BMJ Open

Systematic review of patient-reported measures of treatment burden in stroke

Katie I Gallacher,[1] Terry Quinn,[2] Lisa Kidd,[3] David Eton,[4,5] Megan Dillon, Jennifer Elliot,[6] Natalie Johnston,[1] Patricia J Erwin,[7] Frances Mair[1]

ABSTRACT

Objectives Treatment burden is the workload of healthcare for people with long-term conditions (LTC) and its impact on well-being. A method of measurement is required to identify those experiencing high burden and to measure intervention efficacy. Our aim was to identify, examine and appraise validated patient-reported measures (PRMs) of treatment burden in stroke. Here, stroke serves as an exemplar LTC of older adults.

Design A systematic review of published studies that describe the development and validation of PRMs measuring treatment burden in stroke survivors.

Data sources We searched MEDLINE, Embase, CINAHL and PsycINFO electronic databases.

Eligibility criteria Studies published between January 2000 and 12 April 2019 inclusive, in English language. No restrictions were set based on clinical setting or geographical location.

Data extraction and synthesis Screening, data extraction and quality appraisal were conducted by two independent reviewers. Content of the PRMs was compared with a published taxonomy of treatment burden. Quality appraisal was conducted using International Society for Quality of Life Research standards.

Results From 3993 articles, 6 relevant PRMs were identified: 3 were stroke specific: The Satisfaction with Stroke Care questionnaire; The Stroke Patient-Reported Outcome Measure and The Barriers to Physical Activity after Stroke scale. Three were generic but validated in stroke: The WHO Quality of Life-100; The Patient's Questionnaire on Participation in Discharge Planning and The Chao Perception of Continuity scale. None comprehensively measured treatment burden. Examples of omitted burdens included developing coping strategies, managing finances and returning to driving. The most notable issue regarding quality appraisal was that three PRMs lacked any underpinning qualitative research relevant to the sample.

Conclusion There is a need to develop a comprehensive PRM of treatment burden for use in stroke, with potential for use in other older populations.

INTRODUCTION

Stroke is a common condition of older adults; in Europe, the incidence of stroke increases by a factor of 100 between the ages of 40 and 80 years.[1] Stroke treatments, particularly newer rehabilitative therapies, are often complex with multiple interacting components or

Strengths and limitations of this study

► The first systematic review to examine patient-reported measures of treatment burden in stroke, an important aspect of patient care.
► Thorough literature search of four major electronic databases and reporting as per Preferred Reporting Items for Systematic Reviews and Meta-Analysis guidelines.
► Exclusion of non-English language papers and publication pre-2000 (the latter due to the rapidly evolving nature of stroke management over recent decades).
► Treatment burden is a recently recognised concept influencing quality of care and quality of life, making literature searches challenging.

people involved in their delivery. Multidisciplinary therapy and early discharge from hospital are now recommended in guidelines, and these are becoming more commonplace in practice.[2,3] Engaging with and accessing such treatments, however, can demand considerable time and effort from stroke survivors, and this can be difficult for those who are older or frail.

Our previous qualitative research and systematic review demonstrated that people who have had a stroke can feel overwhelmed by managing their recovery and that this is exacerbated when health services are fragmented and lacking in a person-centred approach.[4,5] Treatment burden is defined as the workload of healthcare for people with long-term conditions and its impact on well-being.[5,6] It is becoming increasingly recognised by governing bodies and clinical guidelines as an important barometer of quality of care requiring attention.[7] Healthcare workload encompasses all tasks relating to health including those recommended by health professionals and those required to maintain or improve health status.[8] Through our previous research, we created a taxonomy of treatment burden in stroke and a conceptual model,[4,5] the latter is shown in figure 1. Our research showed that treatment burden
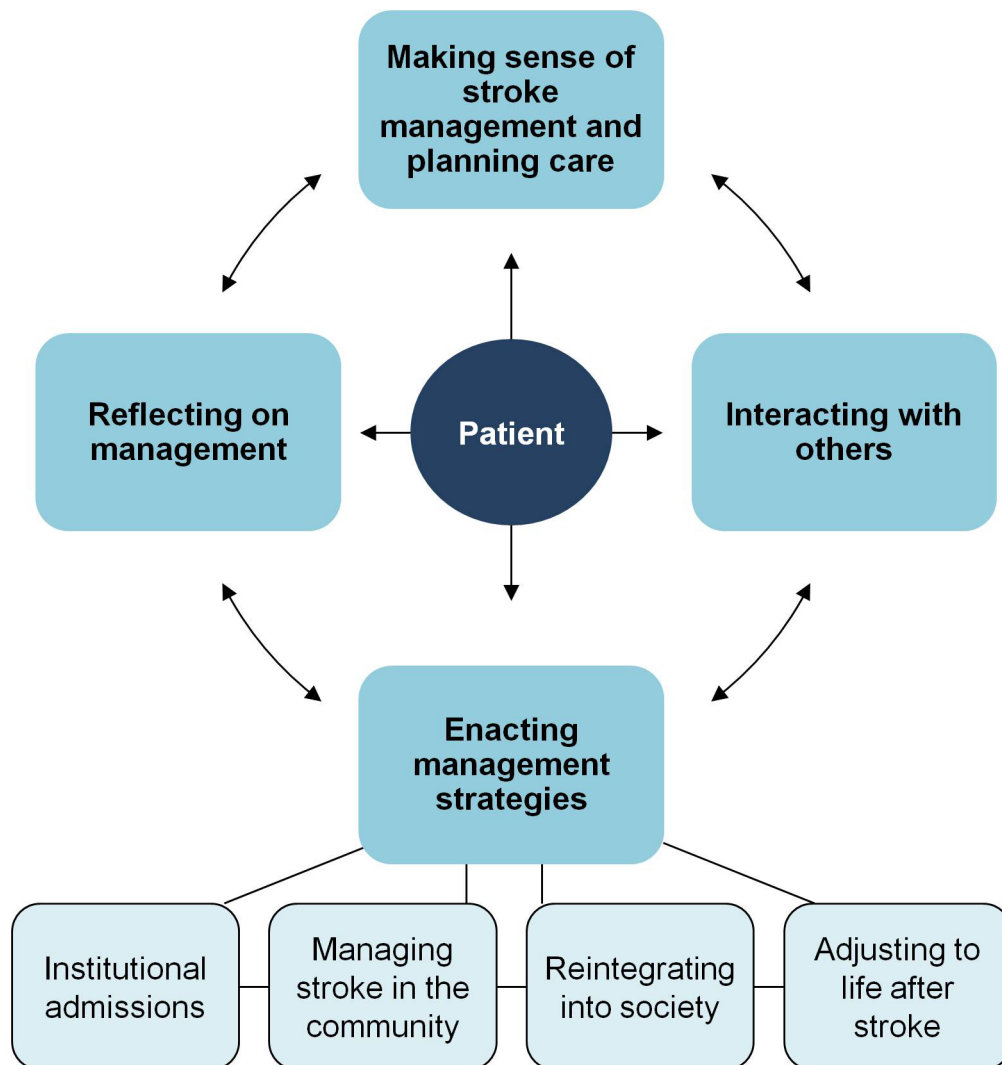
Figure 1 Conceptual model of stroke treatment burden. The arrows represent the possible pathways between components that stroke patients may follow. The 'enacting management strategies' component has four subcomponents. Reproduced with permission from *Plos MED*[4] (creative commons Attribution-Non-commercial 4.0 license).

