-awareness and goal setting) and/or external strategies (eg use of electronic organizers or pagers, or use of written checklists) ideally in the context of a clinical trial. C. When a patient's activities are affected by an executive disorder, the nature and effects of the impairment and ways of supporting and helping the patient should be discussed with others involved (eg family and staff).

(continued)

Table 16.7 (continued)

Agnosia: Australia	The presence of agnosia should be assessed by appropriately trained personnel and communicated to the stroke team.
Perception: UK[a]	A. Any person who appears to have perceptual difficulties should have a formal perceptual assessment (eg using the Visual Object and Space Perception battery (VOSP)) B. Any person found to have agnosia should: – have the impairment explained to them, their carers and their treating team – be offered a perceptual intervention, ideally within the context of a clinical trial

[a]Covers all of the UK except Scotland, which has a separate guideline

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- Lindsay MP, Gubitz G, Bayley M, Hill MD, Davies-Schinkel C, Singh S, and Phillips S. Canadian Best Practice Recommendations for Stroke Care (Update 2010). Prepared by the Canadian Stroke Strategy Best Practices and Standards Writing Group, on behalf of the Canadian Stroke Strategy (a joint initiative of the Canadian Stroke Network and the Heart and Stroke Foundation of Canada). 2010; Ottawa, Ontario, Canada: Canadian Stroke Network [13]

Models of Comprehensive Neuropsychological Rehabilitation

It is clinically intuitive that for maximum efficacy a program of cognitive rehabilitation must be delivered as part of a comprehensive neuropsychological approach and within a clear pathway specifying different levels of involvement by differently skilled professionals. Comprehensive programs are sometimes referred to, especially within the US traumatic brain injury rehabilitation literature, as "holistic" [22] although in Europe the term holistic usually relates to alternative medicine.

The inclusion of recommendations on a comprehensive neuropsychological approach is very new in national stroke guidelines, appearing for the first time in 2012 [14]. It is based on a biopsychosocial model of illness for the organization and delivery of psychological care after stroke. As stated in the preamble to the forthcoming UK (except Scotland) guideline:

> The comprehensive model was developed because domain-specific cognitive rehabilitation interventions (e.g. memory rehabilitation) tend not to address the complexity of life after stroke. The same limitation applies to interventions that focus on a specific mood disorder and this may lead to ineffective treatment (e.g., cognitive problems misdiagnosed as depression).

> Comprehensive-holistic rehabilitation programmes integrate evaluations of cognition, behaviour and mood to formulate the individual's difficulties. They then assist in the development of alternative or compensatory expectations and behaviours, leading towards independent self-management. They acknowledge that people with stroke may have limited awareness of their impairments or their impact (anosognosia), and that many therapies require motivation for engagement. [14]

The evidence base for comprehensive rehabilitation is mostly at the level of case series or cohort studies and largely focused on rehabilitation after acquired brain injury. There have also been two randomized controlled trials, the findings which support the integration of cognitive, interpersonal, and functional skil 67]. However, there is no unequivocal evidence that benefits are long-lasting (i.e., beyond the end of the treatment), which is a key requirement of an effective rehabilitation program. Interested readers are referred to two recent reviews of this topic [22, 23]. The UK (except Scotland) guideline is therefore largely at the level of consensus and based on extrapolation from promising research with younger, traumatically brain injured samples. The main recommendation concerns how multidisciplinary team (MDT) services are delivered, by whom and when, advocating a dynamic, rather than linear, stepped care approach, whereby patients move up and down the following steps of the model as required:

- *Step 1* comprises the routine assessments conducted within the MDT of all admitted patients, and the more detailed assessment of patients exhibiting symptoms of psychological disorder at any time after stroke.
- *Step 2* comprises the management of mild or moderate problems by MDT members who have been appropriately trained and where possible working under specialist supervision.
- *Step 3* comprises the management of more severe or persistent disorder, usually by a specialist.

