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# BMJ Open Health-related preferences of older patients with multimorbidity: an evidence map

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#### ABSTRACT

**Objectives** To systematically identify knowledge clusters and research gaps in the health-related preferences of older patients with multimorbidity by mapping current evidence.

**Design** Evidence map (systematic review variant). Data sources MEDLINE, EMBASE, PsycINFO, PSYNDEX, CINAHL and Science Citation Index/Social Science Citation Index/-Expanded from inception to April 2018.

Study selection Studies reporting primary research on health-related preferences of older patients (mean age ≥60 years) with multimorbidity (≥2 chronic/acute conditions).

Data extraction Two independent reviewers assessed studies for eligibility, extracted data and clustered the studies using MAXQDA-18 content analysis software. Results The 152 included studies (62% from North America, 28% from Europe) comprised 57 093 patients overall (range 9-9105). All used an observational design except for one interventional study: 63 (41%) were qualitative (59 cross-sectional, 4 longitudinal), 85 (57%) quantitative (63 cross-sectional, 22 longitudinal) and 3 (2%) used mixed methods. The setting was specialised care in 85 (56%) and primary care in 54 (36%) studies. We identified seven clusters of studies on preferences: end-of-life care (n=51, 34%), self-management (n=34, 22%), treatment (n=32, 21%), involvement in shared decision making (n=25, 17%), health outcome prioritisation/goal setting (n=19, 13%), healthcare service (n=12, 8%) and screening/ diagnostic testing (n=1, 1%). Terminology (eg, preferences, views and perspectives) and concepts (eg. trade-offs, decision regret, goal setting) used to describe health-related preferences varied substantially between studies. Conclusion Our study provides the first evidence map on the preferences of older patients with multimorbidity. Included studies were mostly conducted in developed countries and covered a broad range of issues. Evidence on patient preferences concerning decision-making on screening and diagnostic testing was scarce. Differences in employed terminology, decision-making components

and concepts, as well as the sparsity of intervention studies, are challenges for future research into evidencebased decision support seeking to elicit the preferences of older patients with multimorbidity and help them construct preferences.

## Strengths and limitations of this study

- This evidence map presents a systematic overview of studies addressing a variety of health-related preferences in older patients with multimorbidity.
- We identified clusters of studies on, for example, health outcome prioritisation and end-of-life care preferences; few studies addressed preferencesensitive decisions on screening and diagnostic
- The terminology and concepts used to address health-related preferences varied considerably in the included studies, highlighting a need for more standardisation to improve further research.

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## INTRODUCTION

Multimorbidity, defined as the co-occurrence of multiple medical conditions in a person, is a growing public health concern that affects approximately two-thirds of people over the age of 60 years.<sup>2 3</sup> Patients with multimorbidity generally experience a higher burden of disease, physical disabilities, adverse drug reactions, more frequent hospital admissions, reduced quality of life and increased mortality compared with those with a single condition. 45 As patients face new and growing demands to organise and coordinate their own care to comply with treatment regimens, multiple chronic conditions are often associated with high treatment burden in addition to the burden of the diseases themselves.<sup>6</sup> If patients are overwhelmed by the burden, they limit their compliance to their preferred tasks. Moreover, the care of patients with multimorbidity is challenging, as treatments for one condition may adversely affect another.<sup>8</sup> Robust evidence supporting





decision-making in these patients is scarce,<sup>2</sup> and the use of multiple disease-based guidelines is inappropriate, as they do not adequately consider potentially interacting conditions and treatments.<sup>910</sup>

The delivery of healthcare in patients with multimorbidity requires a patient-centred approach, that is 'respectful of and responsive to individual patient preferences, needs and values, and ensuring that patient values guide all clinical decisions'. 11 The 'Ariadne principles' 12 stress the importance of physicians and patients sharing realistic treatment goals, and of individualising management and follow-up by taking patients' preferences into consideration when making clinical decisions. Recent clinical guidelines on multimorbidity have embraced this approach and emphasise the incorporation of patients' preferences in clinical decision-making, for example in the selection of appropriate self-management activities and treatment options, as well as in the prioritisation of health outcomes. 13 Similarly, the consideration of patients' views in the form of patient-reported experiences and care outcomes have been recognised as critical to the achievement of high-performing health systems that are responsive to the needs of people with multimorbidity.14