is not purely dependent on volume of healthcare work; the way that services are planned, delivered and co-ordinated also influences the perceived burden felt by an individual.[5] For example, poor co-ordination or communication between healthcare providers can result in duplication or omission of important aspects of care. Capacity to manage healthcare workload will vary greatly between individuals due to personal abilities and circumstances; therefore, two people with the same treatment workload may cope very differently.[5 8]

Treatment burden is important because it may reduce quality of life and result in non-adherence to recommended treatments, resulting in suboptimal health-related outcomes and wasted health service resources.[9] For example, intentional and non-intentional non-adherence to medications may result because of an excessively complex or poorly planned medication regimen.[10] Consideration of treatment burden is particularly important in older people who may have a decreased ability to self-manage health due to physical, cognitive

and emotional difficulties. To date, treatment burden has been under-researched[4 11] and may not be adequately considered by clinicians involved in the provision of care for older people with long-term conditions.

Stroke can be used as an exemplar long-term condition for describing treatment burden. It is common in older adults,[1] often occurs in the context of other comorbidity[12] and can result in complex physical and cognitive impairments.[13] In previous work, we created a conceptual model of treatment burden in stroke through qualitative systematic review and analysis of interviews with stroke survivors.[4 5] This provided important insights into the lived experience of treatment burden in this group and highlighted the need for a method to quantify this burden. There are many potential applications of a tool that measures treatment burden. It could enable clinicians to identify those experiencing high levels of burden who may need additional support with self-management. It could facilitate inclusion of treatment burden as an outcome in clinical trials of stroke therapies alongside

measures of efficacy to ascertain if treatments are not only effective, but manageable in older, frailer patients in the 'real world'. Lastly, measurement of treatment burden would allow analysis of the effectiveness of interventions aimed at decreasing burden.

Treatment burden is experienced on an individual level; therefore, a patient-reported measure (PRM) is the best approach to measurement. A PRM is a report of the patient's health experience or experience with healthcare that comes directly from the patient, without interpretation of the response by anyone else.[14] It is a common and useful way to measure experiences or outcomes that require information directly from the patient, for example, health-related quality of life or satisfaction with health services. To help clinicians and researchers select which PRMs to use in a given setting, The International Society for Quality of Life Research (ISOQOL) has published a set of standards that can be used in the quality appraisal of PRMs.[14]

The aim of this systematic review is to collate and appraise published, validated PRMs of treatment burden in stroke, including discrete portions of PRMs that measure burden (eg, scales, scorable single items). We were particularly interested in content of the tool, the extent to which the PRMs aligned with our previously developed conceptual model of treatment burden in stroke[4 5] and the quality of the supporting research.[14]

METHODS

We followed Preferred Reporting Items for Systematic Reviews and Meta-Analysis (PRISMA)[15] guidance in the design, conduct and reporting of our review (online supplementary appendix 1). A protocol was developed and is available at https://www.crd.york.ac.uk/prospero/. All aspects of screening and data extraction were performed by two independent reviewers (KIG, MD, TJQ, LK, JE, NJ, DTE) with recourse to a third arbitrator as necessary (FSM).

Searching strategy

An initial scoping search was carried out to identify relevant terms and phrases, which would be used in the formal electronic search. This consisted of a preliminary search of personal files; MEDLINE via Ovid and the use of the 'related articles' function in PubMed. A formal search strategy was then created with an information scientist (PJE). We used a concept-based approach to design the search syntax, using validated search filters where available. The concepts were stroke, treatment burden and PRM. The full search strategy can be found in online supplementary appendix 2. As 'patient-reported measure' is not a recognised subject heading, 'patient-reported outcome measure' was used and in addition 'patient reported' was entered as a textword or subject heading or author keyword. Databases searched were as follows: MEDLINE (Ovid), Embase (Ovid), CINAHL (EBSCO) and PsycINFO (Ovid) from first January 2000 up to and including 12 April 2019. PRMs predating 2000 were deemed to be irrelevant to the current experiences of stroke survivors due to considerable changes in stroke management over recent decades.

