Patient engagement in multimorbidity: a systematic review of patient-reported outcome measures.

Short title: A systematic review of patient-reported engagement measures

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Abstract

Background. People with multimorbidity are increasingly engaged, enabled, and empowered to take responsibility for managing their health status. The purpose of the study was to systematically review and appraise the psychometric properties of tools measuring patient engagement in adults with multimorbidity and their applicability for use within engagement programs.

Method. PubMed, Scopus, Web of Science, and PsycInfo were searched from inception to 1 July 2021. Grey literature was searched using EBSCO host-database "Open dissertation". The reference lists of studies meeting the inclusion criteria were searched to identify additional eligible studies. The screening of the search results and the data extraction were performed independently by two reviewers. The methodological quality of the included studies was evaluated with the COSMIN checklist. Relevant data from all included articles were extracted and summarized in evidence synthesis tables.

Results. Twenty articles on eight tools were included. We included tools that measure all four dimensions of patient engagement (i.e., engagement, empowerment, activation, and participation). Their psychometric properties were analyzed separately. Most tools were developed in the last 10 years in Europe or the USA. The comparison of the estimated psychometric properties of the retrieved tools highlighted a significant lack of reliable patient engagement measures for people with multimorbidity. Available measures capture a diversity of constructs and have very limited evidence of psychometric properties that are vital for patient-reported measures, such as invariance, reliability, and responsiveness.

Conclusion. This review clarifies how patient engagement, as operationalized in measures purporting to capture this concept, overlaps with, and differs from other related constructs in adults with multimorbidity. The methodological quality of psychometric tools measuring patient engagement in adults with multimorbidity could be improved.

Keywords: patient engagement; patient empowerment; multimorbidity; assessment; measures; PROMS; PREMS

Background

In recent years the population ageing has led to increase the proportion of people with multiple chronic conditions (i.e. multimorbidity) [1]. Risky habits and lifestyles, longer life expectancy, and improved health care have led one in three adults to suffer from multimorbidity [2]. People with multimorbidity are individuals who live with two or more long-term conditions, one of which is either physical non-communicable disease or a mental health condition, or an infectious disease of long duration [1]. People with multiple long-term conditions are challenging to treat, are prone to experience complications such as readmissions, adverse drug interactions or death, and often require a great deal of social and psychological support [1,2]. Moreover, the risk of being diagnosed with multiple long-term conditions rises with age, is more common among women and in people of lower socio-economic status [1,2]. People with multimorbidity often report difficulties in managing their care pathways that are often designed to control and treat single health conditions [3]. Collectively this makes caring for these people, particularly challenging. Clinicians often struggle to find, personalize, and provide the best therapeutic pathways, interventions, and protocols for people with multiple long-term conditions [4].

Simultaneously, Western culture has gradually shifted from a paternalistic care approach toward patient-centered care and participatory medicine [5, 6]. People with multimorbidity are increasingly engaged, enabled, and empowered to take responsibility for managing their health [7]. Health researchers and stakeholders have started to design, test, and implement engagement interventions for people with multiple long-term conditions, showing their positive effects on health outcomes, user satisfaction, communication between patients and health professionals, adherence to treatment regimes, and healthcare resources usage [8, 9]. This has led to the increased relevance of the concept of patient engagement and its synonyms (e.g., patient empowerment, activation, participation) in the literature [11, 12]. In the last ten years, several studies have attempted to clarify the concept of patient engagement [13-15]. Menichetti et al. [16] highlighted that many concepts in the current literature overlap with patient engagement, such as patient enablement, empowerment,

activation, and participation, since all these concepts refer to people' proactive role in the management of their own healthcare.

ln this context, the use of tools designed and tested to engage people with multiple long-term diseases should be promoted among clinicians. Despite longstanding calls for greater engagement of older adults with multiple long-term conditions in healthcare, current evidence suggests that this population can be successfully engaged [17,18]. People with multiple long-term diseases are a diverse group, ranging from relatively healthy, independent living individuals to very frail individuals with poor physical functioning and cognitive problems, which often can make patient engagement in healthcare a challenging goal.

Therefore, a systematic review of the available engagement measurement tools to evaluate and monitor the benefits of engagement programs for people with multiple long-term conditions may help clinicians improve their care pathways. In particular, the examination of reliability, validity, feasibility, and clinical utility of engagement tools is required to inform the selection of appropriate instruments and address how to effectively enhance engagement in individuals and groups. Thus, the main object of the study was to systematically review and appraise the psychometric properties of tools measuring patient engagement in adults with multimorbidity and their applicability for use within empowerment programs, with a distinct focus on tools which have been validated in people with cardiovascular diseases.

This systematic review has been guided by the following research questions:

- What tools have been developed and validated in the literature to measure patient engagement in adults with multiple long-term conditions?
- What are the best tools, in terms of methodological quality and goodness-of-fit, to measure patient engagement in adults with multiple long-term conditions?
- What are the main conceptual components of engagement tools to shape future engagement interventions in this population?

Methods

2.1 Design

This study was performed in two steps: (i) a systematic review of the psychometric properties of engagement scales and tools was performed; then (ii) the psychometric properties were assessed by following the COnsensus-based Standards for the selection of health Measurement Instruments (COSMIN) guideline for systematic reviews of patient-reported outcome measures [19, 20]. The study protocol was registered on PROSPERO (registration number: CRD42021259968).

2.2 Search methods

A search strategy was designed to retrieve published and unpublished studies measuring patient engagement in adults with long-term conditions (Supplemental File 1). The search filters developed by the Oxford PROM group and Terwee et al. were then used to refine the search strategy [21]. Pubmed, Scopus, Web of Science, and PsycInfo were searched from their inception to April 2024. Grey literature was checked on EBSCOhost-database "Open dissertation" to identify any other significant publications. A forward and backward snowball search was performed to identify additional relevant publications.

The following eligibility criteria were used to select studies: (a) concerned with the development and/or evaluation of measurement properties of instruments that measure engagement and all the related concept such as empowerment, patient participation and patient involvement; (b) including adults with long-term conditions, including either instruments validated on people with multiple long term conditions or validated on people with at least three different long-term conditions; (c) published or unpublished up to April 2024; and (d) available in a language accessible to the authors (English and Italian). Tools were excluded if they: (a) were based on a single item. The literature search was performed by one researcher and then two researchers independently screened the records

based on the title and abstract against the inclusion criteria. For eligible studies, the full texts were retrieved, and the same two researchers independently evaluated the eligibility of each study, and decisions on study inclusion were based on joint agreement.

Data extraction was performed by two researchers and the following data was recorded: (i) author, year and country; (ii) language and setting; (iii) study design; (iv) key characteristics of study subjects; (v) name of measurement instruments and domains measured; (vi) number of items and (sub)scales and number and type of response categories; (vii) recall period and time needed for administration; (viii) scoring algorithm; (ix) mode of administration; (x) instructions given to those who complete the questionnaire; and (xi) licensing information and costs. The psychometric properties reported in the studies were independently extracted by four authors. Then, another researcher independently revised the data extracted for accuracy. Any changes were discussed, and a full agreement was reached among the researchers.

