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QUANTITATIVE PATIENT PREFERENCE EVIDENCE FOR HEALTH TECHNOLOGY ASSESSMENT: A CASE STUDY

Ann-Sylvia Brooker, Steven Carcone, William Witteman, Murray Krahn
THETA (Toronto Health Economics and Technology Assessment) collaborative

Objectives: We conducted a systematic review of quantitative research regarding patients' preferences, perspectives and values for ventilation among chronic obstructive pulmonary disease (COPD) patients. Our objective was to explore the feasibility and desirability of incorporating patient preferences within the health technology assessment (HTA) process by working through a case study.
Methods: Medical and economic databases were searched for studies published in English from 1990 through March 4, 2011. Studies were selected based on title and abstract. Due to the heterogeneity of the studies, data were analyzed using a narrative synthesis approach.
Results: Among 1833 identified citations, twelve studies met our inclusion criteria. Ten of these studies pertained to COPD patient preferences for ventilation. Results indicate that a significant proportion of COPD patients are willing to forgo ventilation, particularly when it is expressed as "indefinite life support" (60–78 percent) rather than as temporary modality. Results indicate that patient preferences for mechanical or noninvasive ventilation cannot be predicted by covariates (e.g., age, quality of life) or by others who are frequently called upon to make decisions are their behalf.
Conclusions: We found that it is indeed feasible to conduct a systematic review of quantitative preference-related evidence for an HTA topic. However, the process of conducting this preference-related case study also revealed several challenges because there is a high degree of variation in taxonomy, instrumentation, and study design. Therefore, we do not recommend it as a routine part of the HTA process, but we suggest that it is a promising area to pursue for preference-sensitive technological decisions.

Keywords: Health technology, Patient preference, COPD, Ventilation, Review

In the past decade, the role of patient values and preferences within health technology assessment (HTA) has received increased attention. HTA has been defined as multidisciplinary policy analysis that investigates the intended and unintended consequences of using a health technology (health intervention) (1). Despite this broad mandate, some observers have suggested that the standard methods of assessing technologies place too great an emphasis on clinical outcomes and societal perspectives, and that patient perspectives are not adequately addressed (1–4). The HTA process has been influenced by evidence-based medicine, in which research that uses cost effectiveness analysis (CEA) and randomized controlled trials (RCTs) are privileged (1;3). Yet, these types of analyses often fail to capture the complexity of patient perspectives (3). CEAs often use utility-based measures such as quality-adjusted life-years (QALYs). Utilities are a value-based measure; but, given that these utilities are generally derived from random samples of the general population, one could argue that they are more adept at reflecting social values, rather than patient values. RCTs assess outcomes that are often defined by the scientific and medical community, rather than by patients (3;5). The recently created Patient-Centered Outcomes Research Institute (PCORI) in the United States is trying to reverse this trend, by engaging patients in the process of research and funding research which explores outcomes of interest to patients. However, it is still to be determined whether one institute with a limited funding supply can have sustained and widespread impact across a range of clinical research areas.

To address patients' perspectives within the HTA process, two main processes have been suggested. First, many have suggested the need for greater involvement of patients in the HTA process, such as through participation in HTA groups that help define the research questions, evaluate evidence and communicate findings (1;6). The rationale is that patients have unique knowledge and perspectives that can usefully inform the process, and furthermore, that those who are directly affected by these policies (i.e. patients), should be involved in shaping them (1). These types of opportunities are increasingly being considered by HTA agencies, although observers note that this process is often neglected (1;6). The second suggestion is to systematically gather and assess scientific evidence on patient perspectives as part of the HTA process (1). Systematically gathering

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such research evidence can provide a more representative view of patients' perspectives than the perhaps unusually vocal patients who choose to be directly involved in the HTA process (2). However, a recent international study found that only 38 percent of full HTA reports included any organizational and/or patient related issue (7). Although in Denmark, the proportion is much higher, as 88 percent of HTA reports include "patient aspects" (8). The international study found that when patient aspects are included in HTA reports, the decision about which of these aspects to explore is often quite arbitrary; methods are often not reported, and the research questions were presented in only a minority of cases (7).

Several authors have suggested that a systematic review of qualitative research can provide important insights into patients' preferences, perspectives, needs, and experiences and that these should be brought into the HTA process (8;9). We agree with this assertion; however, we also believe that there is substantial research on preferences, attitudes and perspectives that use quantitative research methods that can also to be explored.