Screening

Papers found were uploaded to a web-based systematic software programme DistillerSR (Evidence Partners) to facilitate screening and data extraction. Inclusion criteria are shown in online supplementary appendix 3.

Consistent with a prior systematic review of treatment burden measures,[11] potentially relevant scales were assessed at the level of individual items or domains. Items in each measure were scrutinised to ascertain if they were consistent with the definition of treatment burden outlined in the inclusion criteria (which is in line with the taxonomy of treatment burden and conceptual model created in our previous research).[4 5] If less than 50% of items in the PRM reflected treatment burden, then it was excluded. An exception to this was the presence of an independently scorable item or subscale that was deemed relevant, regardless of size. If a study had used a potentially relevant PRM but no information on development or validation was given in that paper, then the original development paper was sought and examined. If the original development paper was published pre-2000 but the PRM had been used in a published study after that date, then the PRM was included. References of included papers were scrutinised for relevant articles and the above process repeated until no new titles were found.

Data extraction and evidence synthesis

The data extraction and quality appraisal form is provided in online supplementary appendix 4. Data extracted were as follows: descriptive data about the study (eg, sample size and details of participant characteristics), items relating to treatment burden and information on PRM development or validation including psychometric testing. Content of the items on treatment burden was mapped to the taxonomy of treatment burden created in our previous work.[4 5] This was undertaken to ascertain which aspects of treatment burden were included in the PRMs and to scrutinise if any burdens were omitted. Quality appraisal was conducted using ISOQOL standards[14] as a reference. ISOQOL standards include whether the PRM was developed from underpinning qualitative research; reliability (how much it is free from measurement error); content validity (whether it measures what is relevant and important to the patient); construct validity (whether it measures what it purports to measure); responsiveness (whether it can detect changes over time); interpretability of scores (whether scores are meaningful to those using it) and patient and investigator burden (how difficult it is to use). We did not create a summative quality score and did not exclude papers due to perceived risk of bias; rather it served as a point of discussion.
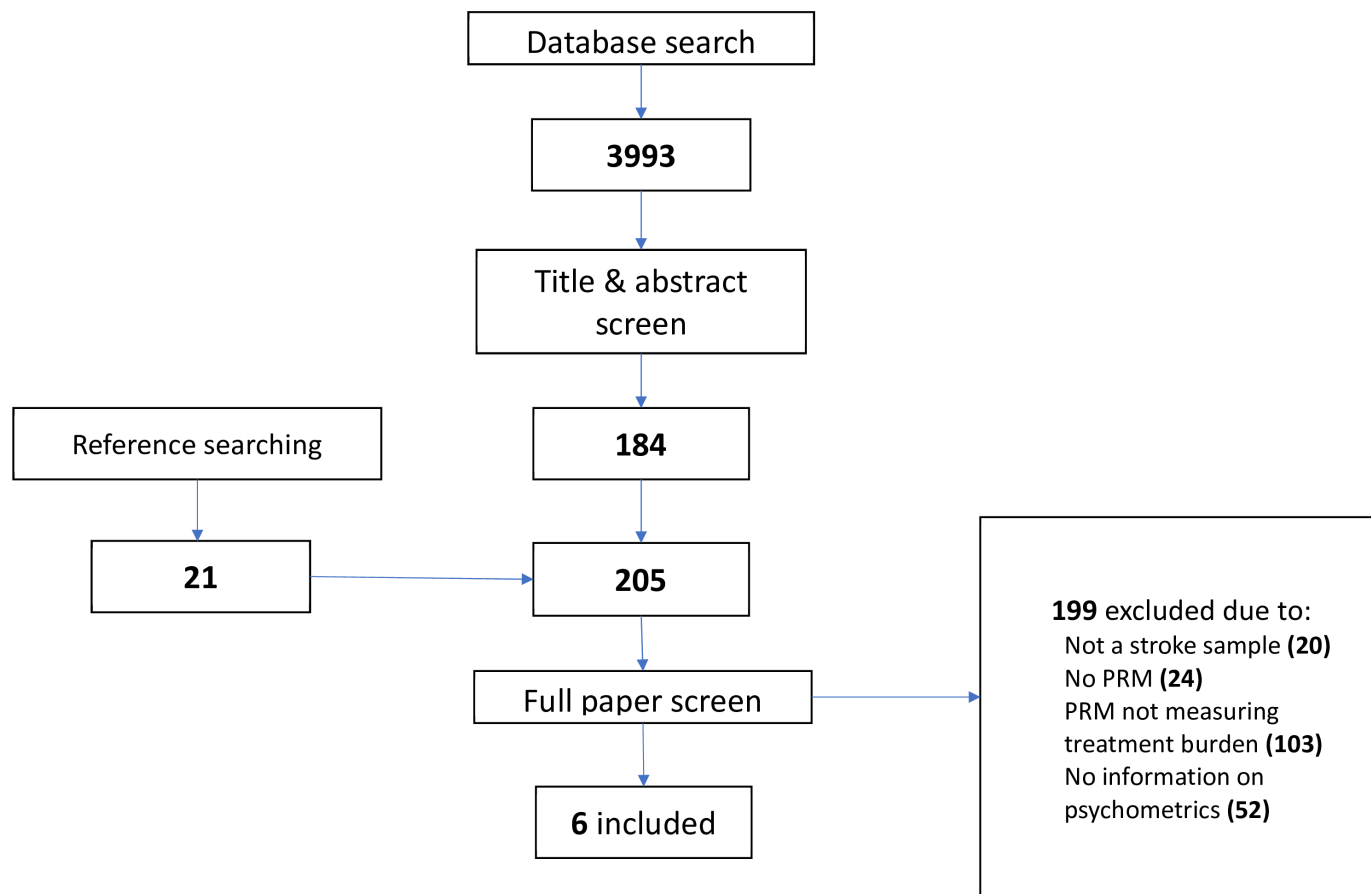
```
                    ┌─────────────────────┐
                    │   Database search   │
                    └─────────────────────┘
                               │
                               ▼
                    ┌─────────────────────┐
                    │       3993          │
                    └─────────────────────┘
                               │
                               ▼
                    ┌─────────────────────┐
                    │  Title & abstract   │
                    │      screen         │
                    └─────────────────────┘
                               │
                               ▼
┌─────────────────────┐   ┌─────────────────────┐
│ Reference searching │   │       184           │
└─────────────────────┘   └─────────────────────┘
           │                       │
           ▼                       ▼
┌─────────────────────┐   ┌─────────────────────┐
│        21           │──▶│       205           │
└─────────────────────┘   └─────────────────────┘
                               │
                               ▼
                    ┌─────────────────────┐        ┌──────────────────────────────┐
                    │ Full paper screen   │───────▶│ 199 excluded due to:         │
                    └─────────────────────┘        │ Not a stroke sample (20)     │
                               │                   │ No PRM (24)                  │
                               ▼                   │ PRM not measuring            │
                    ┌─────────────────────┐        │ treatment burden (103)       │
                    │     6 included      │        │ No information on            │
                    └─────────────────────┘        │ psychometrics (52)           │
                                                    └──────────────────────────────┘
```