It remains unclear how health-related preferences can be elicited from older patients with multimorbidity, as patients may be unfamiliar with the decision elements. 15 Moreover, concerns have been raised that patients are often provided with too little information about the benefits and harms of a treatment, 16 17 may find it difficult to prioritise health outcomes and make trade-offs, and in consequence, may refrain from participating in the decision making process. 18 As evidence maps allow a systematic approach to be used to collate evidence on a broad topic, we used this emerging method to map the health-related preferences of older patients with multimorbidity. 19 In particular, we aimed to (1) systematically identify and describe key characteristics of research on health-related preferences of older patients with multimorbidity, (2) display the landscape of existing research in visual formats, (3) identify evidence clusters to guide any subsequent knowledge synthesis (systematic reviews and meta-analysis) and (4) identify evidence gaps and encourage relevant stakeholders and funding agencies to prioritise these in future research.

#### **METHODS**

## Reporting protocol and guideline

We described the methods in a study protocol<sup>20</sup> that has since been subject to no amendments, registered the evidence map in Open Science Framework (OSF, DOI 10.17605/OSF.IO/MCRWQ) and adhered to the Preferred Reporting Items for Systematic Reviews and Meta-Analyses Extension for Scoping Reviews checklist<sup>21</sup> where possible (see online supplementary table S1).

## **Systematic literature search**

We searched the electronic databases MEDLINE and EMBASE (via Wolters Kluwer's search interface Ovid), PsycINFO, PSYNDEX and CINAHL (via EBSCOhost) and Social Science Citation Index and Science Citation Index Expanded (via Web of Science from Clarivate Analytics) from inception until April 2018. In our search, we combined medical subject headings with keywords covering old age, multimorbidity, polypharmacy and search terms related to patient preferences. The search strategy was adapted to suit the database under review (see online supplementary table S2 on Search strategy in MEDLINE—Ovid).

Based on the 32 most relevant studies identified in our initial search (ie, when keywords provided by the author contained the terms 'multimorbidity' and 'patient preferences' or 'patient priorities' and/or described a specific method for eliciting patients' preferences, such as 'conjoint analysis'), we conducted a cited reference search (forward citation tracking) using the Web of Science Core Collection. We also checked the reference lists of included studies, the reference lists of systematic reviews on related topics for further studies (hand search) and contacted the authors of conference proceedings that had not published a full set of results. We searched for ongoing trials in the Register for Clinical Trials<sup>22</sup> and the WHO International Clinical Trials Registry.<sup>23</sup>

## Inclusion and exclusion criteria

We included qualitative and quantitative studies involving older patients of 60 years and older with multimorbidity (two or more simultaneous chronic or acute conditions<sup>1</sup>) that addressed health-related patient preferences. We also included studies involving older patients with chronic conditions that are frequently associated with multimorbidity, even if they were not reported in detail (chronic heart failure, chronic obstructive pulmonary disease (COPD), chronic kidney disease, advanced cancer and frailty). <sup>24–26</sup>

We excluded studies investigating preferences relating to interventions of limited availability or whose legal status was unclear (eg, euthanasia, which is not legal or available in most countries), studies addressing the preferences of caregivers, family or medical and/or other professionals as well as case reports, narrative reviews and editorials. We did not apply any restrictions to the geographical location of the study or language of publication.

## **Study selection**

Two reviewers (AIG and JN) screened the titles and abstracts of all references identified by electronic searches. Before screening, stepwise calibration was performed on a sample of 50 studies, with the aim of achieving 80% agreement between the two reviewers.<sup>20</sup> If 80% agreement had not been reached, our inclusion and exclusion criteria would have been refined to reach this cut-off. The new criteria would then have required further calibration using a new sample of 50 studies until the threshold was



reached. We also obtained full texts of potentially relevant articles, and two reviewers (AIG, JN or CS) independently assessed these for inclusion. Conflicts were resolved by discussion among reviewers.

## **Mapping the evidence**

## **Data extraction**

Following the calibration of five full text articles, two reviewers (AIG, JN or CS) independently extracted data on (1) study characteristics including study design (observational (qualitative, quantitative or mixed-methods, cross-sectional or longitudinal) and interventional), geographical area, study setting (eg, primary care), sample size, (2) study aim, (3) patient population (eg, definition of multimorbidity, age, sex) and (4) characteristics of preferences, such as methods used to elucidate patients' preferences, and definition of preferences according to the authors.