2.3 Quality appraisal

The COSMIN checklist [22] was used to evaluate the methodological quality of studies on measurement properties. The checklist uses a standardized descriptive framework to assess the measurement properties against quality markers in ten boxes [22]. Each box includes a pool of items (from five to 18) scored on a four-point scale (from 1 ‘poor’ to 4 ‘excellent’). The overall score is obtained by taking the lowest score indicated by the items in the box: therefore, a final score is given for each psychometric property, ranging from ‘poor’ to ‘excellent’. The measurement property ‘criterion validity’ was not considered in this systematic review since no “gold standard” exists for measuring engagement; therefore, eight boxes were rated. One researcher underwent training in the use of the COSMIN guidelines while the second reviewer had previous experience in the field. The inter-rater agreement between the two reviewers for the quality appraisal was 86.36% (k=0.79).

2.4 Synthesis

Included validation studies have been summarized according to the data extracted. The values of the psychometric properties evaluated, and the quality of the methodologies used in assessing these psychometric properties have been also summarized using a descriptive approach. The conceptual components for future engagement interventions were synthesized based on the conceptual framework underlying the single engagement tools.

Results

The literature search produced 6561 results, of which 942 duplicates were excluded. A total of 5473 articles were excluded at the title and abstract screening stage, while other 123 articles were excluded at the full-text stage. Twenty-three articles [23-42;] met the inclusion criteria describing eight families of tools as reported in Figure 1.

---- *Please insert Figure 1 here* ----

3.1 Study features

The main characteristics of the 23 articles [23-42] are reported in Table 1. The eight families of tools were categorized as those used to measure patient engagement in managing their own health and those used to measure patient engagement in managing their healthcare pathways (Table 1). Most studies validated or investigated the psychometric properties of the following tools: (i) the Patient Activation Measurement (PAM) (n=10) [29-37]; (ii) The Patient Assessment Care for Chronic Conditions (PACIC) (n=3) [38-40]; and (iii) The Patient Health Engagement Scale (PHE-S®) (n=5) [23-26].

The majority (78%) of the included studies were published in the last 10 years and included patients from 15 different countries, mainly North America (e.g., USA, Canada) and Europe (e.g., Denmark, Netherlands, UK, Italy) (Table 1). Six studies focused on the development and validation of these tools, while the others were adaptation, translation, and evaluation of their psychometric properties (Table 1). Among primary studies, the first data collection was performed in 2003 [37].

Overall, the number of participants involved ranged from 114 [23] to 5184 patients [31]. The response rate was only reported in ten studies and ranged from 48% [37] to 96.2% [24]. As shown in Table 1, tools were mainly validated among patients with diabetes (66%), hypertension and other cardiovascular morbidities (52%), or on people with multiple long-term conditions (23%). Most participants were female, and the mean age of participants varied from 37 [25] to 74 years old [59]. The ethnicity of participants was only reported in eleven studies, and most participants were Caucasian. Most of the scales required patients to have a basic level of health literacy. Patients with cognitive or mental health problems were often excluded from the validation studies.

Almost all tools were validated either in hospitalized (35%) or in primary care populations (65%), except Rademakers et al. [31] which employed data from both settings. All the included tools were self-report questionnaires. Few studies reported the completion time and ranged from less seven minutes [39] to 12 minutes [23]; only one study reported the administration time [52].

Table 1. Characteristics of the included studies

Prom	References	Aim	Language	Final number of item and subscale	Type of response	Population, (%)	N	Age, mean (D) yrs	Setting
Tools to measure patient engagement in managing their health									
PHE-S®	Usta et al., 2019	To assess the psychometric properties of PHE-s in Turkish patients with chronic diseases.	Turkish	5 items	7-point Likert scale	Diabetes mellitus (33); hypertension (28.9); Cancer (21.9%); Cardiovascular disorders (18.4); chronic renal failure (13.2), rheumatologic disorders (9.7), Chronic obstructive pulmonary disease (7.9%)	114	55.9 (14.5)	Hospital
	Zhang et al., 2017	To translate the original, PHE-s into Chinese Mandarin and to evaluate its psychometric properties in a group of patients with chronic disease in China.	Chinese	5 items	7-point Likert scale	Hypertension (71), diabetes (29.2); cardiovascular disease (27.1.); cerebrovascular disease (13.3); Chronic obstructive pulmonary	377	53.8(11)	Primary care

Prom	References	Aim	Language	Final number of item and subscale	Type of response	Population, (%)	N	Age, mean (D) yrs	Setting
						disease (10.4), cancer (2.4)			
	Magallares et al., 2017	To adapt the Patient Health Engagement scale to the Spanish population (S.PHE-s) following the guidelines for cross-cultural adaptations.	Spanish	5 items	7-point Likert scale	Hypothyroidism (16.9); Hypertension (12.3%); Crohn disease (7); asthma (6.8); migraine (6.5); diabetes (4.8), others	413	37.1(11.8)	primary care
	Graffigna et al., 2015	to validate the patient Health Engagement Scale.	Italian	5 items	7-point Likert scale	Asthma (16.4); Hypertension (35.6), Cardiovascular disorder (15.3); chronic obstructive pulmonary disorder (4), cancer (21), fibromialgy (5.2), artritereumatoide (7.3); osteoarthritis (7.3); hypercholesterolemia (10.3); allergy (16.6)	430	51.3(NR)	hospital
	Changizi et al., 2023	To evaluate the psychometric features of the PHE-scale in	Iranian	5 items	7-point Likert scale	Long-term breast cancer	128	26-65 (8.11)	Hospital

Prom	References	Aim	Language	Final number of item and subscale	Type of response	Population, (%)	N	Age, mean (D) yrs	Setting
		Iranian patients with breast cancer							
PAM-13	Rademakers et al., 2016	to compare the psychometric properties in studies from the different countries and establish whether the scores on the PAM vary between the studies.	Danish; Dutch; German; Norwegian; English	13 items	five possible responses, scoring ranging from 0-4	Adults with multiple chronic diseases from five different countries	5184	45-97*	primary care & hospital
	Schmaderer et al., 2015	to investigate the psychometric properties of the PAM in patients with multimorbidity in the hospital setting.	English	13 items	five possible responses, scoring ranging from 1-4	Adults discharged from an acute care facility with three or more chronic diseases	313	62.7(15)	hospital
	Skolasky et al., 2010	to determine the psychometric properties of PAM among multimorbid older adults and evaluate a theoretical, four-stage model of patient activation.	English	13 items	five possible responses, scoring ranging from 1-4	Adults with an average of four multiple chronic diseases each	853	56.6 (12.9)	primary care

Prom	References	Aim	Language	Final number of item and subscale	Type of response	Population, (%)	N	Age, mean (D) yrs	Setting
	Kosar et al., 2019	to test the reliability and validity of a Patient Activation Measure.	Turkish	13 items	five possible responses, scoring ranging from 0-4	Adults with multiple chronic diseases	130	56.7(13.8)	primary care
	Zeng et al., 2019	to assess the reliability and validity of the PAM13 in Chinese patients with hypertension and/or diabetes in a community management setting.	Chinese	13 items	five possible responses, scoring ranging from 0-4	Hypertension (59.3), diabetes (17.9), hypertension and diabetes (22.8)	509	67.2(8.9)	primary care
	Moreno-Chico et al., 2017	To develop a European Spanish adaptation of the original PAM-13 and to examine its psychometric properties in a sample of chronic patients.	Spanish	13 items	five possible responses, scoring ranging from 1-4	High blood-pressure (69.2); diabetes (66.3); dyslipidemia (49) and COPD (25.5)	208	65.8(9.45)	primary care
	Graffigna et al., 2015	to validate a culturally adapted Italian Patient Activation Measure (PAM13-	Italian	13 items & 1 dimensions	5-point Likert scale	Hypertension (20.2), Cardiovascular disorder (29.1), asthma	529	53.0(17.1)	hospital