Figure 2 Preferred Reporting Items for Systematic Reviews and Meta-Analysis (PRISMA) flow chart. PRM, patient-reported measure.

Patient and public involvement

The Research Advisory Group that guides this programme of research includes four stroke survivors or carers of stroke survivors. Their input has helped to guide the aims, objectives and methods of this study. Additionally, the results of this study were presented to three individuals with cardiovascular disease and discussed in a focus-group setting, which informed our discussion in the paper.

RESULTS
Screening
The PRISMA diagram of included studies is shown in figure 2. The database search yielded 3993 articles, of which 184 were retrieved for full-text review. The text of a further 21 papers, which were identified through reference list searching or were the original validation studies of PRMs identified within the full text papers, was also fully reviewed.

Identified PRMs
Six papers were identified that each described the development or testing of a relevant PRM[16–21] and were included in the review (table 1).

The Satisfaction with Stroke Care (SASC) questionnaire[16] was developed to measure stroke survivor's satisfaction with inpatient and outpatient health and social care services; 12 out of 13 items measure treatment burden. This PRM was originally developed and validated in a sample of stroke survivors. The Stroke Patient Reported Outcome Measure (Stroke-PROM)[17] was developed for use as an outcome measure in stroke drug trials and measures the effects of stroke and its treatments on an individual. Only 4 out of 46 items are relevant to treatment burden, but this was an independently scorable domain on satisfaction with treatments. This PRM was developed and validated in a sample of stroke survivors. The Barriers to Physical Activity after Stroke scale (BAPAS)[18] was developed to measure the perceived barriers to regular physical exercise after a stroke. It was developed and validated in stroke survivors, and 7 out of 14 items were deemed relevant to treatment burden. The other three measures were originally developed in non-stroke populations and subsequently validated in stroke survivors. The WHO Quality of Life-100 (WHOQOL-100)[19] is aimed at measuring quality of life and 4 out of its 100 items are relevant; they measure accessibility and quality of health and social care and constitute an independently scorable domain. The Patient's Questionnaire on Participation in Discharge Planning (P-QPD)[20] measures perceived patient involvement in the discharge planning process, all items are relevant to treatment burden, and all focus

Table 1 Included Patient Reported Measures

Name	Country of study	Purpose of PRM	Structure of PRM	Maximum score	Items relevant to treatment burden	Treatment burdens in PRM
SASC16	UK	Patient satisfaction with stroke services	2 domains (inpatient, outpatient), 13 items	39	12	Interactions with healthcare staff (kindness, personal care, communication); information provision about illness/services available after discharge; type, amount and adequacy of hospital treatments and therapies; preparation for return home; access to social and medical support in the community; adequacy of outpatient and ambulance services.
Stroke-PROM[17]	China	Effects of stroke on patients participating in drug trials	4 domains (physical, psychological, social, therapeutic), 46 items	230	4	Satisfaction with effects of treatments and services received.
BAPAS[18]	France	Patient perceived barriers to regular physical exercise after stroke	2 subscales (behavioural barriers, physical barriers), 14 items	70	7	Information provision; transport problems; lack of motivation; fear of another stroke; fear of falling; lack of finances; activity not suited to individual (patient not sporty).
WHOQOL-100[19]	Turkey	Quality of life	6 domains, 24 facets, 100 items	500	4	Accessibility and quality of health and social care.
P-QPD[20]	Sweden	Patient perceived involvement in discharge planning	3 subscales (information, medical treatment, goals and needs), 14 items	56	14	Information provision on illness/tests/examinations/treatments/medication/rehabilitation; ability to ask questions; ability to participate in discussions about treatments/goals/social support/rehabilitation needs after discharge; participation in working out discharge plan.
Chao-PC[21]	UK	Patient perceived continuity of care	2 domains, 23 items	115	17	Doctor's knowledge of medical history and family; location of medical care; continuity of doctor; fragmentation of care; relationship with doctor; communication with doctor; access to other specialist; emergency care; trust in doctor.