## Types of preference

We conducted qualitative relational content analysis<sup>27</sup> to derive overarching themes. The analysis was based on coding by two independent reviewers (AIG, JN or CS) using MAXODA-18, which were further scrutinised by CM, JWB, MvdA, TSN and MSB.<sup>20</sup> The initial step was to scrutinise title and abstract (focusing on the study aim) of the included studies to gain a general understanding of what the study was about. The full text was then read and re-read and codes assigned (eg, resuscitation preferences<sup>28</sup>), which were later grouped according to overarching themes (eg, life-sustaining treatment preferences).<sup>27</sup> <sup>28</sup> Reviewers' categorisation of preference types was partly based on a previous classification (ie, end-of-life preferences, prioritisation of health problems, prioritisation of medication, preferences regarding the role played in decision-making, preferences in surgical treatment methods, prioritisation of treatment goals, determinants of preference, changes in preferences and preferences concerning the organisation of healthcare) obtained from a pilot study (published elsewhere) of the evidence map.

## **Mapping**

We tabulated the identified studies, summarised study and patient characteristics, as well as study publications per year, and used bubble plots to display evidence clusters in terms of preference type and study characteristics.

#### Patient and public involvement

A patient representative (KR) from the Federal Joint Committee 'Gemeinsamer Bundesausschuss (G-BA)' was involved in the conception and development of the evidence map, in the interpretation of the findings, and in writing the manuscript. KR has considerable expertise in evidence-based medicine in a healthcare context, and an understanding of the pivotal role of patients' preferences in the provision of effective healthcare.

## **RESULTS**

## **Literature search and selection process**

Among the 9145 unique screened references, 152 studies (comprising over 57 000 patients) were included in the evidence map. As 80% agreement between the two reviewers was achieved in the first calibration exercise, inclusion and exclusion criteria remained unchanged. We contacted 48 authors of conference papers (13% answered) and included one further study that had already been identified in our electronic search (figure 1). Online supplementary tables S3a-3f show key characteristics of the included studies. Online supplementary table S4 presents excluded studies and reasons for exclusion.

## Key characteristics of the included studies and participants

Of the included studies, all but one were observational (151/152), and nearly all were conducted in developed countries (147/152) (table 1, online supplementary tables S3a-S3f) and published in 2007 or thereafter (128/152) (see online supplementary figure S1). All studies were written in English.

The sample size ranged from 9 to 9105 patients and captured both sexes (51% female). The mean age of participants ranged from 60 to 85 years. Eight studies<sup>29–36</sup> included different age groups but only data from patients aged 60 years and older were included in the evidence map. Three studies<sup>36–38</sup> provided no age estimate but were included because they clarified that they had only included older patients. In 87 of the included studies, patients with multimorbidity (no index disease defined) or comorbidity (index disease defined plus at least one other associated condition) were investigated, and in the remaining 65 studies, patients with conditions known to be highly associated with multimorbidity were included.

## Types of preference and evidence clusters

Content analysis<sup>27</sup> enabled us to identify seven major types of preference (table 2). We assigned 130 studies (85 %) to one of these types of preference and 22 (15 %) studies<sup>37 39–57</sup> to two types of preference. Terminology (eg, preferences, views and perspectives) and concepts (eg, trade-offs, decision regret and goal setting) varied substantially among studies.

## End-of-life care preferences

The largest evidence cluster comprised the 51 studies (34 %) addressing end-of-life care preferences, most of which were in specialised care settings (41/51 studies) (figure 2). Content analysis of this preference revealed that advance care planning, 42 in which multimorbid patients with advanced chronic diseases were asked how they would like to be cared for in the final months of their lives, was the main theme (table 2). The most common theme within this cluster concerned preferences for specific life-sustaining treatments (29/51), 28 32-34 36 58-81 such as cardiopulmonary resuscitation or mechanical ventilation. Additional topics in this cluster addressed