Prom	References	Aim	Language	Final number of item and subscale	Type of response	Population, (%)	N	Age, mean (D) yrs	Setting
		I) for patients with chronic conditions.				(16.4) COPD (4) diabetes (16.2) cardiovascular disorder (29.1) oncology (21) fibromyalgia (5.2) osteoarthrosis (7.3) artritereumatoide (7.3); hypercholesterolemia (10.2) allergy (16.6)			
	Kerari et al., 2023	to determine the psychometric properties of the Arabic version of the Patient Activation Measure.	Arabic	13 items	five possible responses, scoring ranging from 1-4	Adults with chronic conditions (40)	225	53 (12.5)	Primary care
	Zakeri et al., 2023	to translate the American versions of the PAM-13 into Persian and test the psychometric properties of the Persian version among chronic patients	Persian			ischemic heart disease (IHD) (42,9), diabetes mellitus (DM) (12.6), hypertension (16.7), congestive heart failure (CHF) (10.3), chronic obstructive pulmonary	438	62.21 (13.39)	Hospital

Prom	References	Aim	Language	Final number of item and subscale	Type of response	Population, (%)	N	Age, mean (D) yrs	Setting
						disease (COPD) (9.4), other (8.2): chronic kidney disease (CKD), multiple sclerosis (MS), rheumatoid arthritis (RA), cancer, psychological disorders			
PAM-22	Cunha et al., 2018	To adapt and validate the Patient Activation Measure (PAM22) in a sample of Brazilians with chronic diseases under outpatient monitoring.	Portuguese	22 items, 4 subscales	five possible responses, scoring ranging from 1-4	Cancer (13.6) HIV/Aids (9.7) rheumatoid arthritis (9.9) systemic lupus erythematosus (6.8) Cron's disease (7.8) diabetes (9.7) ulcerative RECTOCOLITIS (4.9) OBESITY (5.8) coronary insufficiency (8) chronic renal insufficiency (5.5) systemic arterial	513	49.9(14.6)	primary care

Prom	References	Aim	Language	Final number of item and subscale	Type of response	Population, (%)	N	Age, mean (D) yrs	Setting
						hypertension (9.6) cardiac failure (8.9) Cardiac failure (8.6%)			
	Hibbard et al., 2004	to develop a measure for assessing ‘‘activation,’’ and the psychometric properties of that measure.	English	22 items, 4 subscales	5-point Likert scale	Angina/heart problem (13), Hypertension (34) arthritis (38) chronic pain(25) depression (15) diabetes (11) lung disease (12) cancer (5) high cholesterol (30)	1515	45–54*	primary care
HES	Serrani et al., 2014	to translate and adapt the Health Empowerment Scale (HES) for a Spanish-speaking older adults’ sample and perform its psychometric validation.	Spanish	8 items	5-point Likert Scale from 5 to 1	Hypertension (58.8) arthritis (40.3) diabetes (20.7) hyperlipidemia (17.1)	648	74.8(11.6)	primary care
Small’s scale	Small et al., 2013	to report on two empirical studies conducted to understand and measure empowerment in patients with long-term conditions in primary care.	English	8 items	4-point Likert scale	Diabetes (46.2) COPD (13.2) irritable bowel syndrome (21.8) arthritis (52.3) anxiety and depression (26.9) asthma (15.7)	197	62.8(14.3)	primary care

Prom	References	Aim	Language	Final number of item and subscale	Type of response	Population, (%)	N	Age, mean (D) yrs	Setting
						Coronary heart disease (16.8) Heart problems or high blood pressure (52.8)			
Tools to measure patient engagement in managing their healthcare pathways									
PACIC	Wensing et al., 2008	to develop and test a Dutch version of the PACIC questionnaire, a measure for patient reported structured chronic care.	Dutch	20 item & 5 subscales	five-point response scale, ranging from 1 to 5	Adults with diabetes and/or COPD	165	68(10.3)	primary care
	Glasgow et al., 2005	To develop and validate the Patient Assessment of Chronic Illness Care (PACIC)	English	20 items & 5 subscales	five-point response scale, ranging from 1 to 5	Adults with two different chronic conditions	266	64.2(10.5)	primary care
PPQ	Berg et al., 2020	To develop an instrument to measure patient participation in health care and to investigate the measurement properties of the Patient	Danish	16 items & 4 subscales	4-point Likert Scale from 1 to 4	Hypertension (33) diabetes (13) cancer (5) depression (4)	378	<65	hospital

Prom	References	Aim	Language	Final number of item and subscale	Type of response	Population, (%)	N	Age, mean (D) yrs	Setting
		Participation Questionnaire (PPQ).							
PPET	Jerofke-Owen & Garnier-Villarreal, 2020	to develop and psychometrically test the Patient Preferences for Engagement Tool (PPET).	English	29 items	5-point Likert rating scale	Hypertension (34.7); heart disease (24.4); dyslipidemia (20.5); asthma (11); COPD (8.5) diabetes mellitus (22.7); arthritis (17.2); cancer (26.6)	308	58.2 (17.1)	hospital
PRE-HIT	Koopman et al., 2014	to measure patient readiness to engage with health technologies among adult patients with chronic conditions.	English	28 items	4-point Likert scale	Hypertension (81), coronary artery disease (12) diabetes mellitus (39) heart failure (11)	200	54(14)	primary care

Note: NR= not reported; * = age range in years.

The number of evaluated psychometric properties ranged from two to six (Table 2). The most commonly assessed properties were structural validity and internal consistency. Only two studies evaluated measurement error [26,37]. None of the included studies evaluated measurement variance. However, given that the items included are a manifestation of different underlying constructs, these properties were evaluated individually for each group of tools (Table 2).

3.1.1 Tools to measure patient engagement in managing their health

Five tools to measure patient engagement in managing their health were retrieved (Table 1).

The Patient Health Engagement Scale (PHE-S®) is a patient self-administered short psychometric questionnaire developed to measure the level of patient engagement in their healthcare function [26]. It consists of five items measured on a 7-point Likert scale, that allows patients to easily mirror their current emotional states and illnesses experience. The PHE-S® has a robust theoretical foundation since it was developed from the Patient Health Engagement model [26]. Currently, six versions of this scale are available: Italian [26]; English [26]; Turkish [23]; Spanish [25]; Chinese [24]; Persian [XXX]. Across these tools, the psychometric properties remain the same as the original version (Table 2), demonstrating the consistency of PHE-S®. All the validation studies tested the internal consistency of the tool. Structural validity was evaluated using the Categorical Principal Component Analysis (CATPCA), a confirmatory factor analysis (CFA) and a RASCH model (Table 2). Reliability was evaluated in three studies (from acceptable to very good), while cross-cultural validity was assessed in two (Table 2). All the PHE-S psychometric properties were judged as good or adequate. The only exception was the reliability of the Turkish version which was judged as doubtful (Table 2).

The Patient Activation Measure (PAM) [37] is a well-known tool to assess patients' knowledge, skills, and confidence for managing their health. There are currently two versions of the PAM, the original 22-item (PAM-22) and the 13-item short form (PAM-13). The PAM measures patient activation on a 0-100 scale, and the patients' responses are measured on a 5-point Likert scale.