BAPAS, Barriers to Physical Activity after Stroke scale; PRM, patient-reported measure; WHOQOL-100, WHO Quality of Life-100.

on the discharge process. The Chao Perception of Continuity scale (Chao-PC)[21] has 23 items; 17 of which measure treatment burden, all items focus on continuity of care.

Details of the participants included in each study are given in online supplementary appendix 5. Five of the studies reported a mean age over 60 years,[16 18–21] the other stated that 48% were between 45 and 65 years and that 40% were 65 years or over.[17] Two studies were conducted in the UK (SASC and Chao-PC),[16 21] both were community based with a majority of white participants. The other studies were conducted in France (BAPAS),[18] China (Stroke-PROM),[17] Sweden (P-QPD)[20] and Turkey[19] (WHOQOL-100). All studies included a balanced mix of men and women. Level of participant education varied

between studies, the Chinese sample had the highest[17] and one of the UK samples had the lowest.[21]

Content of the PRMS
Table 2 shows the aspects of treatment burden included in and missing from the PRMs found. All aspects of treatment burden found within the included PRMs fell inside our previously published conceptual framework and taxonomy.

Quality appraisal
A detailed account of quality appraisal is given in table 3. The three PRMs that were developed in stroke populations (SASC, Stroke-PROM and BAPAS)[16–18] were

Table 2 Our taxonomy of treatment burden in stroke		
Type of treatment burden	**Healthcare workload**	**Care deficiency**
Making sense of stroke management and planning care	Understanding symptoms, investigations, treatments, risk factors. Information gathering. *Taking responsibility for health management.* Goal setting and prioritising. *Problem solving.* *Managing uncertainty.* Maintaining motivation. *Developing coping strategies.* Coping with negative emotions.	Lack of information provision and poor signposting. Information hard to understand. Poorly timed information. Not enough verbal information. Information not tailored to individual. Lack of support with care planning.
Interacting with others	Seeking advice or help from health and social care professionals. *Gaining emotional and practical support from friends, family, fellow patients.* *Strained relationships due to treatments.* *Protecting carers from stress.* *Dealing with stigma, for example, of walking aids.*	*Misdiagnosis.* Paternalism from HPs. Lack of empathy from HPs. Mismatch in ideas between patient and HP. *Poor access to a GP.* Poorly co-ordinated care. Poor continuity. *Poor communication from GP.*
Enacting management strategies	Undergoing acute care. Inpatient rehabilitation. Discharge process. Community rehabilitation. Attending outpatient appointments/therapies. Taking medications. Risk factor modification. *Managing comorbidities.* Adaptations to home. Organising and receiving home care. *Return to driving and employment.* Using mobility aids. *Managing finances.* Paying for treatments. *Enacting coping strategies.* *Using alternative therapies.*	*Waiting times for inpatient tests.* Unpleasant ward. Poorly supported discharge. *Poor GP follow-up.* Lack of help with transport to appointments. Complicated medication regimens. Poor access to home adaptations and walking aids. Substandard home care. *Poor access to driving assessment.* *Complicated benefits system.* *Lack of psychological support and support groups.*
Reflecting on management	Attending review appointments. Joint healthcare decisions with HPs. *Reflecting on progress.* *Deciding on adherence to HP advice.* *Keeping up to date with new treatments available.* Managing worry about another stroke.	*Poor short-term follow-up for milder cases.* *Poor long-term follow-up for all.*

Aspects of treatment burden not included in any of the PRMs found are shown in italics.
GP, general practitioner; HP, health professional; PRM, patient-reported measure.

Table 3 Quality appraisal of included papers using ISOQOL standards as a reference