Many of the terms pertaining to patients' perspective are inter-related, and are sometimes used interchangeably. Generally, authors have used the terms "patient perspectives" or "patient aspects" as umbrella terms. Thus, "patient perspectives" has been used to refer to patients' experiences, attitudes, and, beliefs, values and expectations about health, illness, and health technologies (1). "Patient aspects" has been used to refer to patients' perspectives, needs, experiences, preferences and acceptance of a technology (8). For this study, we will use the term "patient perspectives" as an umbrella term, to indicate all of the aforementioned categories (patients' experiences, attitudes, beliefs, values etc.). We will use the word patient "preferences" in a more precise way to denote the patients' view of the desirability of a health-related outcome, process or treatment choice.

In this study, we chose to explore how quantitative, patient preference data could contribute to the HTA process. The international review of HTA reports identified only one report that used data from preference instruments (7). However, including patient preference data within an HTA report can indicate the general acceptability of a technology to patients, which can be useful for policy makers. Including preference data within an HTA report may also encourage readers to recognize that an optimal decision is not uniform across patients, but is determined by patients' values concerning treatments or outcomes. Given that many of these concepts are inter-related, we chose to explore all aspects of patients' perspectives that might have a bearing on the evaluation of a technology, not only patient preferences, but also patients' attitudes and values. To explore this topic, we chose to work through a case study. At the development stages of this project, the Ontario Health Technology Advisory Committee commissioned a series of systematic reviews on the topic of Chronic Obstructive Pulmonary Disease (COPD) and a series of interventions. In addition, another research group at McMaster University initiated a synthesis of the qualitative research associated with the experience of COPD patients (10). This provided us with an ideal opportunity to explore how a systematic review of the quantitative research on preference-related issues of COPD patients could compliment the typical HTA process.

We chose to further restrict the systematic review to an appropriate intervention. Patient preferences are most relevant in several situations, such as when interventions have only marginal benefits, or when interventions involve trade-offs with significant negative and positive consequences (11). In these situations, patient choices can vary considerably based on individual patient preferences for outcome or process (4). We selected ventilation treatment for COPD as an appropriate case study, because the survival rates for COPD patients who receive intubation and mechanical ventilation (MV) are fairly poor—approximately 70 percent of patients will come off ventilation and survive to hospital discharge (contrasted to a 95 percent chance of death for those who refuse ventilation) (12). In addition, the treatment is uncomfortable, as the presence of a ventilation tube impedes the patient from speaking and eating normally. Therefore, the decision to use MV is a preference-sensitive decision; patients will vary in terms of the value they place on prolonging life versus the value of avoiding an unpleasant procedure with only limited, long-term efficacy.

METHODS

General Objective

The general study objective was to explore the feasibility and desirability of conducting systematic reviews on patient perspectives, preferences and values on HTA topics. Specific objectives included: (i) to develop an appropriate search strategy with high sensitivity for finding quantitative research related to patient preferences; (ii) to summarize and synthesize the literature on patient preferences for ventilation among COPD patients; and (iii) to discuss the advantages and disadvantages of including quantitative, patient preference related data within HTAs based on the results of our case study.

Search Strategy

We searched MEDLINE, Cumulative Index to Nursing and Allied Health Literature (CINAHL), Cochrane, American Economic Association's electronic bibliography (EconLit), EMBASE, Health Economic Evaluations (HEED), and PsycINFO databases for relevant studies published in English from 1990 through March 4, 2011. We developed a comprehensive list of terms that relate to patient perspectives, satisfaction, preferences and values (see Supplementary Tables, which can be viewed online at www.journals.cambridge.org/thc2013119, for complete list). Search terms for COPD and for ventilation were developed by referring to systematic reviews of these topics (e.g., Cochrane systematic reviews). Next, we searched abstracts and set aside all relevant quantitative articles. The

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reference lists for these articles were scrutinized to ensure that we did not omit any relevant literature. This process was repeated until we had developed a search strategy with high sensitivity (see Supplementary Tables for complete list).