Several translations and validations of the PAM are available (Table 1), as well as the original version developed by Hibbard et al. [37]. The PAM shows different judgements of its psychometric properties among its validations: in some of the studies, the PAM demonstrated good construct validity, reliability, and internal consistency overall, in others the judgement is doubtful or inadequate (Table 2). However, the PAM is the only patient activation measures retrieved that has been validated in a wide range of chronic or multimorbid populations (Table 1).

The Health Empowerment Scale (HES) is a survey that measures patients' self-management skills and decision-making abilities [41]. The HES was adapted from the Diabetes Empowerment Short Form Scale (DES-SSF) and has 8 items measured on a 5-point Likert scale. The HES shows good internal consistency, construct validity and adequate reliability (Table 2). Small floor and ceiling effects were reported (Table 2). Its content validity and theoretical conceptualization were judged as doubtful since the HES has no real underlying conceptual model. Other studies are needed to evaluate the consistency of the HES psychometric properties.

Small et al. [42] developed a short questionnaire to measure empowerment in patients with long-term conditions (primarily diabetes, irritable bowel syndrome, coronary heart disease, or chronic obstructive pulmonary disease). It has 8 items measured on a 4-point Likert scale. Its structural validity appears to be doubtful, and no content validity was provided (Table 2).

3.1.2 Tools to measure patient engagement in managing their healthcare pathways

Four tools measuring patient engagement in healthcare were identified.

The Patient Assessment of Care for Chronic Conditions (PACIC) is a survey that measures specific actions that chronic patients report they have experienced in the healthcare system [39]. The PACIC was developed from the Patient Centered model and has five subscales, measuring patients' activation, delivery system experience, goal setting, problem-solving, and coordination involvement. Five studies utilizing the PACIC were retrieved (Table 1). The PACIC is a 20-item questionnaire, and it uses a 5-point response scale, with higher scores indicating better quality of care. Similar to the

PAM, the various PACIC validation studies report different judgments of its psychometric properties (Table 2). The PACIC content validity has been assessed by Glasgow et al. [39] and was rated as inadequate. Its' structural validity was judged as very good only by two studies (Table 2). PACIC reliability was only assessed by three studies with two deeming its reliability as inadequate or doubtful.

The Patient Participation Questionnaire (PPQ) is an instrument developed to measure patient participation in their treatment and care [40]. It has been validated in patients with multi-morbidity, where one-third of the sample were patients with hypertension [40]. The PPQ is a short questionnaire with 16 items and four subscales, measured on a 4-point Likert scale. The PPQ has a good internal consistency, but its structural validity has been judged as doubtful, and no measures of its reliability have been provided yet (Table 2).

The Patient Readiness to Engage in Health Internet Technology (PRE-HIT) is a tool developed to measure the likelihood of using health information technology among patients with chronic conditions [28]. The PRE-HIT focuses on the measurement of patients' engagement in specific conditions and 28 items measured on a 4-point Likert scale. Only its content validity, internal consistency and reliability were reported (Table 2).

The Patient Preferences for Engagement (PPET) tool was developed to assess patients' preferences for engaging in healthcare [27]. The PPET was designed to inform the planning and delivery of individualized healthcare. The PPET consists of 29 items weighted with a 5-point Likert scale. No PPET composite score has been computed yet. The content validity was judged doubtful, while its reliability, structural validity, and internal consistency were rated as adequate or very good (Table 2). Other studies are needed to further evaluate the consistency of the PPET psychometric properties.

Table 2. Quality assessment of the included studies.

Instrument	**Authors and year**	**Internal consistency**	**Reliability**	**Content validity**	**Structural validity**	**Hypotheses testing**		**Cross-cultural validity**	**Floor and/or ceiling effect**
		α Cronbach	*ICC*	*S-ICV*	*Variance explained %, methods*	*Hypotheses*	*sub-groups*	*DIF analyses and forward-backward*	
PHE-s	Graffigna et al., 2015	0.87	0.95	NA	$\chi2 = 10.98$, CFI = 0.981, RMR = 0.018, RMSEA = 0.059	Invariance in the two subsamples divided by gender	by age and educational level	DIF backward-forward	small floor effect (range 1.7%-4.5%) moderate ceiling effect (range 27.6%-55%)
	Magallares et al., 2017	0.85.	NA	NA	$\chi2 = 1.88$, df = 4, p = .75; CFI = .99, RMR = .01, GFI = .99, RMSEA = .05	correlations with life satisfaction, medicine adherence behavior, anxiety, depression	by gender	Multigroup analyses forward-backward	no severe floor or ceiling effect
	Zhang et al., 2017	0.89	0.52-0.79.	0.92	$\chi2 = 6.65$, df = 4, p = 0.156; (CFI = 0.983, SRMR= 0.014, GFI = 0.979, RMSEA = 0.067	Positive correlation with patient activation and medication adherence	NA	NA forward-backward	no severe floor or ceiling effect
	Usta et al., 2019	0.80	0.61	0.89	CATPCA and Rasch analysis (varied 0.62 to 1.14)	NA	NA	NA forward-backward	NA

	Changizi et al., 2023	NA	NA	0.81	CATPCA and Rasch analysis (varied 0.658-0.932)	NA	NA	NA forward-backward	NA
PPET	Jorfke-Owen and Garnier-Villarreal, 2020	>0.7	NA	0.8	EFA = 45%, $\chi 2$ (309) = 453.35, CFI = 0.892, TLI = 0.878, RMSEA = 0.056, 90% CI [0.045, 0.067], SRMR = 0.125, gamma-hat = 0.933, gamma-hatadj = 0.918.	NA	by age, comorbidities, educational level, health perception	Multi group comparisons forward-backward	NA
PRE-HIT	Koopman et al., 2014	>70	0.60-0.85	Face validity	NA	NA	NA	NA backward-forward	NA
PPQ	Berg et al., 2020	0.89.	NA	NA	RMSEA=0.043, CFI=0.98; TLI=0.98	NA	NA	NA backward-forward	strong ceiling effect (range 34% to 94%)
SDM-Q-9	Scholl et al., 2012	0.92	.68	Face validity	NA	Correlation between OPTION and SDM-Q-9	NA	NA backward-forward	low variance due to ceiling

									effects and floor effects
HES	Serrani et al., 2014	α= 0.89	0.92	0.98	CFI, GFI and NNFI ≥0.90, and RMSEA ≤0.06; $\chi 2(634) = 5425.72$; $p< 0.001$; KMO = 0.890	correlations between the HES total and item scores and the General Self Efficacy Scale, Swedish Rheumatic Disease Empowerment Scale and Making Decisions Empowerment Scale	NA	NA backward-forward	Floor and ceiling effects were small (<20%)
Small's scale	Small et al., 2013	0.82	NA	NA	EFA =45.7%	hypothesize relationships with overall empowerment (or individual dimensions) based on existing theory or empirical data (self-efficacy; gender; patient enablement; quality of chronic care; age; ethnicity; level of education; etc.)	by comorbidities, gender, age, ethnicity, living arrangements, education, current work, depression, general health, and self-efficacy	Multi group comparisons backward-forward	NA
PACIC	Tusek-Bunc et al., 2014	0.93	Spearman correlation	NA	NA	NA	NA	NA forward-backward	NA