The model in Fig. 16.2 illustrates the approach recommended by the NHS Stroke Improvement Program for England [31] and was developed from the stepped care model for adults with depression described by the National Institute for Health and Clinical Excellence (NICE) [68]. The latter defines stepped care as providing "a framework in which to organize the provision of services supporting patients, [caregivers] and healthcare professionals in identifying and accessing the most effective interventions." The NHS Improvement publication includes more details on operationalizing the stepped care model for people with stroke, including cognitive problems [31]. One of the core aspects of the model concerns skill mix and the employment of trained non-psychologists at certain steps of the model. This is a specific issue in the UK where difficulty accessing clinical psychologists has been a common and persisting finding from national audits [17, 69].

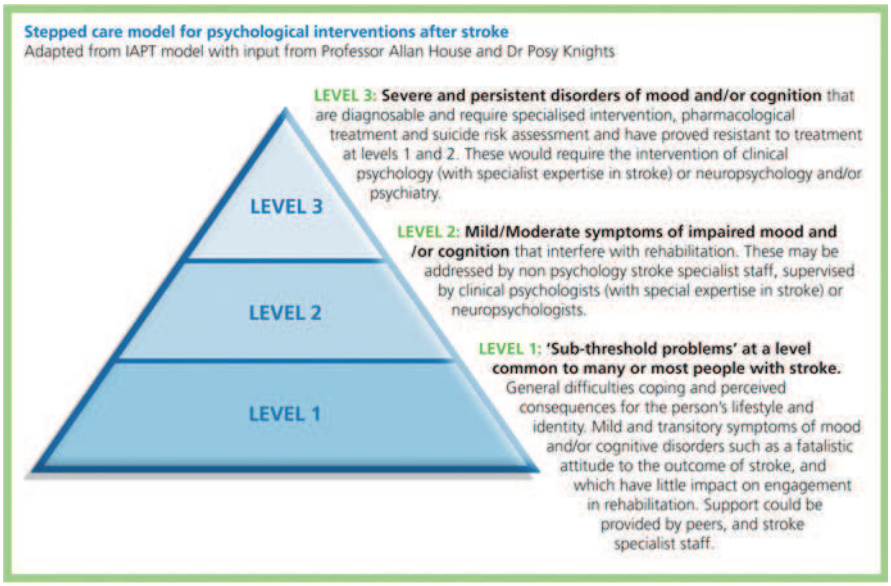

Fig. 16.2 Stepped care model for psychological interventions after stroke. Reprinted with permission from Gillham S, Clark L. Psychological care after stroke—improving stroke services for people with cognitive and mood disorders. NHS Improvement—Stroke, 2011. http://www.improvement.nhs.uk/stroke/Psychologicalcareafterstroke/tabid/177/Default.aspx

Summary

There is much to celebrate in the achievements of those working to develop an evidence-based approach for the rehabilitation of people with cognitive problems after stroke. Certain cognitive domains (e.g., neglect and aphasia) have attracted considerable research interest resulting in a range of interventions, many trials, and other levels of evidence. These feed into Cochrane systematic reviews and inform national clinical guidelines. These are exciting times with great potential for significant service improvement through emerging evidence for comprehensive neuropsychological rehabilitation approaches. In addition, practical recommendations for service delivery and organization are beginning to appear such as through recent modifications to the English stepped care model of psychological services.

On the other hand, even within heavily researched topics such as aphasia and neglect, there is still considerable uncertainty about which interventions to use, for which subgroup, when in the stroke pathway, and at what intensity. These are important questions. Furthermore, it is not clear why some topics (e.g., apraxia and memory) are, relatively speaking, under-researched and it certainly does not appear to be linked to either low prevalence or minimal impact on activity or social role

Implications for Research

Future studies should:

1. Provide a sufficiently detailed theoretical rationale for, and description of, the interventions including type and amount to allow implementation into clinical practice and research replication.
2. Provide a standard care control group, carefully documenting the content and amount of standard care, which can be highly variable.
3. Include detailed diagnostic information on individuals' perceptual problems given the heterogeneity in perceptual problems in terms of type, severity, and likely impact on everyday function.
4. Ensure low risk of study bias through rigorous methodological development and reporting, e.g., ensure allocation concealment, attempt to blind outcome assessors and report the success or failure, report all loss to follow-up, report results from all outcome measures, and control for other possible sources of bias.
5. Be of sufficient size to have adequate statistical power to answer clinically important questions about long-term functional outcomes.
6. Specify a primary endpoint and include analysis of other key outcomes such as adverse events, psychosocial benefits, and other outcomes deemed important by service users.
7. Adopt an intention-to-treat approach to measurement of outcomes in all individuals as well as to analysis of measured outcomes by treatment group.
8. Include a health economic assessment.

participation. Nor is it certain that simply producing "more of the same" research is the most productive way forward. As suggested in several of the Cochrane reviews of cognitive rehabilitation (see following for a recent example from the perception review [29]), future research could greatly improve clinical care through certain methodological and reporting changes:

Several countries now produce and audit against national clinical guidelines. In terms of cognitive rehabilitation there is reasonable consistency between the nations. Sometimes their differences are simply due to their publication date, with less evidence available to the older guidelines. The Scottish, Canadian, and Australian publications were in 2010, whereas the UK (excluding Scotland) guideline from the Royal College of Physicians London was updated for publication in 2012. Other differences result from the choice of either a wide or more focused breadth of topics and of course judgments about the standards set for accepting a piece of evidence, the criteria for which are described within each guideline.

Finally, the oft-repeated conclusion when examining the evidence is that we need more evidence! However there is also a need—and indeed it is already being

met—for a paradigm shift in how we think about rehabilitation for people with cognitive problems. We need to reach a balance between domain-specific research (essential for helping us understand specific impairments and mechanisms for recovery) and research into broad-based comprehensive approaches (that treat the person's cognitive deficits within the broader perspective of impact on everyday life and well-being). We must also engage in implementation research, so that the emerging evidence is translated into clinical practice.

Acknowledgements We are grateful to the authors of the national clinical guidelines (Australia, Canada, Scotland, RCP London) and the NHS Improvement—Stroke program for England and to Wiley for allowing us to reprint sections of their work. Our ideas are the result of collaborative working with the RCP London's Intercollegiate Stroke Working Party, and especially the members of the cognitive psychology subgroup: Janet Cockburn, David Gillespie, Andrew Bateman, and Peter Knapp whose input we wish to acknowledge.

Potential Conflict of Interest: Audrey Bowen is a member of the RCP London Intercollegiate Working Party for Stroke that produces the National Clinical Guideline referred to as UK (except Scotland), and author of some of the studies referred to in this chapter.

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Appendix 2 – example of communication aids used in qualitative study (study 1)