Inclusion and Exclusion Criteria

Studies published in English were selected if they met the following criteria: (i) study participants (or a subsample) had COPD; (ii) study interventions included ventilation, MV and/or NIV (noninvasive ventilation) for the treatment of COPD; (iii) patient preferences, perspectives, attitudes, etc., were assessed by means of structured questionnaires or medical records. Our initial search yielded many articles that measured "quality of life" (QoL) indicators. Although quality of life overlaps conceptually with patient preferences, we decided to exclude these studies because of tractability and so that the more neglected topics relevant to preferences, attitudes and values could be emphasized.

Selection of Studies and Data Extraction

Identified studies were selected based on title and abstract by two authors independently (A.B. and S.C.). Full articles were retrieved if a decision could not be made based on the abstracts. The same authors (A.B. and S.C.) independently extracted data from each study into tables (Tables 1 and 2). Any disagreements were resolved by discussion or consensus with a third party (M.K.).

Analysis

The heterogeneity among studies in terms of instruments and outcomes precluded the possibility of a meta-analytic strategy. Data were analyzed using narrative synthesis, appropriate when the studies are too dissimilar for meta-analysis. A narrative synthesis consists of organizing, describing, exploring, and interpreting study findings and attempting to find explanations for these findings (13).

RESULTS

Study Characteristics

Among 1833 identified citations, twelve studies were eligible for data abstraction (Figure 1). These studies took place in a broad range of settings (Table 1, column 2): community (two studies); hospital (seven studies); community and hospital (two studies); and a rehabilitation center (one study).

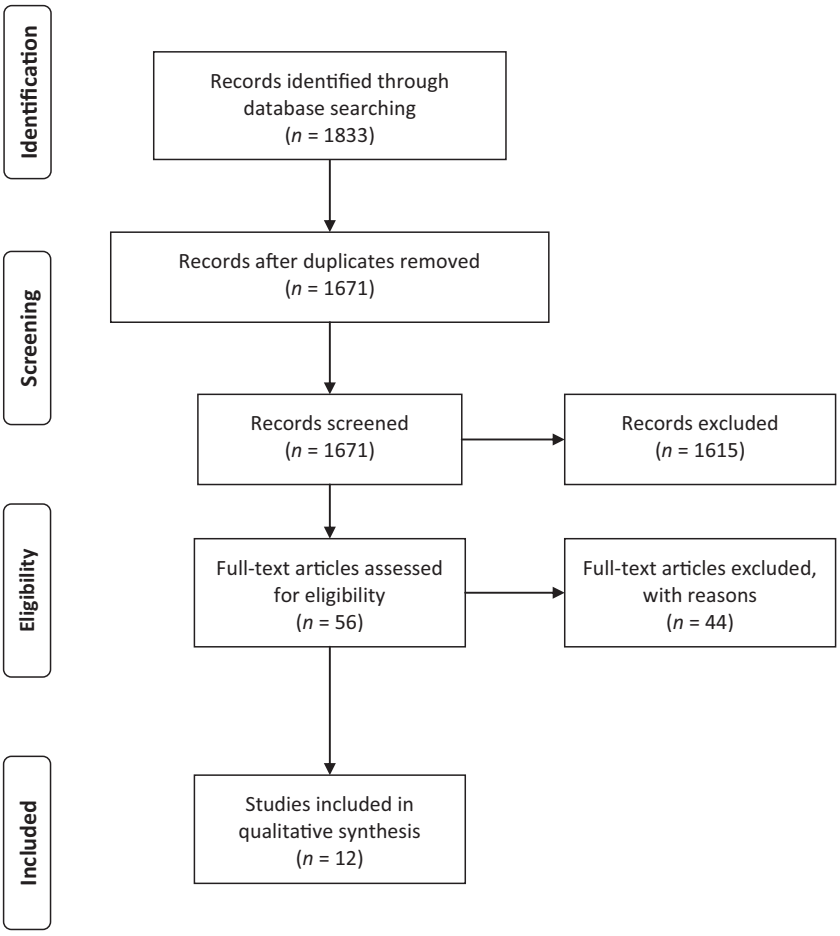


Figure 1. PRISMA flow diagram.