	Wensing et al., 2008	0.71-0.83	>0.70	NA	CFA=70% KMO = 0.844; Bartlett's test of spherity p= 0.000	higher PACIC scores positively correlated to both patients' perceived enablement after the latest visit to the GP and to patients' overall evaluations of general practice.	NA	NA forward-backward	several items might have floor or ceiling effects.
	Fan et al., 2014	0.96	NA	NA	CFA=74% RMSEA estimate of 0.09; CFI, 0.91; NFI, 0.90; and NNFI, 0.89.	NA	NA	NA forward-backward	floor and ceiling effects (range from 1.8% to 2%)
	Iglesias et al., 2014	NA	NA	NA	RMSEA <0.08, WRMR <0.1.00, CFI >0.97	correlation with demographic variable	by age, gender, education, comorbidities, annual blood pressure, weight and lipid measure	Multi group comparisons forward-backward	floor effect (range from 7-67%) & ceiling effect (range from 4-46%)
	Glasgow et al., 2005	0.84	test-retest reliability	Expert panel	NA	The PACIC and its scales would (a) generally not be related to patient demographics (eg, gender, age, education) but (b) would be related to disease characteristics (eg, number of comorbid conditions). The PACIC would be moderately related to, but not redundant, with measures of primary care and patient activation.	NA	NA backward-forward	no items had ceiling effect

PAM-13	Rademakers et al., 2016	0.80-0.88	test-retest reliability	NA	NA	NA	NA	NA forward-backward	NA
	Schmaderer et al., 2015	0.88	NA	0.91	$\chi 2$= 5 400.41, df 5 65, p.0.01.; SRMR=.087, RMSEA =.08 CFI =.89	PAM scores would have (a) an inverse relationship with depression, (b) a positive relationship with physical functional status and health care quality, and (c) no relationship with number of comorbidities or severity of illness.	by depression, functional status, and comorbidities	Multi group comparisons forward-backward	NA
	Skolasky et al., 2010	0.87	NA	NA	KMO=0.96	higher PAM scores are related to greater adherence to desirable health-related behaviors, higher functional status, and better health care quality. Patients' level of activation is not correlated with their number of comorbid conditions. Negative correlation between the PAM and comorbid conditions.	NA	NA forward-backward	NA
	Stepleman et al., 2010	NA	NA	NA	CFA	Correlation with MSSE, BDI-II and MS QOL, lower depression, and higher well-being	by age, educational level	Multi group comparisons forward-backward	NA

	Zeng et al., 2019	0.92	NA	NA	$\chi 2 = 139.3$, df = 59, P <0.001, RMSEA = 0.060, CFI = 0.957	NA	NA	NA forward-backward	floor effect (range 1.8-5.2%) and ceiling effect (range 21.4-28.1)
	Eyles et al., 2020	0.92	NA	NA	χ 2 =3901.0644, 3927 − 5 degrees of freedom, P = 0.61 (Kaiser-Meyer-Olkin value = 0.88 and Bartlett's Test of Sphericity $\chi 2 = 1404.0$, df 78, $p < 0.001$	moderate correlations between DASS and AQoL scores with PAM-13. Weak correlations (between PAM-13 and HOOS/KOOS 'Pain' and 'Function in daily living' subscale scores.	NA	DIF analysis forward-backward	no floor or ceiling effect
	Maindal et al., 2009	0.89	NA	NA	CFA=43.2%	NA	NA	DIF analysis forward-backward	Floor effect was small (range 0.6–3.6%), but the ceiling effect was above 15% for all items (range 18.6–62.7%).

	Graffigna et al., 2015	0.88	NA	NA	$\chi 2$= 2129.7, df = 78, p < 0.001; Kaiser-Mayer-Olkin measure of sampling adequacy was equal to 0.89.	NA	NA	DIF analysis forward-backward	small floor effect (range 1.7–4.5 %) and a moderate ceiling effect (range 27.6–55.0 %).
	Kapoor et al., 2020	0.84	NA	NA	NA	NA	NA	NA forward-backward	NA
	Kosar et al., 2019	0.81	0.98	NA	x2/df: 1.59, RMSEA: 0.071, CFI: 0.96, NNFI: 0.95, Kaiser Meyer Olkin coefficient was .75 and Barlett test was x2: 646.870; p: 0. 000.	NA	NA	NA forward-backward	NA
	Moreno-Chico et al., 2017	NA	NA	NA	Data showed a fit to the Rasch model	correlation between self-efficacy, quality of life, visits to the emergency room and number of hospitalisations	NA	DIF analysis forward-backward	NA
	Ngooi et al., 2016	0.86	NA	NA	CFA=77%	correlation with depression and self-efficacy	NA	DIF analysis forward-backward	All items had a small floor effect, but nine out of 13 items had a ceiling

									effect larger than 15 %.
	Laranjo et al., 2018	NA	NA	NA	The Rasch dimension explained 39.1% of the variance in the data.	NA	NA	DIF analysis forward-backward	no floor or ceiling effects.
	Hashim et al., 2020	0.87	NA	Face validity	EFA=60% KMO value was 0.86 and the p-value was < 0.0001 for Bartlett's test of sphericity.	NA	NA	NA forward-backward	small floor effect (range 0–3.1 %) and a moderate ceiling effect (range 5.4–26.9 %)
	Kerari et al., 2023	McDonald's omega 0.80	0.31 (item 2) to 0.57 (item 11)	NA	$\chi2 = 76.76$, df = 51, $p < 0.01$; TLI = 0.94; CFI = 0.96; RMSEA = 0.04 [90% CI = 0.02–0.07	NA	NA	Multi group comparisons forward-backward	N/A
	Zakeri et al., 2023	0.88	0.96	0.91	EFA $\chi2 = 1265.85$, df = 78, $p < 0.001$ KMO= 0.84 CFA $\chi2$/d.f. = 1.82, RMSEA = 0.077,	NA	NA	Multi group comparisons forward-backward	The floor effect was 5.2% (ranging from 2.3 to 10.3%), but the ceiling effect was

					SRMR = 0.055, GFI = 0.91, CFI = 0.97, IFI = 0.97, NNFI = 0.96, PNFI = 0.70)				26.19% (ranging from 17.3 to 33.7%).
PAM-22	Cunha et al., 2018	NA	0.26-0.64	NA	Rasch model	no relationship between activation, gender, and age of the participants. Positive correlation between activation and time of diagnosis of the chronic disease	NA	NA forward-backward	NA
	Hibbard et al., 2004	0.87	test retest reliability	assessed by expert panel	Rasch model	those with higher activation would be more likely to engage in specific self-care and preventive behaviors. Further, those with higher activation who have a specific chronic disease should be more likely to engage in the self-care behaviors specific to their condition (e.g., exercising to control arthritis pain). Similarly, it was hypothesized that those with higher measured activation should engage in other health ‘‘consumeristic’’ behaviors, such as seeking relevant health care information, being persistent in getting clear answers from providers, and using comparative	NA	NA backward-forward	NA

						performance information to make health care choices. Those with more activation would indicate less fatalism about their future health.			

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3.1.3 Conceptual components for future engagement interventions

According to the synthesis of the conceptual models or frameworks behind the tools included in this review, we extracted eight main conceptual components to be considered for future patient engagement interventions. The conceptual components are emotional adjustment, self-efficacy, self-management, health literacy, shared decision making, collaborative goal setting, proactive communication with the care teams, and problem solving (Table 3).