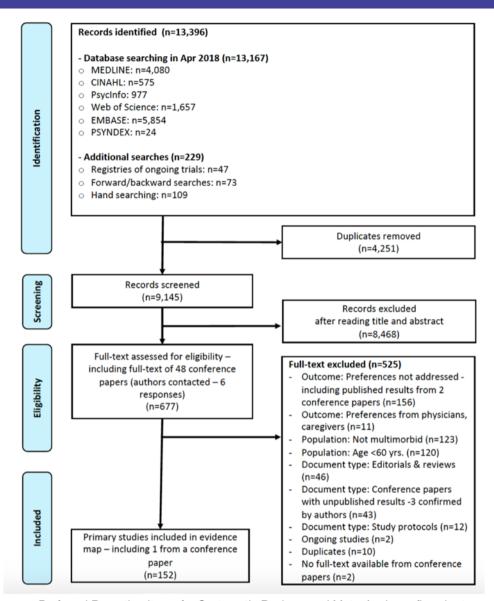


Figure 1 Evidence map Preferred Reporting Items for Systematic Reviews and Meta-Analyses flowchart.

themes such as the preferred place of death (eg, home vs hospice).  $^{60}$ 

## Self-management preferences

The second largest evidence cluster included 34 studies (22 %) and addressed patients' self-management priorities, defined as activities that an individual undertakes to maintain or reduce the effect of a disease(s) on their health status.<sup>82</sup> Most studies about self-management preferences were conducted in primary care (21/34). The only intervention study in the evidence map (113) used a cluster-randomised design to evaluate whether structured priority-setting consultations led to a sustainable reconciliation of diverging physician-patient views on the importance of health problems. Overall, content analysis of this evidence cluster revealed five key themes: (1) patients' prioritisation of their multiple health problems (20/34),  $^{30.45.47.54.83-98}$  as an example of which patients were asked how they prioritised their osteoarthritis over their other conditions, 97 (2) patients' preferences regarding

self-management of their medications  $(8/34)^{42 \cdot 43 \cdot 47 \cdot 54 \cdot 99 - 102}$  and, for instance, its association with treatment adherence,  $^{42}$  (3) patients' self-care behaviours  $(3/34)^{44 \cdot 55 \cdot 103}$  aimed at accomplishing their life goals,  $^{44}$  (4) characteristics of eHealth support tools  $(2/34)^{30 \cdot 104}$  to help patients self-manage their multiple health conditions  $^{104}$  and (5) changes in patients' choices resulting from changing circumstances (2/34).

## Treatment preferences

Thirty-two studies (22 %) investigated a variety of treatment preferences concerning (1) medication (13/32),<sup>37</sup> <sup>41-43</sup> <sup>47</sup> <sup>49</sup> <sup>53</sup> <sup>54</sup> <sup>105-108</sup> perhaps for a specific blood pressure-lowering drug due to its characteristics (eg, effects and dose schedule),<sup>106</sup> (2) dialysis as a treatment option in end-stage renal disease (6/32),<sup>48</sup> <sup>109-113</sup> (3) surgery (4/32),<sup>114-117</sup> such as a decision in favour of implantable cardioverter-defibrillators or joint replacement, (4) chemotherapy (5/32),<sup>35</sup> <sup>57</sup> <sup>118-120</sup> for which studies may have examined preferences in adjuvant cancer



Table 1 Descriptive summary of included	studies
Variable	Total-n (%)
Study characteristics	
Geographical location	
North America	94 (62%)
Europe	43 (28%)
Australia and New Zealand	10 (7%)
Asia	5 (3%)
Setting	
Primary care	54 (36%)
Outpatient specialised	59 (39%)
Hospital (inpatient and emergency)	26 (17%)
Nursing homes	5 (3%)
Interdisciplinary	8 (5%)
Study design/method	
Qualitative (observational)	63 (42%)
Cross-sectional (observational)	59 (39%)
Longitudinal (observational)	4 (6%)
Quantitative	86 (57%)
Cross-sectional (observational)	63 (41%)
Longitudinal (observational)	22 (15%)
Interventional	1 (1%)
Mixed methods (qualitative and quantitative)	3 (2%)
Observational (total)	151 (99%)
Interventional (total)	1 (1%)
Sample size—median (range)	83 (9–9105)
Observational	
Qualitative	30 (9–160)
Quantitative	196 (11–9105)
Mixed methods	50 (32-60)
Interventional	317
Patients' characteristics	
Type of condition	
Studies describing multimorbid patients*	58 (38%)
Studies describing patients with an index disease and comorbidity	29 (19%)
Diabetes	7 (5%)
Hypertension	5 (3%)
Depression/mental illness	4 (3%)
Cardiovascular disease	4 (3%)
Osteoarthritis	3 (2%)
Other	6 (4%)
Studies describing patients with chronic conditions often associated with multimorbidity	65 (43%)
Chronic heart failure	10 (7%)
Advanced cancer	16 (11%)
	Continued