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Table 1. Summary of Study Methods

Article	Setting	Sample size of the COPD participants only	Outcome measures	Methods	Sample questions
1. Chakrabarti et al. (2009)	UK; hospital	50	Patient attitudes towards receiving IMV and NIV.	Five-stage structured interview by a study investigator (MD).	Willingness to receive NIV in an acute setting after being provided with a: verbal description (stage 1); photograph (stage 2); demonstration (stage 3); willingness to receive IMV after detailed explanation (stage 4); after explanation of alternatives (stage 5).
2. Claessens et al. (2000)	USA; multi-center study in 5 teaching hospitals	445 = patient only; 730 = surrogate substitution	Preference for having MV indefinitely was assessed by interview.	Interviews.	"Would you be very willing, somewhat willing, somewhat unwilling, very unwilling, or rather die than put up with (or continue to put up with) being attached to a ventilator or respirator all the time?"
3. Dales et al. (1999)	Canada; hospital	19	Preference for intubation and MV.	Scenario-based decision aid. Patients were interviewed on two occasions, with a second interview taking place a year later.	*I. To be intubated and receive MV:* ". . . there will be a 50% chance of coming off of the machine and living for one 1 year in a state of health that is no better or worse than before the attack which brought them to hospital" *II. Not to be intubated:* ". . . there will be a 100% chance of natural death."
4. Gaber et al. (2004)	UK; community	100	Stated preference for IMV, NIV or CPR.	Survey was conducted by two nurse specialists in patients' homes. A second survey (part 2) was mailed by post.	Patients were asked to imagine a hypothetical scenario in which "They were admitted to hospital with worsening of their condition . . . They were asked "on reaching that stage, would you wish to have NIV, IV or CPR?" "Do you think this sensitive issue should be discussed with all patients?" (part 2)
5. Goldstein et al. (1995)	Canada; community	9	Patient experience with use of Home MV.	Standardized format questionnaire conducted by a research assistant.	1. How has Home MV affected your lifestyle? 2. Do you feel that you made an informed choice when you first started home IMV or when it became permanent?
6. Jones et al. (1998)	UK; hospital	11	Patient satisfaction with domiciliary NIPPV.	Postal questionnaire completed after 2 years of treatment.	No details provided.

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Table 1. Continued

Article	Setting	Sample size of the COPD participants only	Outcome measures	Methods	Sample questions
7. Lynn et al. (2000)	USA; 5 teaching hospitals	416	Preference for MV.	Medical records and interviews with patients and their identified surrogates. Patients were interviewed at three instances in time: after study entry; during week 2 (or discharge), and then follow up conducted at 2 and 6 months after enrolment.	"preferences to die rather than spend all of the time on a ventilator" was assessed – interview.
8. Norris et al. (2005)	USA; community and hospital	111	Stated preference for intubation and MV and CPR.	Interviewer administered questionnaire.	"Participants were asked whether they would want intubation and MV and whether they would want CPR in four health states . . ."
9. Rocker et al. (2008)	Canada; multi-center study in 5 teaching hospitals	118	Patients' attitudes towards receiving information about MV.	A research coordinator at each site administered a 28-item questionnaire in person.	Patients had to rate whether it was extremely important that they receive information about MV.
10. Stapleton et al. (2005)	USA; community and hospital	101	Patient preferences for intubation and MV or CPR given their current health status.	Patients were interviewed in person by a trained study interviewer	". . . If you were in your current health and were unable to breathe on your own, would you want to be on a breathing machine for a few days?"
11. Travaline and Silverman (1995)	USA; hospital	37	Patient preferences for indefinite MV.	Patients were interviewed in person following their appointment with their physician – a questionnaire.	". . . The patient was then presented with the hypothetical situation of requiring MV as 'indefinite life-support' rather than temporary modality . . ."
12. Wilson et al. (2005)	Canada; rehabilitation centre	33	Preference for MV and intubation in the event of a life-threatening exacerbation.	The patient is provided with a decision aid (DA) to review at home. The DA comprised of a portable, self-administered and self-paced audio booklet. A second session was conducted to ask participants about their preferences for IMV.	Key information from the decision aid includes: (1) the average duration of MV; (2) proportion of patients who come off ventilation; (3) proportion of patients who will die without ever coming off MV, and; (4) proportion of patients who will be weaned from MV but die during the period of hospitalization.