Emotional adjustment, mainly related to the "patient engagement" domain, - refers to the patients' ability to cope with the diagnosis and to elaborate their own role in the disease management. Self-management and self-efficacy – mainly related to the "patient activation domain" - are two well-known components of engagement interventions and refer to patients' ability to effectively recognize their needs and act proactively to fulfill them. Health literacy, mainly linked to the "patient empowerment" domain, refers to patients' knowledge and ability to understand information provided by the healthcare providers or caregivers about the disease and treatment journey. Also shared decision making and proactive communication are common conceptual components of engagement measurement tools. Indeed, shared decision making – which is mainly related to the "patient participation" domain - is essential in making them able to proactively manage their disease by enabling an open dialogue with the healthcare team about therapeutic choices and strategies. Collaborative goal setting and problem-solving, mainly related to the patient are crucial skills that make patients able to effectively plan self-care activities and to engage in proactive behaviors towards their disease management.

Table 3. Components of engagement interventions for patients diagnosed with multiple chronic diseases

Domain	Tool	Pillars for patient engagement interventions
Patient engagement		
	PHE-s	Emotional adjustment, proactive communication with the care team
	PPET	Health literacy, self-efficacy
Patient activation		
	PAM-13	Shared decision-making, health literacy, self-efficacy, self-management, goal setting, problem solving
	PAM-22	Shared decision-making, health literacy, self-efficacy, self-management, goal setting, problem solving
Patient participation		
	PACIC	Collaborative goal setting, problem solving, self-efficacy
	PRE-HIT	health literacy, self-efficacy, emotional adjustment
	PPQ	Shared decision making, self-efficacy
	SDM-Q-9	Shared decision making
Patient empowerment		
	HES	Shared decision making, self-efficacy, self-management skills, health literacy
	Small's scale	Emotional adjustment, shared decision making, self-management

Discussion

This systematic review retrieved eight different tools that measure patient engagement in people with multiple long-term diseases. The tools were analyzed separately, based on the construct they measured. Half of the tools retrieved focused on measuring patient engagement as the process of emotional adjustment and the acquisition of motivation to manage their disease or as a general process of acquisition of a higher level of power. The other half measured people's ability to take an active part in their consultations with healthcare professionals. Overall, the structure of the instruments was heterogeneous, as were their psychometric properties. Many tools only partially described their psychometric properties, with few outlining their theoretical foundation. The best psychometric properties were reported by the PAM® [37] and the PHE-S® [26], which are the most tested and cross-culturally validated measures of patient engagement in managing their health to date.

Most of the tools retrieved were developed and/or adapted in the last ten years, highlighting the growing importance of the concept of patient engagement in healthcare. The tools were tested mainly in populations with diabetes or hypertension. This is not surprising given the mean age of people with long-term conditions [43] and the importance of engaging with these people to help them achieve a suitable quality of life [44,45]. Most instruments were short (< 15 items) and had a short completion time (less than 10 minutes). The psychometric properties most often measured and reported were internal validity, content validity and construct validity. Many tools which showed a good theoretical foundation and reliability (Table 2), lacked a formal assessment of their structural validity. It is important that future studies further clarify the construct validity of these tools. Floor and ceiling effects were reported with some tools, and this may be problematic as the response scale of these instruments was all measured using Likert scales. Only three tools (PAM, PACIC, and PHE-S®) were tested in more than two different populations. This highlights the importance of increasing the dissemination of the concept of engagement and its measurement tools across healthcare conditions and especially in developing countries.

None of the identified tools measured both patient engagement in managing their own health and the healthcare pathways. This may be due to the lack of consensus on a unique definition of patient engagement [13-15]. Patient engagement is a construct that in the literature overlaps with other psychological constructs such as activation, participation, and empowerment. However, even if many of these concepts are strongly intersecting (e.g., patient engagement and patient empowerment), others clearly measure different aspects of the process of engagement (e.g., patient participation). This problem was originally highlighted by Fumagalli and colleagues in 2015 [13] and almost seven years later remains unresolved. The development of a single tool that measures all the different constructs underlying the concept of patient engagement may be an effective way to ease the process of measuring engagement.

To our knowledge, only one previous review has focused on measuring the concept of patient engagement in healthcare. Jerofke-Owen et al. [46] limited their review on tools measuring patients' preferences for engagement in healthcare; however, they did not systematically retrieve and evaluated also the tools measuring patients' engagement in managing their own health. While this approach may increase accuracy in the analysis of the finding, given the lack of clarity on the concept of engagement it could also limit the ability to synthesize the concept's use in the literature and lead to the loss of many valuable tools. Instead, we choose to use an inclusive approach to gain a deeper understanding of all the tools available to measure the concept of patient engagement.

This review allowed us to reflect on the components that should characterize engagement interventions in the future. The conceptual models and frameworks of the engagement tools are characterized by components such as emotional adjustment, self-efficacy, self-management, health literacy, shared decision making, collaborative goal setting, proactive communication with the care teams, and problem-solving. Some of these components (e.g., shared decision making, and proactive communication with the care team) are particularly important to identify the best care pathways for people with multiple chronic conditions. Others instead (e.g., emotional adjustment, self-efficacy, self-management) are necessary to guarantee that people with multiple chronic conditions are

confident and able to partake in complex decisions on prognosis, treatment options and prioritizing care driven by their own perspective on what is acceptable, feasible or meaningful. These findings suggest that future engagement interventions should consider all these components to be effective. Current literature on patient engagement intervention for people with multiple long-term conditions is very heterogeneous [45]. This diversity in the evidence base challenges the ability to draw robust conclusions and the increasing interest in patient engagement in the last ten years in Europe and America sets the stage for reflection.

This review has some limitations. Firstly, while there are many different related concepts of engagement, some central terms might be lacking. Therefore, we excluded some concepts, for instance, self-care, patient adherence, or patient compliance although they have been used as related concepts of engagement. From our perspective, these concepts are outcomes of engagement. We chose the concepts which have in recent years been used as describing the active role of patients in healthcare [13,25], assuming they had an up-to-date view of related concepts. Secondly, some measures were rather new, and their validation process may be still ongoing. Lastly, it is possible that some relevant articles written in languages other than English or Italian may have been missed.

Conclusions

This systematic review highlights the need for a more comprehensive measure of patient engagement which includes all its related concepts (i.e., patient empowerment, patient activation, patient participation) and addresses all the possible components of patient engagement (i.e., emotional adjustment, self-efficacy, self-management, health literacy, shared decision making, collaborative goal setting, proactive communication with the care teams, problem-solving). Despite policy interest and initiatives relating to patient engagement, there is limited evidence to support the reliability and validity of existing tools and for the specific application to people with multiple long-term conditions. Moreover, retrieved studies often lack cross-cultural validation of the measures. This is particularly relevant as research suggests that there are ethnic differences in illness perception and

management [47,48]. Future research could usefully develop a definitive more comprehensive measure of patient engagement.

Declarations:

Ethics approval and consent to participate: Not applicable

Consent for publication: Not applicable

Availability of data and materials: The datasets used and/or analysed during the current study are available from the corresponding author on reasonable request.