Table 1 Continued	
Variable	Total-n (%)
Chronic kidney disease	15 (10%)
COPD	4 (3%)
Mixed (heart failure, COPD)	20 (13%)
Age (range)†	60–85
Sex (% female)†	28 905 (51%)

<sup>\*</sup>No further details of included conditions were reported in the majority of studies.

treatments and (5) non-pharmacological/conservative interventions (3/32), 121-123 such as studies exploring preferences for activity interventions. 121

## Involvement in the shared decision making process

Twenty-five (17 %) studies explored how patients preferred to be involved in the shared decision making process. Studies in this cluster investigated preferred (1) patterns of engagement (21/25),  $^{37\ 41\ 46\ 48-52\ 57\ 124-133}$  (2) information (4/25),  $^{39\ 52\ 128\ 134}$  (3) communication with providers  $(1/25)^{40}$  and (4) patient decision aids  $(1/25)^{.56}$ 

#### Healthcare service preferences

Twelve studies (8%) focused on preferences for certain healthcare services, and specifically (1) preferred care processes (10/12), <sup>45</sup> <sup>135–143</sup> such as continuity of care, accessibility and acceptance of the substitution of a physician by nurses and (2) service models (2/12), <sup>31</sup> <sup>144</sup> perhaps asking patients about their preferences regarding Chronic Care Model recommendations. <sup>31</sup>

## Health outcome prioritisation and goal setting

Nineteen studies (13%) investigated health outcome prioritisation and goal setting. These may have been (1) patients' holistic goals for their lives or with respect to their various diseases (6/19),  $^{44}$   $^{55}$   $^{145-148}$  (2) health outcome prioritisation  $(10/19)^{53}$   $^{149-157}$ —one study in particular addressed the tools patients preferred to use to prioritise health outcomes  $^{151}$  and (3) collaborative goal setting among patients, physicians and caregivers (3/19).  $^{103}$   $^{158}$   $^{159}$ 

## Screening and diagnostic tests

One study<sup>160</sup> investigated cancer screening preferences among patients with multimorbidity.

## **DISCUSSION AND CONCLUSION**

This work provides a systematic overview of research on health-related preferences of older patients with multimorbidity.

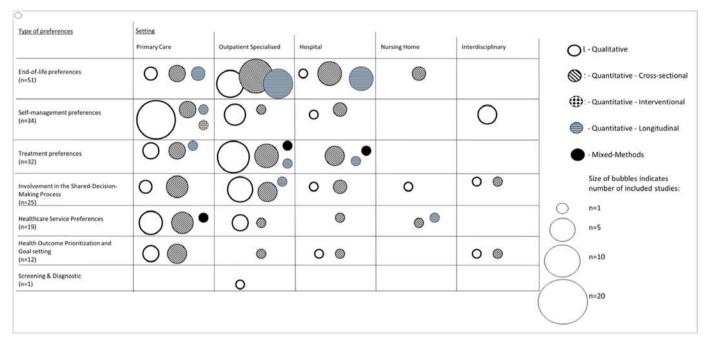
<sup>†</sup>Studies with overlapping population were excluded (n=10). <sup>36</sup> 58 59 83 84 105 109 149 161 169

COPD, chronic obstructive pulmonary disease.