Patient-reported measure	Qualitative work relevant to sample	Reliability	Content validity	Construct validity	Responsiveness	Interpretability	Feasibility
SASC	Patient/health professional interviews; literature search.	Internal consistency: Cronbachs alpha=0.86 for hospital satisfaction and 0.77 for home satisfaction. Test-retest: weighted kappa=good reliability for 11 questions. Mean difference on test-retest=0.59 (SD=2.40) hospital satisfaction; 0.32 (SD=2.1) home satisfaction.	By post—28 then 23 participants.	Principle components analysis revealed two factors. High correlation between hospital satisfaction and Faces Scale satisfaction measure (r=0.67; p<0.00005). No strong correlations found between hospital satisfaction and measures of function or quality of life. Weak negative correlation found with the Geriatric Depression score (r=−0.26; p=−0.0015). No strong correlations found between home satisfaction and other measures apart from a Nottingham extended ADL scale (r=0.30; p=0.00098).	Not tested.	High score=greater satisfaction Score if answering 'satisfied' to all questions: hospital satisfaction=16/24, home satisfaction=10/15	Response rate to postal questionnaire 87%.
Stroke-PROM	Patient/health professional interviews; literature search.	Internal consistency: Cronbach's alpha=0.905 for the total score and for the four domains it ranged from 0.861 to 0.908. No test-retest.	By referring to literature, consulting questionnaires, interviewing patients and consulting with patients, physician experts and one psychometric expert. Confirmed using the CVI.	Confirmatory factor analysis: index of fit met the standard requirements. Discriminant validity: mRS assessed disability and scale could differentiate between healthy controls and stroke patients with different degrees of disability.	Not tested.	Higher score=more positive responses.	Response rate, completion rate were over 97%. Time to completion=8.9 min.
BAPAS	Patient interviews and health professional expert panel.	Internal consistency: Cronbachs alpha=0.86. Test-retest: Intraclass correlation coefficient model 2,1=0.91 (95% CI 0.79 to 0.97).	Panel of experts in the field and 10 patients.	Principal component analysis with number of factors fixed at 8—showed original structure (BAPAS-27) was replicated in the final BAPAS scale. The eight factors explained 84% of total variance of the BAPAS scale. Also assessed the proper construct of the BAPAS scale—two factors were obtained that explored physical dimensions and two that explore behavioural. A two-part scale was constructed (physical and behavioural). Criterion validity tested using correlation with mRS score: r=0.65 (p<0.001).	Not tested.	Higher score=more barriers.	Time to complete if naive=4 min.
WHOQOL-100	Expert review and focus groups but not stroke survivors specifically (results not given).	Internal consistency: Cronbach's alpha for relevant domain (environment)=0.92. Test-retest not done in stroke survivors and not given.	Yes but not in stroke survivors and results not given.	Convergent validity: correlations found between WHOQOL-100 and SF36. Fair to good for relevant domains.	Not in stroke patients and results not given.	Higher=better QOL.	Long—100 items.
P-QPD	Unclear.	Internal consistency: Cronbach's alpha=information 0.82; goals needs 0.87; medical treatment 0.66. No test-retest.	Face validity established with 'patients and experts'.	Factor analysis: three factors extracted. Comparisons of scores across known groups:subscale differences found on age, length of hospital stay, ADL (independent vs dependent). No differences based on gender, education, living arrangement or prior experience of stroke.	Not tested.	Higher=greater participation.	Not discussed.
Chao-PC	Unclear.	Internal consistency: Cronbach's alpha ranged from 0.7 to 0.76 for interpersonal trust, interpersonal knowledge and provider consistent care. No test-retest.	Face-to-face delivery of questionnaire for 110 participants.	Exploratory factor analysis: three factors supported (interpersonal trust, interpersonal knowledge, provider consistent care). Known-groups validity comparing distress and disability groups—no significant differences in scores identified.	Not tested	Higher=better continuity.	Low response rate in postal questionnaire. Deemed not easily transferable to a UK setting without further modification.

ADL, activites of daily living; BAPAS, Barriers to Physical Activity after Stroke scale; CVI, content validity index; ISOQOL, International Society for Quality of Life Research; mRS, modified Rankin score; WHOQOL-100, WHO Quality of Life-100.

developed from qualitative work relevant to the sample, that is, in stroke survivors. The WHOQOL-100 had been originally developed from qualitative work undertaken in non-stroke populations and did not include any qualitative research examining stroke survivors' experiences. It was unclear if the Chao-PC or the P-QPD had been developed from any underpinning qualitative work with stroke survivors; however, development of the Chao-PC was underpinned by Banahan's conceptual model of continuity of care.[22 23]

Regarding reliability, all PRMs had been tested for internal consistency and all were found to be suitably reliable (Cronbach's alpha >0.70). Only two studies provided information on test-retest reliability (SASC and BAPAS).[16 18] The SASC study assessed this by weighted kappa in a sample of 21 patients who repeated the PRM 2 weeks after the original mailing.[16] Eleven out of the 13 items had a weighted kappa ≥0.3 and the authors reported that cut-off as acceptable. The BAPAS study repeated the measure in 21 participants after 4–6 days and found good reproducibility with an intraclass coefficient of 0.9.[18] However, authors note the short interval between test and retest, therefore the possibility of recollection bias.

Content validity had been assessed in all studies, although in the case of the WHOQOL-100[19] this was not with stroke survivors and the SASC[16] had not been tested face-to-face. All studies had assessed construct validity using various methods including factor analysis, correlations with other PRMs and known-group validity. Results are detailed in table 3.

None of the studies had tested for responsiveness to change, but this was deemed appropriate as none were measuring outcomes longitudinally.

Regarding interpretability, all papers described the meaning of high and low scores but only one described a cut-off (SASC).[16]

Three studies (SASC, Stroke-PROM and Chao-PC)[16 17 21] reported that they assessed participant burden by analysing response rates; the two with high response rates were deemed to have low burden (SASC, Stroke-PROM)[16 17] and the other with a low response rate (Chao-PC) was deemed not easily transferable to a UK population without further modification due to the structure and content of its items.[21] Two studies reported time to complete the measure (Stroke-PROM and BAPAS).[17 18] None assessed investigator burden, that is, ease of use for the researcher using the PRM.

DISCUSSION
PRMs found
We found six PRMs that measure treatment burden in stroke. All had been tested in older stroke survivors in a mix of hospital and community settings within developed countries. None of the PRMs found had been developed to comprehensively evaluate treatment burden and none of the included studies were aimed at doing so. Rather, they were aimed at assessing related concepts or narrow aspects of treatment burden. One PRM, which was developed before treatment burden had been conceptualised in the medical literature, was aimed at measuring patient satisfaction (SASC),[16] and this covered the most aspects of treatment burden out of the PRMs found, however, many treatment burdens were missing. Three PRMs focused on important but limited aspects of treatment burden: barriers to participation in physical activity (BAPAS),[18] continuity of care (Chao-PC)[21] and participation in discharge planning (P-QPD).[20] The other two (WHOQOL-100 and Stroke-PROM)[17 19] are longer measures that included a small minority of items that were relevant to the issue of treatment burden and independently scorable. In summary, none of the published PRMs comprehensively measured treatment burden in stroke.