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196 **References**

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198 1. World Health Organization, 2016. Multimorbidity. Available from:
199 https://apps.who.int/iris/bitstream/handle/10665/252275/9789241511650-eng.pdf
200 2. Divo MJ, Martinez CH, Mannino DM. Ageing and the epidemiology of multimorbidity. Eur
201 Respir J. 2014 Oct;44(4):1055-68. doi: 10.1183/09031936.00059814. Epub 2014 Aug 19.
202 PMID: 25142482; PMCID: PMC4918092.
203 3. Dhere A. Managing complex long-term conditions and multimorbidity. Clin Med (Lond).
204 2016 Dec;16(6):545-547. doi: 10.7861/clinmedicine.16-6-545. PMID: 27927819; PMCID:
205 PMC6297343.
206 4. Committee on the Learning Health Care System in America; Institute of Medicine; Smith M,
207 Saunders R, Stuckhardt L, et al., editors.Washington (DC): National Academies Press (US);
208 2013 May 10.
209 5. deBronkart D. The patient's voice in the emerging era of participatory medicine. Int J
210 Psychiatry Med. 2018 Nov;53(5-6):350-360. doi: 10.1177/0091217418791461. Epub 2018
211 Aug 16. PMID: 30114957.
212 6. Weil AR. The Patient Engagement Imperative. Health Aff (Millwood). 2016 Apr;35(4):563.
213 doi: 10.1377/hlthaff.2016.0337. PMID: 27044951.
214 7. Pushparajah DS. Making Patient Engagement a Reality. Patient. 2018 Feb;11(1):1-8. doi:
215 10.1007/s40271-017-0264-6. PMID: 28741235; PMCID: PMC5766722.
216 8. Bombard Y, Baker GR, Orlando E, Fancott C, Bhatia P, Casalino S, Onate K, Denis JL,
217 Pomey MP. Engaging patients to improve quality of care: a systematic review. Implement
218 Sci. 2018 Jul 26;13(1):98. doi: 10.1186/s13012-018-0784-z. PMID: 30045735; PMCID:
219 PMC6060529.

9. Barello S, Triberti S, Graffigna G, Libreri C, Serino S, Hibbard J, Riva G. eHealth for Patient Engagement: A Systematic Review. Front Psychol. 2016 Jan 8;6:2013. doi: 10.3389/fpsyg.2015.02013. PMID: 26779108; PMCID: PMC4705444.
10. Rossi MC, Lucisano G, Funnell M, Pintaudi B, Bulotta A, Gentile S, Scardapane M, Skovlund SE, Vespasiani G, Nicolucci A; BENCH-D Study Group. Interplay among patient empowerment and clinical and person-centered outcomes in type 2 diabetes. The BENCH-D study. Patient Educ Couns. 2015 Sep;98(9):1142-9. doi: 10.1016/j.pec.2015.05.012. Epub 2015 May 21. PMID: 26049679.
11. Náfrádi L, Nakamoto K, Schulz PJ. Is patient empowerment the key to promote adherence? A systematic review of the relationship between self-efficacy, health locus of control and medication adherence. PLoS One. 2017 Oct 17;12(10):e0186458. doi: 10.1371/journal.pone.0186458. PMID: 29040335; PMCID: PMC5645121.
12. Castro EM, Van Regenmortel T, Vanhaecht K, Sermeus W, Van Hecke A. Patient empowerment, patient participation and patient-centeredness in hospital care: A concept analysis based on a literature review. Patient Educ Couns. 2016 Dec;99(12):1923-1939. doi: 10.1016/j.pec.2016.07.026. Epub 2016 Jul 18. PMID: 27450481
13. Fumagalli LP, Radaelli G, Lettieri E, Bertele' P, Masella C. Patient Empowerment and its neighbours: clarifying the boundaries and their mutual relationships. Health Policy. 2015 Mar;119(3):384-94. doi: 10.1016/j.healthpol.2014.10.017. Epub 2014 Nov 5. PMID: 25467286.
14. Higgins T, Larson E, Schnall R. Unraveling the meaning of patient engagement: A concept analysis. Patient Educ Couns. 2017 Jan;100(1):30-36. doi: 10.1016/j.pec.2016.09.002. Epub 2016 Sep 3. PMID: 27665500.
15. Barello S, Graffigna G, Vegni E. Patient engagement as an emerging challenge for healthcare services: mapping the literature. Nurs Res Pract. 2012;2012:905934. doi: 10.1155/2012/905934. Epub 2012 Oct 31. PMID: 23213497; PMCID: PMC3504449.

16. Menichetti J, Libreri C, Lozza E, Graffigna G. Giving patients a starring role in their own care: a bibliometric analysis of the on-going literature debate. Health Expect. 2016 Jun;19(3):516-26. doi: 10.1111/hex.12299. Epub 2014 Nov 4. PMID: 25369557; PMCID: PMC5055237.
17. Markle-Reid M, Ganann R, Ploeg J, Heald-Taylor G, Kennedy L, McAiney C, Valaitis R. Engagement of older adults with multimorbidity as patient research partners: Lessons from a patient-oriented research program. J Comorb. 2021 Mar 17;11:2633556521999508. doi: 10.1177/2633556521999508. PMID: 33796472; PMCID: PMC7975523.
18. Dambha-Miller H, Simpson G, Hobson L, Roderick P, Little P, Everitt H, Santer M. Integrated primary care and social services for older adults with multimorbidity in England: a scoping review. BMC Geriatr. 2021 Dec 3;21(1):674. doi: 10.1186/s12877-021-02618-8. PMID: 34861831; PMCID: PMC8642958.
19. Mokkink LB, Prinsen CA, Bouter LM, Vet HC, Terwee CB. The COnsensus-based Standards for the selection of health Measurement INstruments (COSMIN) and how to select an outcome measurement instrument. Braz J Phys Ther. 2016 Jan 19;20(2):105-13. doi: 10.1590/bjpt-rbf.2014.0143. PMID: 26786084; PMCID: PMC4900032.
20. Prinsen CAC, Mokkink LB, Bouter LM, Alonso J, Patrick DL, de Vet HCW, Terwee CB. COSMIN guideline for systematic reviews of patient-reported outcome measures. Qual Life Res. 2018 May;27(5):1147-1157. doi: 10.1007/s11136-018-1798-3. Epub 2018 Feb 12. PMID: 29435801; PMCID: PMC5891568.
21. Terwee CB, Bot SD, de Boer MR, van der Windt DA, Knol DL, Dekker J, Bouter LM, de Vet HC. Quality criteria were proposed for measurement properties of health status questionnaires. J Clin Epidemiol. 2007 Jan;60(1):34-42. doi: 10.1016/j.jclinepi.2006.03.012. Epub 2006 Aug 24. PMID: 17161752.
22. Mokkink LB, de Vet HCW, Prinsen CAC, Patrick DL, Alonso J, Bouter LM, Terwee CB. COSMIN Risk of Bias checklist for systematic reviews of Patient-Reported Outcome

Measures. Qual Life Res. 2018 May;27(5):1171-1179. doi: 10.1007/s11136-017-1765-4. Epub 2017 Dec 19. PMID: 29260445; PMCID: PMC5891552.

23. Usta D, Korkmaz F, Akyar I, Bonanomi A. Patient Health Engagement Scale (PHE-s): Validity and reliability for Turkish patients with chronic diseases, *CUKUROVA UNIVERSITESI TIP FAKULTESI DERGISI 2019. 44(*3): 1055-1063. 10.17826/cumj.482420

24. Zhang Y, Graffigna G, Bonanomi A, Choi KC, Barello S, Mao P, Feng H. Adaptation and Validation of a Chinese Version of Patient Health Engagement Scale for Patients with Chronic Disease. Front Psychol. 2017 Feb 6;8:104. doi: 10.3389/fpsyg.2017.00104. PMID: 28220090; PMCID: PMC5292425.

25. Magallares A, Graffigna G, Barello S, Bonanomi A, Lozza E. Spanish adaptation of the Patient Health Engagement scale (S.PHE-s)in patients with chronic diseases. Psicothema. 2017 Aug;29(3):408-413. doi: 10.7334/psicothema2017.75. PMID: 28693715.