Table 2 Description of	Description of the type of preferences investigated in the included studies			
Types of preferences	Definition	Themes	No. of studies	References
End-of-life care preferences	Treatment preferences for resuscitation and critical care. Covers all aspects	Advanced care planning	51	28 32–34 36 39 46 50–52 56 58–81 150 161 169–182
	relating to anticipatory decision-making such as advance directives.	- Life-sustaining treatment preferences	29	28 32-34 36 58-81
Self-management preferences	Preferences related to the ongoing activities that an individual undertakes to	eHealth support	2	30 104
	maintain or reduce the effect of a disease/s on his or her health status. It includes how and under what circumstances, patients prioritise conditions and	Prioritisation of health problems	20	30 45 47 54 83-98
	adjust self-management practices, how priorities might change over time, and	Medication self-management	80	42 43 47 54 99–102
	TOW LIEST ALE CISCUSSED WITH THE THE PROPOSICIALS.	Self-care behaviours	3	44 55 103
		Revisiting choices	2	29 30
Treatment preferences	Preferences that involve a discrete set of effective treatment options (eg. radical	Medication	13	37 41-43 47 49 53 54 105-108
	mastectomy vs lumpectomy with radiation for localised breast cancer). The treatment options can include any intervention with a therapeutic aim.	Dialysis	9	48 109–113
		Surgery	4	114–117
		Chemotherapy	5	35 57 118–120
		Non-pharmacological/conservative	က	121–123
		Medication device	-	183
Involvement in the shared	Preferences regarding the degree of involvement in discussions with health	Patterns of engagement	21	37 41 46 48–52 57 124–133
decision making process preferences	professionals about the options for treatment, the benefits and harms of each therapy, and making collaborative decisions about how to proceed.	Patient decision aid	-	56
		Information	4	39 52 128 134
		Communication with providers	-	40
Healthcare service preferences		Processes of care	10	45 135–143
	services the health system provides.	- Site of care	2	138 142
		- Type of social support	-	139
		- Type of caregiver/provider	က	135 138 143
		- Continuity and access	2	45 135–137
		- Guiding principles	-	141
		Service models	2	31 144
		- Chronic care model	-	31
		- Cardiac rehabilitation	-	144
Health outcome prioritisation	Preferences regarding personal health and life outcomes (eg, function, social	Life and health goals	9	44 55 145–148
allo goal settilig	activities and symptom renet) that people hope to achieve unlough their realisticate. Health outcome goals that patients prioritise within the context of their care preferences.	Health outcome prioritisation	10	53 149–155
		- Preferred tools	-	151
		Collaborative goal setting	ဇ	103 158 159
		<ul> <li>Patient, physician, caregiver agreement</li> </ul>	-	158
Screening and diagnostic tests		Screening test	-	160
preferences	diagnostic test.	- Cancer screening	-	160





**Figure 2** Types of preference by setting and study design. Circle size represents the number of studies; pattern coding represents the study design. \*The bubble plot displays more than the total number of included studies (n=174 vs n=152) because 22 studies were assigned to two different types of preference.

#### **Evidence clusters**

We identified 152 studies, most of which were published within the last decade and conducted in developed countries. The vast majority of studies included in the evidence map used a qualitative or cross-sectional quantitative design (126/152).

Our clustering approach revealed that studies of patient preference focused on seven areas: end-of life care, self-management, treatment, involvement in shared decision making, health outcome prioritisation/goal setting, healthcare service delivery and screening/diagnostic testing. The size of the evidence clusters varied widely (from 1 to 51 studies) and the research objectives and settings differed considerably.

The largest and most homogenous cluster was of end-oflife preferences  $(51/152 \, \text{studies})$  and was largely confined to specialised care (41/51). Furthermore, the study objectives revealed one overarching theme (advance care planning) and were relatively uniform compared with the other clusters.

Self-management and treatment preferences were the second (34/51) and third (32/51) largest clusters respectively. Although studies about self-management preferences were relatively homogeneous in terms of study setting (they were mostly conducted in primary care (21/34)), we found considerable variability in the overarching themes. Treatment preferences were rather heterogeneous, with the cluster containing a variety of settings and themes.

Overall, we identified clusters of evidence. However, as evidence maps do not permit the critical appraisal of the robustness of evidence, the evidence clusters (ie, studies) still require verification. <sup>19</sup>

## **Evidence gaps**

Longitudinal studies were rare and the few that did observe changes in preference over time generally concerned end-of-life care preferences. <sup>33</sup> <sup>59</sup> <sup>63</sup> <sup>64</sup> <sup>66</sup> <sup>161</sup> The only intervention study we identified <sup>93</sup> highlighted the fragility of prioritisation processes over time, and showed that health priorities shared by patients and physicians were often not sustainable 2 weeks after an intervention. Preferences tend to change when chronic conditions worsen, <sup>33</sup> <sup>59</sup> <sup>63</sup> <sup>64</sup> <sup>66</sup> <sup>161</sup> additional diagnoses are made that lead patients to prioritise a new condition over existing ones, <sup>30</sup> or new information about treatment options is obtained. <sup>37</sup> However, although crucial in clinical decision making, it is unclear how and why patient preferences change significantly over time. High quality longitudinal studies are needed to help physicians deal with changing preferences and to reassess preference-sensitive decisions.