Type of problems

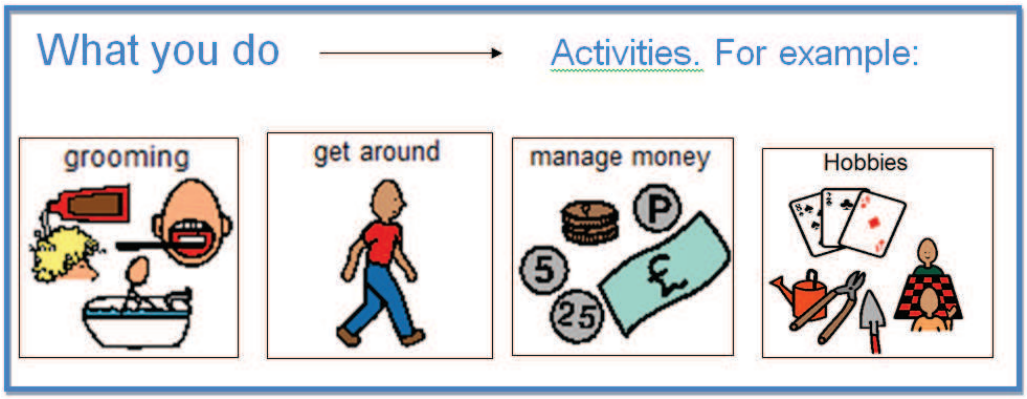
Memory

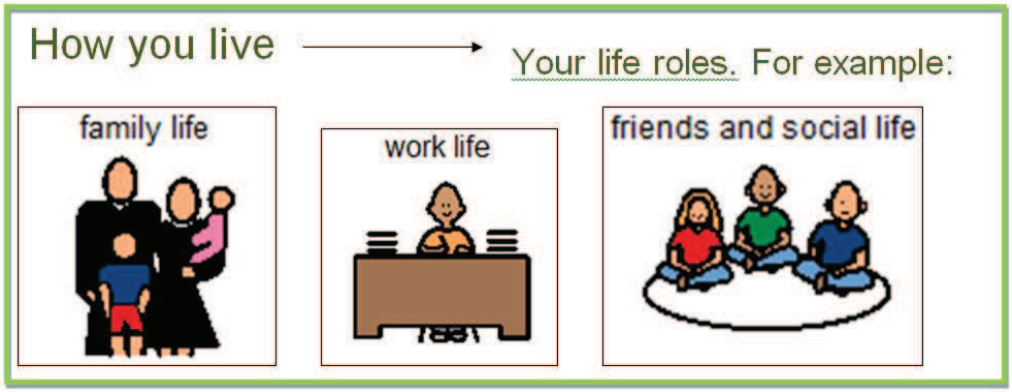
Ordering actions

Due to copyright issues, other cue cards developed for the 'Types of Problems' category' cannot be reproduced here. They were:

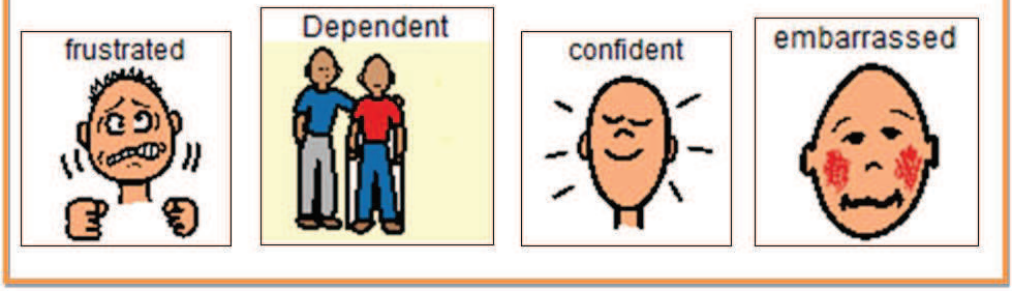
- concentration;
- communication;
- perception – making sense of what you see and hear;
- problem solving;
- noticing things on both sides of you.

Impact of these problems





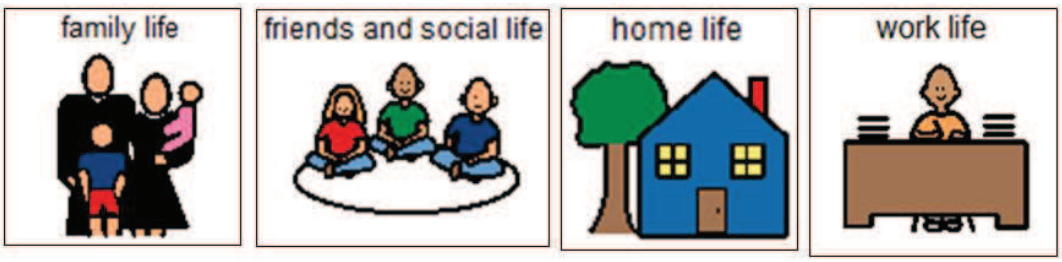
How you feel → About yourself? How others view you? Life? For example:



More of what you do

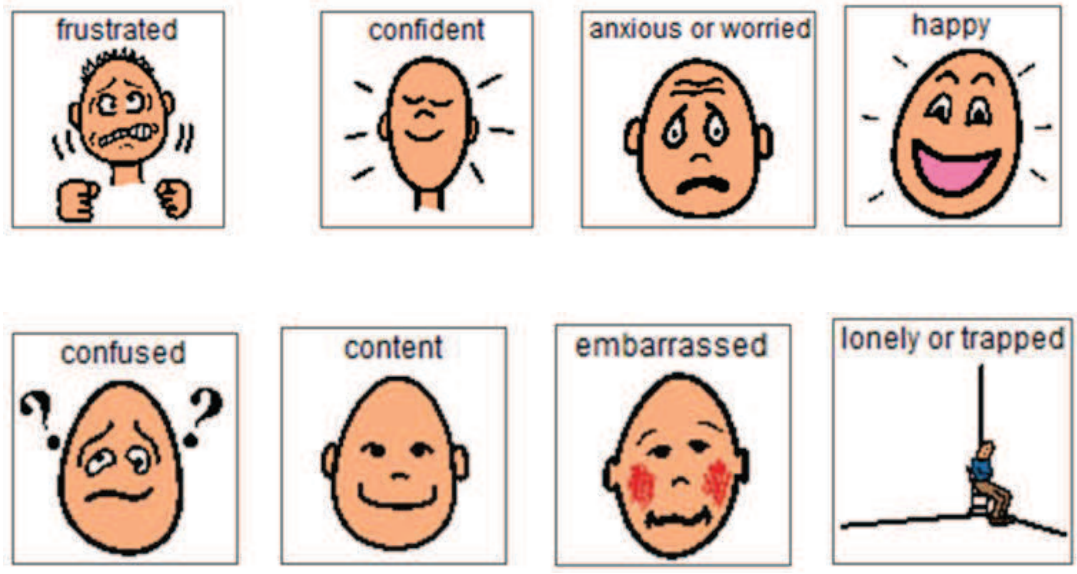
Examples only… more cue cards available

More of how you live



Examples only… more cue cards available

More of how you feel



Examples only… more cue cards available

Most of the aids from the 'impact of problems' onwards were developed through the Boardmaker programme.

Appendix 3. Search strategy used in Systematic Review (study 2)

#	Searches
1	(cogniti$ adj3 (process$ or disorder$ or defect or impair$ or problem$ or abilit$ or difficult$ or deficit$ or dysfunction or disturbance$ or fail$)).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
2	(Aphasi$ or dysphasi$ or anomia).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
3	((language or linguistic or speech or communicat$) adj2 (disorder$ or impair$ or problem$ or abilit$ or difficult$ or deficit$ or dysfunction$ or disurbance$)).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
4	((attention$ or processing or alert$ or distract$ or concentrat$) adj2 (disorder$ or impair$ or problem$ or abilit$ or difficult$ or deficit$ or dysfunction or disturbance$)).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
5	Attention Deficit Disorder with Hyperactivity/ or ADHD.mp.
6	attention deficit disorder.mp. or exp Attention Deficit Disorder/
7	4 not (5 or 6)
8	((memory or recall or remember$) adj2 (disorder$ or impair$ or problem$ or dysfunction or disturbance$)).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
9	((percept$ or visu?percept$ or visu$?spatial or visu$?construct$) adj2 (disorder$ or impair$ or problem$ or abilit$ or difficult$ or deficit$ or dysfunction or disturbance$)).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
10	exp Visual Attention/ or exp Sensory Neglect/ or exp Visual Perception/ or unilateral neglect.mp.
11	hemianopia.mp. or exp Hemianopia/
12	myopia.mp. or exp Myopia/
13	amblyopia.mp. or exp Amblyopia/
14	strabismus.mp. or exp Strabismus/