26. Graffigna G, Barello S, Bonanomi A, Lozza E. Measuring patient engagement: development and psychometric properties of the Patient Health Engagement (PHE) Scale. Front Psychol. 2015 Mar 27;6:274. doi: 10.3389/fpsyg.2015.00274. PMID: 25870566; PMCID: PMC4376060.

27. Jerofke-Owen TA, Garnier-Villarreal M. Development and Psychometric Analysis of the Patient Preferences for Engagement Tool. Nurs Res. 2020 Jul/Aug;69(4):289-298. doi: 10.1097/NNR.0000000000000423. PMID: 31977839.

28. Koopman, R.J., Petroski, G.F., Canfield, S.M. *et al.* Development of the PRE-HIT instrument: patient readiness to engage in health information technology. *BMC Fam Pract* **15,** 18 (2014). https://doi.org/10.1186/1471-2296-15-18

29. Rademakers J, Maindal HT, Steinsbekk A, Gensichen J, Brenk-Franz K, Hendriks M. Patient activation in Europe: an international comparison of psychometric properties and patients' scores on the short form Patient Activation Measure (PAM-13). BMC Health Serv

Res. 2016 Oct 12;16(1):570. doi: 10.1186/s12913-016-1828-1. PMID: 27729079; PMCID: PMC5059995.

30. Schmaderer M, Pozehl B, Hertzog M, & Zimmerman L. Psychometric Properties of the Patient Activation Measure in Multimorbid Hospitalized Patients. *Journal of nursing measurement*, 2015, *23*(3), 128–141. https://doi.org/10.1891/1061-3749.23.3.E128
31. Skolasky RL, Green AF, Scharfstein D, Boult C, Reider L, Wegener ST. Psychometric properties of the patient activation measure among multimorbid older adults. Health Serv Res. 2011 Apr;46(2):457-78. doi: 10.1111/j.1475-6773.2010.01210.x. Epub 2010 Nov 19. PMID: 21091470; PMCID: PMC3064914.
32. Kosar C, Besen DB. Adaptation of a patient activation measure (PAM) into Turkish: reliability and validity test. Afr Health Sci. 2019 Mar;19(1):1811-1820. doi: 10.4314/ahs.v19i1.58. PMID: 31149012; PMCID: PMC6531939.
33. Zeng H, Jiang R, Zhou M, Wu L, Tian B, Zhang Y, Qu F. Measuring patient activation in Chinese patients with hypertension and/or diabetes: reliability and validity of the PAM13. J Int Med Res. 2019 Dec;47(12):5967-5976. doi: 10.1177/0300060519868327. Epub 2019 Oct 10. PMID: 31601130; PMCID: PMC7045661.
34. Moreno-Chico C, González-de Paz L, Monforte-Royo C, Arrighi E, Navarro-Rubio MD, Gallart Fernández-Puebla A. Adaptation to European Spanish and psychometric properties of the Patient Activation Measure 13 in patients with chronic diseases. Fam Pract. 2017 Sep 1;34(5):627-634. doi: 10.1093/fampra/cmx022. PMID: 28379415.
35. Graffigna G, Barello S, Bonanomi A, Lozza E, Hibbard J. Measuring patient activation in Italy: Translation, adaptation and validation of the Italian version of the patient activation measure 13 (PAM13-I). BMC Med Inform Decis Mak. 2015 Dec 23;15:109. doi: 10.1186/s12911-015-0232-9. PMID: 26699852; PMCID: PMC4690217.

36. Cunha CM, da Cunha D, Manzato RO, Nepomuceno E, da Silva D, & Dantas R. Validation of the Brazilian Version of the Patient Activation Measure 13. *Journal of nursing measurement*, 2019, *27*(1), 97–113. https://doi.org/10.1891/1061-3749.27.1.97

37. Hibbard JH, Stockard J, Mahoney ER, Tusler M. Development of the Patient Activation Measure (PAM): conceptualizing and measuring activation in patients and consumers. Health Serv Res. 2004 Aug;39(4 Pt 1):1005-26. doi: 10.1111/j.1475-6773.2004.00269.x. PMID: 15230939; PMCID: PMC1361049.

38. Wensing M, van Lieshout J, Jung HP. *et al.* The Patients Assessment Chronic Illness Care (PACIC) questionnaire in The Netherlands: a validation study in rural general practice. *BMC Health Serv Res* **8,** 182 (2008). https://doi.org/10.1186/1472-6963-8-182

39. Glasgow RE, Wagner EH, Schaefer J, Mahoney LD, Reid RJ, Greene SM. Development and validation of the Patient Assessment of Chronic Illness Care (PACIC). Med Care. 2005 May;43(5):436-44. doi: 10.1097/01.mlr.0000160375.47920.8c. PMID: 15838407.

40. Berg SK, Færch J, Cromhout PF, Tewes M, Pedersen PU, Rasmussen TB, Missel M, Christensen J, Juel K, & Christensen AV. Questionnaire measuring patient participation in health care: Scale development and psychometric evaluation. *European journal of cardiovascular nursing: journal of the Working Group on Cardiovascular Nursing of the European Society of Cardiology*,2020, *19*(7), 600–608 https://doi.org/10.1177/1474515120913809

41. Serrani Azcurra DJ. Elders Health Empowerment Scale: Spanish adaptation and psychometric analysis. *Colombia medica (Cali, Colombia)*, 2014, *45*(4), 179–185.

42. Small N, Bower P, Chew-Graham CA, Whalley D, Protheroe J. Patient empowerment in long-term conditions: development and preliminary testing of a new measure. BMC Health Serv Res. 2013 Jul 8;13:263. doi: 10.1186/1472-6963-13-263. PMID: 23835131; PMCID: PMC3725177.

43. Busse R, Blumel M, Scheller-Kreinsen D, Zentner A. Tackling chronic diseases in Europe. Available from: https://www.euro.who.int/__data/assets/pdf_file/0008/96632/E93736.pdf

44. Yen HY, Lin LJ. Quality of life in older adults: Benefits from the productive engagement in physical activity. J Exerc Sci Fit. 2018 Aug;16(2):49-54. doi: 10.1016/j.jesf.2018.06.001. Epub 2018 Jun 7. PMID: 30662493; PMCID: PMC6323155.

45. Søgaard MB, Andresen K, Kristiansen M. Systematic review of patient-engagement interventions: potentials for enhancing person-centred care for older patients with multimorbidity. BMJ Open. 2021 Dec 16;11(12):e048558. doi: 10.1136/bmjopen-2020-048558. PMID: 34916309; PMCID: PMC8679112.

46. Jerofke-Owen T, Garnier-Villarreal M, Fial A, Tobiano G. Systematic review of psychometric properties of instruments measuring patient preferences for engagement in health care. J Adv Nurs. 2020 Apr 30. doi: 10.1111/jan.14402. Epub ahead of print. PMID: 32350898.

47. Lip, G. Y., Kamath, S., Jafri, M., Mohammed, A., & Bareford, D. (2002). Ethnic differences in patient perceptions of atrial fibrillation and anticoagulation therapy: the West Birmingham Atrial Fibrillation Project. Stroke, 33(1), 238-242.

48. Hillier, S. (1991). The health and health care of ethnic minority groups. Sociology as applied to medicine, 146-159

Figure caption

Figure 1. PRISMA flow diagram of the studies' selection