During quality appraisal, the most notable weakness was that the three studies that involved validation of PRMs originally developed in non-stroke populations (Chao-PC, P-QPD, WHOQOL-100)[19–21] did not describe any qualitative work underpinning their use in stroke survivors. Additionally, the lack of assessment of content validity or consideration of feasibility of the WHOQOL-100 results in uncertainty about whether this long 100-item measure is suitable in a stroke population who are typically older and potentially cognitively impaired, frail or easily fatigued. Only one PRM (SASC)[16] provided a cut-off score, this was for 'satisfactory treatment'. None of the studies were longitudinal therefore none assessed responsiveness; however, testing of this would be required if longitudinal measurement was an intended future use of the PRM, such as in a clinical trial. One measure (Chao-PC)[21] was deemed by the authors as unsuitable for use in a UK population due to the structure and content of the items. For example, items did not distinguish between primary and secondary care and so could be confusing to a UK patient.

Strengths and limitations
Our search was limited to English-language papers, which could be viewed as a limitation, although there is increasing evidence that this may have little effect.[24] Exclusion of papers published pre-2000 could also be viewed as a limitation; however, this was chosen due to the rapidly evolving nature of stroke management over recent decades. One paper published before 2000 was included because the PRM identified had been used in subsequent studies after that date.[25] Searching for papers that examine treatment burden is challenging because it is a relatively new concept that is multifaceted. To combat this, we clearly defined treatment burden prior to the start of our review based on our previous qualitative work.[4 5]

How results fit in with current literature and future research
Treatment burden is a relatively new concept in the medical literature, with robust qualitative work giving us a better understanding of the patient experience of this phenomenon in stroke and other patient groups.[4–6 26 27]

Aspects of care that stroke survivors describe as particularly burdensome include information provision about stroke treatments, care co-ordination and the process of transitioning from hospital into the community.[4 5] These are examples of areas that would benefit from measurement and intervention to lessen treatment burden. Despite this, we still need to understand more about the relationship between treatment burden and health-related outcomes; how burden changes over the patient journey and whether we can lessen treatment burden through altering the way that healthcare is provided. To examine these areas, quantification of treatment burden is required, yet this is not straightforward. Treatment burden is more than simply healthcare workload, it is a complex interplay of healthcare systems, individuals and their social networks that results in a feeling of encumbrance if demand outweighs personal resources.[5 8] This has the potential to lead to disengagement from health services, wasted resources and worsening health outcomes, particularly in vulnerable groups such as those who are older, frail, socioeconomically deprived or socially isolated.[28]

Treatment burden has received attention from researchers interested in individual conditions[29–32] and from those interested in studying people with multiple long-term conditions.[6] PRMs have recently been developed for use in the latter population.[33–35] There is overlap between these generic treatment burden PRMs and our taxonomy of treatment burden in stroke; however, many stroke-specific burdens are not represented in the generic measures, such as robotic upper limb neurorehabilitation, speech and language therapy, management of visual problems and vocational rehabilitation. There is good evidence that stroke survivors obtain better health-related outcomes in treatment pathways designed specifically for stroke survivors,[36] and therefore, some of their healthcare experiences are likely to be different. In this systematic review, we chose to exclude PRMs that have not been validated in stroke survivors because current guidance for PRM selection indicates that it is desirable that chosen PRMs be validated in a sample relevant to the population in question.[14] This means that PRMs not developed for use in stroke specifically may not fully represent all treatment burdens encountered by stroke survivors. Additionally, stroke survivors are typically older individuals who may have cognitive impairment, visual difficulties or aphasia that can make completion of a PRM challenging. It is vital that when PRMs are developed for use in older populations that careful attention is paid to usability in this group.

In conclusion, we found no comprehensive PRMs of treatment burden that had been validated in a stroke population. Further research to develop and validate a new PRM of treatment burden in stroke would be important to enable new insights into the quality of care and quality of life of stroke survivors. Such a tool could also be of value for use in other older populations with similar healthcare challenges.

Author affiliations
[1]General Practice and Primary Care, Institute of Health and Wellbeing, University of Glasgow, Glasgow, UK
[2]Department of Academic Geriatric Medicine, Institute of Cardiovascular and Medical Sciences, University of Glasgow, Glasgow, UK
[3]Nursing & Healthcare School, School of Medicine, Dentistry and Nursing, University of Glasgow, Glasgow, UK
[4]Department of Health Sciences Research, Mayo Clinic, Rochester, Minnesota, USA
[5]Robert D. and Patricia E. Kern Center for the Science of Health Care Delivery, Mayo Clinic, Rochester, Minnesota, USA
[6]Stroke and Brain Imaging, Institute of Neuroscience and Psychology, University of Glasgow, Glasgow, UK
[7]Mayo Medical Libraries, Mayo Clinic College of Medicine, Rochester, Minnesota, USA

REFERENCES

1. Béjot Y, Bailly H, Durier J, *et al*. Epidemiology of stroke in Europe and trends for the 21st century. *Presse Med* 2016;45:e391–8.
2. Royal College of Physicians. Stroke guidelines, 2016. Available: https://www.rcplondon.ac.uk/guidelines-policy/stroke-guidelines [Accessed 1 July 2019].
3. Winstein CJ, Stein J, Arena R, *et al*. Guidelines for adult stroke rehabilitation and recovery a guideline for healthcare professionals from the American heart Association/American stroke association. *Stroke* 2016;47:E98–169.
4. Gallacher K, Morrison D, Jani B, *et al*. Uncovering treatment burden as a key concept for stroke care: a systematic review of qualitative research. *PLoS Med* 2013;10:e1001473.
5. Gallacher KI, May CR, Langhorne P, *et al*. A conceptual model of treatment burden and patient capacity in stroke. *BMC Fam Pract* 2018;19.
6. Eton D, Ridgeway J, Egginton J, *et al*. Finalizing a measurement framework for the burden of treatment in complex patients with chronic conditions. *Patient Relat Outcome Meas* 2015;6:117–26.
7. NICE. Multimorbidity: clinical assessment and management, 2016. Available: https://www.nice.org.uk/guidance/ng56 [Accessed 1 July 2019].
8. May CR, Eton DT, Boehmer K, *et al*. Rethinking the patient: using burden of treatment theory to understand the changing dynamics of illness. *BMC Health Serv Res* 2014;14:281.
9. May C, Montori VM, Mair FS. We need minimally disruptive medicine. *BMJ* 2009;339:b2803.
10. Chambers JA, O'Carrol RE, Hamilton B, *et al*. Adherence to medication in stroke survivors: a qualitative comparison of low and high adherers. *Br J Health Psychol* 2010:592–609.