Table 2. Summary of Key Results and Implications for HTA

Article	Main results	Results that may be pertinent for HTA
1. Chakrabarti et al. (2009)	• Stage 1 – 96% (48/50) stated a willingness to receive NIV if offered in the acute setting. • Stage 2 – 76% (38/50) stating that they would be willing to receive NIV. • Stage 3 – 84% (42/50) of the cohort stated that they would be willing to receive NIV. • Stage 4 – 60% (30/50) stated that they would be willing to receive IMV during an acute exacerbation. • Stage 5 – 58% (29/50) stated that they would be willing to receive IMV. • 48% expressed an interest to issue an Advanced Directive of Care • Patients willing to receive IMV were younger (67 versus 76 years $p = 0.016$) and had a better functional status (NEADL index 20 versus 15 units $p = 0.03$). • Willingness to receive NIV was not significantly associated with any covariates measured such as gender, health status, depression.	• Preferences for ventilation among COPD patients can vary according to how the procedure is presented and described. • 86% found demonstration of NIV helpful in decision making compared to 24% with the photographic aid ($p < 00.001$).
2. Claessens et al. (2000)	• 78% of patients would be "very unwilling" or "would rather die" than be attached to a ventilator "all the time" ($n = 445$). • 77% of patients responded 'yes' when it was the surrogate response b/c patient was unable to communicate ($n = 730$). • 37% of patients had a preference for a do not resuscitate order.	• Majority of COPD patients were willing to forgo a potentially-life saving procedure (ventilation).
3. Dales et al. (1999)	• Two males (20%) and all ten women declined intubation and MV. Three undecided patients did make a decision as a result of the DA (15%). An additional 3 patients changed their decision after reviewing DA (15%). • At 1 year, only 2 patients (11%) had changed their decision.	• For preferences regarding MV, there was: (1) no agreement between patients and next-of-kin and (2) The agreement between physicians and patients was 65%.
4. Gaber et al. (2004)	• 77% of patients said yes to IMV and 67% said yes to NIV. • Forty-eight patients (~48%) wanted all additional treatments (IMV, NIV, CPR) if needed and 12 wanted none (~12%). • 19 patients said 'no' for CPR but 'yes' to NIV and IMV (~19%). • 10 patients said 'no' to CPR and IMV but 'yes' to NIV (~10%). • 11 patients gave mixed answers (11%). • There were no statistical differences between the groups answering 'yes' and 'no' with respect to age, sex, spirometry, the use of antidepressants, the number of hospital admissions over the previous 12 months, being on LTOT or the BPQoL questionnaire score.	• Majority of patients said yes to IMV and NIV • 64 out of 65 patients agreed that preferences for NIV, IV or CPR should be discussed.
5. Goldstein et al. (1995)	• 33% ($n = 3$) felt that they had made an informed decision when they first started Home MV. • In terms of positive statements, 78% ($n = 7$) reported improved symptoms, 44% ($n = 4$) reported greater mobility and freedom and11% ($n = 1$) reported it was a life-sustaining treatment., • In terms of negative statements, 67% ($n = 6$) reported limited mobility and freedom, 0% reported equipment concerns or social implications.	
6. Jones et al. (1998)	• 75% claimed to be "very satisfied" with the treatment, and dissatisfaction in the remainder was related to skin ulceration on the bridge of the nose caused by the mask (a problem which the authors claimed to be resolved in most cases by careful adjustment of the head harness). • All respondents claimed improved sleep quality and relief of morning headache	
7. Lynn et al. (2000)	• Preferences to die rather than living on a ventilator were between 60%–70% of those interviewed. These preferences were stable over the three time windows (6mo-3mo before death; 3mo-1mo before death; 1mo-death).	• Majority of COPD patients were willing to forgo a potentially-life saving procedure (ventilation).