We identified a further research gap in a lack of studies in older patients with multimorbidity that test the effectiveness (1) of interventions using different methods to elicit/construct preferences, and (2) of (complex) interventions that proactively consider patient preferences among patient-relevant outcomes.

The smallest cluster (containing only one study) concerned the preferences of older patients with multimorbidity with respect to screening or diagnostic tests. <sup>160</sup> This finding is surprising, as the additional health-related burden of screening and diagnostic tests can be substantial, and it is well-known that the risk-benefit ratio of such tests can be highly preference-sensitive. <sup>13</sup>

It is worthy of note that end-of-life care preferences were mostly assessed in specialised ambulatory care. As palliative care is a core task in primary care, we would



have expected more studies to address such end-of-life preferences in this setting. <sup>162</sup>

## **Comparison with other studies**

This is the first evidence map of health-related preferences in older patients with multimorbidity. Although previously published evidence summaries, such as scoping or systematic reviews, partially addressed specific topics relating to some of the clusters identified in this evidence map, none focused on older patients with multimorbidity.

Four systematic reviews explored preferences in end-of-life care (as well as other preferences, such as involvement in shared decision making and goal setting): (1) Puts  $et\ al^{163\ 164}$  systematically reviewed factors influencing older adults' (not necessarily multimorbid) decision to accept or refuse cancer treatment, (2) de Decker  $et\ al^{165}$  confirmed an association of the wish not to be resuscitated with multimorbidity, (3) Singh  $et\ al^{166}$  conducted a meta-analysis on the roles cancer patients (not necessarily multimorbid) prefer to play in treatment decision-making and (4) Vermunt  $et\ al^{167}$  evaluated studies of the effects of interventions that support collaborative goal setting in elderly people with a chronic health condition or multimorbidity, including our only intervention study. 93

Most of the evidence summaries of health-related preferences focused on end-of-life care preferences, and specifically its determinants. Further research should concentrate on the clusters and gaps identified in our evidence map in order to enhance our understanding of the preferences of older patients with multimorbidity.

## **Strengths and limitations**

A major strength of our approach is that we used a sensitive strategy that combined controlled terms (ie, a defined vocabulary to index and retrieve information from the included electronic databases) and free-text searches in all relevant databases. Furthermore, we did not apply any restrictions to publication language, design or geographical location of the studies. Additionally, we searched for unpublished studies in registries and contacted authors of conference papers.

However, we addressed a broad topic with incomplete indexing (both, multimorbidity and patient preferences) and may have missed studies. In particular, we did not include search terms for specific measures of preference (eg, analytic hierarchy process, discrete choice experiment and conjoint analyses) in electronic searches, because test searches including them did not increase sensitivity. Furthermore, we did not search grey literature, as this approach would not have identified additional relevant studies that could have justified the enormous effort involved. <sup>168</sup>

Despite the experience gathered in the pilot study (published elsewhere), the use of a lower age limit of at least 60 years was difficult to operationalise, as studies often included a wide age range but did not always report separate results for older patients. When the age group was unclear, we did not include the study.

Furthermore, we used an iterative process to develop our evidence clusters and the identified clusters and their definitions were agreed on by all authors. However, inherent to the methods used, we cannot rule out some subjectivity.

## **CONCLUSIONS AND FURTHER RESEARCH OUTLOOK**

This evidence map provides the first systematic overview of empirical investigations concerning health-related preferences of older patients with multimorbidity. Their objectives addressed a broad range of relevant topics across all settings and used predominantly cross-sectional and observational qualitative and quantitative methods. Our evidence map also revealed gaps, both in general such as the scarcity of longitudinal studies to investigate changes in preferences over time, and of intervention studies, which, with one exception, 93 failed to develop and test interventions to support the construction of healthrelated preferences in this population. More specifically, we found a remarkably low number of studies addressing preferences concerning end-of-life care in a primary care setting, as well as preferences related to screening and diagnostic testing. Furthermore, the included studies varied considerably in terms of terminology (eg, preferences, priorities, views and perceptions) and decisionmaking components and concepts (eg, trade-offs, decision regret and goal setting). These require further elucidation.

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