15	(9 or 10) not (11 or 12 or 13 or 14)
16	(aprax$ or dysprax$ or prax$ or practic).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
17	(executive dysfunction or dysexecutive syndrome or dysexecutive function).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
18	((executive function$ or initiat$ or awareness) adj2 (disorder$ or dysfunction or impair$ or difficult$ or problem$ or deficit$ or disturbance$ or disabilit$)).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
19	((goal management or goal selection or goal setting or goal directed behaviour or goal directed activit$) adj3 (disorder$ or dysfunction or impair$ or difficult$ or problem$ or deficit$ or disturbance$ or disabilit$)).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
20	((strategy?formation or planning or organi#ation or problem?solving or decision?making or sequenc$) adj2 (disorder$ or dysfunction or impair$ or difficult$ or problem$ or deficit$ or disturbance$ or disabilit$)).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
21	((functional activities or functional task$ or execut$ task$) adj3 (disorder$ or dysfunction or impair$ or difficult$ or problem$ or deficit$ or disturbance$ or disabilit$)).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
22	((activit$?of?daily?living or ADL) adj3 (disorder$ or dysfunction or impair$ or difficult$ or problem$ or deficit$ or disturbance$ or disabilit$)).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
23	1 or 2 or 3 or 7 or 8 or 15 or 16 or 17 or 18 or 19 or 20 or 21 or 22
24	quality of life.mp. or "Quality of Life"/
25	(QL or QoLor SF-36 or health index or health status).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
26	((disability or function$ or well being) adj2 (index or indices or instrument$ or measure$ or questionnaire$ or profile$ or scale$ or score$ or status or survey$ or test or assess$ or tool or checklist)).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
27	24 or 25 or 26

28	((patient or self$ or subjective) adj1 (rate$ or report$ or base$ or assess$ or evaluat$)).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
29	23 and 27 and 28
30	((disability or function$ or well being) adj5 (index or indices or instrument$ or measure$ or questionnaire$ or profile$ or scale$ or score$ or status or survey$ or test or assess$ or tool or checklist)).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
31	24 and 25 and 30
32	(acceptability or effect size$ or factor analys$ or factor loading$ or feasibility or item selection or interpretability or item response theory or latent trait theory or precision or psychometric$ or rasch or ROC or AUC or reproduc$ or reliabilit$ or replicab$ or repeatab$ or responsive$ or scaling or sensitivity or valid$ or weighting$).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
33	23 and 28 and 32
34	limit 33 to (human and english language and adulthood <18+ years> and "300 adulthood <age 18 yrs and older>" and human)

Appendix 4. Critical appraisal checklist for assessment in Systematic Review (Study 2)

Review Areas	Review Questions	
(A) Descriptive:		
Purpose what was the instrument designed to measure?		
Background. What was the rationale behind its design? (include settings)		
Description of tool N items & subscales, main domains, response format, method of administration, training, scoring, any normative data		
(B) Evaluative:	0 = not reported; + = some limited evident in favour; ++ = some good aspects in favour but some aspects do not meet criteria; +++ = good evidence in favour; - = evidence available does not support criteria.	
	Description	**Score**
Acceptability to respondents Any quantitative or qualitative data e.g. missing data, mode of administration, floor/ceiling		
Reliability Test-retest (reproducibility) correlations for summary scores ideally 0.7 Internal consistency (homogeneity e.g. alpha ≥0.7; item total correlations ≥0.2		
Validity Content (focus on **user-centredness**) Construct (correlation with other measures + known groups differences) Any other e.g. criterion (gold standard)		
Responsiveness Any evidence? Is 'significant change' defined (e.g. effect size, t-tests)? Does tool differentiate pts (e.g. floor ./ ceiling effects)?		
Feasibility Availability, time taken, scoring / interpretation, data good quality		

Pages 168 to 178 are removed from this version of my thesis due to copyright.

These pages contained:

- **Appendix 5a PRECiS questionnaire**
- **Appendix 5b - PRECiS Administrator guide**

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http://www.click2go.umip.com/i/coa/precis.html

You will be required to register to download; the process is free and very simple.