11. Eton D, Elraiyah TA, Yost K, *et al*. A systematic review of patient-reported measures of burden of treatment in three chronic diseases. *Patient Relat Outcome Meas* 2013;4:7–20.

12. Gallacher KI, Batty GD, McLean G, *et al*. Stroke, multimorbidity and polypharmacy in a nationally representative sample of 1,424,378 patients in Scotland: implications for treatment burden. *BMC Med* 2014;12:151.

13. Stroke Association. State of the nation - Stroke statistics, 2018. Available: https://www.stroke.org.uk/resources/state-nation-stroke-statistics [Accessed 1 July 2019].

14. Reeve BB, Wyrwich KW, Wu AW, *et al*. ISOQOL recommends minimum standards for patient-reported outcome measures used in patient-centered outcomes and comparative effectiveness research. *Qual Life Res* 2013;22:1889–905.

15. PRISMA. Transparent reporting of systematic reviews and meta-analysis, 2009. Available: http://www.prisma-statement.org/ [Accessed 1 July 2019].

16. Pound P, Gompertz P, Ebrahim S. Patients' satisfaction with stroke services. *Clin Rehabil* 1994;8:7–17.

17. Luo Y, Yang J, Zhang Y. Development and validation of a patient-reported outcome measure for stroke patients. *Health Qual Life Outcomes* 2015;13.

18. Drigny J, Joussain C, Gremeaux V, *et al*. Development and validation of a questionnaire to assess barriers to physical activity after stroke: the barriers to physical activity after stroke scale. *Arch Phys Med Rehabil* 2019. doi:10.1016/j.apmr.2018.12.034. [Epub ahead of print: 24 Jan 2019].

19. Unalan D, Soyuer F, Ozturk A, *et al*. Comparison of SF-36 and WHOQOL-100 in patients with stroke. *Neurol India* 2008;56:426–32.

20. Almborg A-H, Ulander K, Thulin A, *et al*. Patients' perceptions of their participation in discharge planning after acute stroke. *J Clin Nurs* 2009;18:199–209.

21. Hill KM, Twiddy M, Hewison J, *et al*. Measuring patient-perceived continuity of care for patients with long-term conditions in primary care. *BMC Fam Pract* 2014;15:191.

22. Chao J. Continuity of care: incorporating patient perceptions. *Fam Med* 1988;20:333–7.

23. Banahan BF, Banahan BF. Continuity as an attitudinal contract. *J Fam Pract* 1981;12:767–8.

24. Morrison A, Polisena J, Husereau D, *et al*. The effect of English-language restriction on systematic review-based meta-analyses: a systematic review of empirical studies. *Int J Technol Assess Health Care* 2012;28:138–44.

25. Sulch D, Melbourn A, Perez I, *et al*. Integrated care pathways and quality of life on a stroke rehabilitation unit. *Stroke* 2002;33:1600–4.

26. Sav A, Kendall E, McMillan SS, *et al*. 'You say treatment, I say hard work': treatment burden among people with chronic illness and their carers in Australia. *Health Soc Care Community* 2013.

27. Gallacher K, May C, Montori VM, *et al*. Understanding treatment burden in chronic heart failure patients. A qualitative study. *Ann Fam Med* 2011;9:235–43.

28. Mair FS, May CR. Thinking about the burden of treatment. *BMJ* 2014;349:g6680.

29. Kahn LS, Vest BM, Madurai N, *et al*. Chronic kidney disease (CKD) treatment burden among low-income primary care patients. *Chronic Illn* 2015;11:171–83.

30. Eton DT, Anderson RT, Cohn WF, *et al*. Risk factors for poor health-related quality of life in cancer survivors with multiple chronic conditions: exploring the role of treatment burden as a mediator]]&gt. *Patient Relat Outcome Meas* 2019;10:89–99.

31. Lorenz EC, Egginton JS, Stegall MD, *et al*. Patient experience after kidney transplant: a conceptual framework of treatment burden. *J Patient Rep Outcomes* 2019;3.

32. Rogers E, Yost K, Rosedahl J, *et al*. Validating the patient experience with treatment and self-management (pets), a patient-reported measure of treatment burden, in people with diabetes. *Patient Relat Outcome Meas* 2017;8:143–56.

33. Eton DT, Yost KJ, Lai JS, *et al*. Development and validation of the patient experience with treatment and self-management (pets): a patient-reported measure of treatment burden. *Qual Life Res* 2017.

34. Tran V-T, Harrington M, Montori VM, *et al*. Adaptation and validation of the treatment burden questionnaire (TBQ) in English using an Internet platform. *BMC Med* 2014;12:109.

35. Duncan P, Murphy M, Man MS, *et al*. Development and validation of the multimorbidity treatment burden questionnaire (MTBQ). *BMJ Open* 2018;8:e019413.

36. Langhorne P, Williams BO, Gilchrist W, *et al*. Do stroke units save lives? *The Lancet* 1993;342:395–8.