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Table 2. Continued

Article	Main results	Results that may be pertinent for HTA
	• In terms of decisions to use the ventilator as documented by medical records: At six to three months before death, 33% (n = 38) of patients made a decision to use MV, while 12% (n = 14) chose not to have MV. At three to one month prior to death, 46% (n = 76) chose MV, while 15% (n = 24) chose not to use MV. At one month before death, 40% (n = 103) chose to use MV, while 31% (n-81) chose not to use MV.	
8. Norris et al. (2005)	• Preferences for not wanting intubation and MV in four health states: (1) 39.6%; (2) 93.6%; (3) 84.5%; (4) 83.6%. • Preferences for not receiving CPR in the four health states were: (1) 38.4%; (2) 91%; (3) 81.7%; (4) 82.1%. • There were no significant differences between men and women in terms of these treatment preferences.	• Expressed preferences for MV vary depending on hypothetical health state.
9. Rocker et al. (2008)	• The aspect of End of Life care that was most important to patients was "not to be kept alive on life support when there is little hope for meaningful recovery" (rated as "extremely important" for 55% of participants). • 29% of patients thought that it was extremely important to have information about MV. (No data provided for the four other response categories.)	
10. Stapleton et al. (2005)	• A majority of the patients surveyed reported they would want MV (62.2%) and CPR (63.6%). • Age, number of co-morbid illnesses, FEV_1, gender, evidence of depression, HRQoL, were not associated with preferences for IMV in a multivariate model.	• A majority of patients surveyed reported that they would want MV.
11. Travaline and Silverman (1995)	• 40% (n = 15) responded they would use MV as indefinite support, 22% (n = 8) would not use MV, and 38% (n = 14) were unsure. • Age, sex, race, clinic or co-morbid conditions did not predict preference for MV.	• Patients who had discussions with their physicians were more likely to specify a choice concerning MV when compared with those patients who had no prior discussions. • Discussions concerning life-sustaining treatment with MV occurred infrequently between patients and their physicians (among only 14% of study participants).
12. Wilson et al. (2005)	• Only two participants (6%) reported that they had already made an advanced decision about MV at baseline. • After reviewing the decision aid, 94% (31) reported they made a choice. • Of those who made a decision, 74% (23) decided to forego MV in the event of a serious exacerbation. • 27% found the experience of the DA to be "a little" or "quite upsetting" and a further 18% found it to be "very upsetting". 12% found that the decision was "quite," "very," or "extremely" difficult to make. • A total of 22 participants (67%) reported that they had discussed their ventilation decision with another person. This was usually the spouse or other family member (21/22 = 96%), although in some cases it also included the family physician (3/22 = 14%). • Of the 23 patients who would forego MV, 5 (22%) cited a low likelihood of survival as being the most important reason for their decision. • For those who were inclined to accept MV (or who were uncertain), the most common reason was that these individuals had a strong personal will to prolong life (40%) or important external social reasons for staying alive (20%). • Those who would accept MV were significantly younger. However, the two groups did not vary by gender, pulmonary function, dyspnea, HRQoL, depressive symptoms.	

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The studies that satisfied the inclusion criteria generally focused on COPD patients' preferences for ventilation (Table 1, column 4). Three studies measured patient attitudes toward either receiving information about MV, or discussing preferences concerning MV/CPR with their health care professional (14–16). Nine studies assessed patient's stated preferences for the MV intervention (Table 1). One study measured patient decisions regarding MV retrospectively, by means of patient medical record review (17). In addition, many of these studies provided additional details by exploring whether ventilation decisions vary by patient characteristics or by how the question was addressed to patients. Two studies did not pertain to patient preferences; as they queried patients who had received home ventilation about their views on the intervention (18;19).

The questions that were posed to individuals about their preferences varied significantly from one study to another (Table 1, column 6). In some cases, patients were asked about their preferences for indefinite ventilation ("would you be willing, somewhat willing, somewhat unwilling, or rather die than put up with being attached to a ventilator all the time.") (20). In other cases, patients were asked about their preferences for being on a "breathing machine for a few days" (21). These are different hypothetical scenarios, one referring to a temporary use of ventilation, the other scenario suggesting indefinite dependence. One person with stable preferences may respond negatively to the first question, yet respond affirmatively to the second question. There is a further complication, as research has revealed that presenting individuals with exactly the same hypothetical situation, but with altered wording can produce different results (22). For these reasons, we developed data extraction tables (Tables 1, and 2) that would be suitable for this type of research. Tables should show the pertinent preference-related questions, so that results can be properly interpreted and understood.

Study Findings

Key study results are summarized in Table 2.

Do COPD Patients Want to Discuss Their Prognosis and Possible Interventions?. Three studies explored whether COPD patients want to discuss preferences for MV, or Advanced Directives of Care (ADC). Rocker et al. (16) found that only 29 percent rated it as extremely important to know about mechanical ventilation. Another study found that 48 percent of study participants were interested in issuing an ADC (14). In contrast, a UK study found that 98 percent of respondents thought that the sensitive issue of preferences for NIV, IMV (Invasive Mechanical Ventilation), and CPR should be discussed with patients (15). It is worth noting that only Gaber et al. (15) had patient preferences elicited by a respiratory nurse who was responsible for coordinating the patients' care needs; whereas, patients were queried about their preferences by study personnel in the previous two studies (14;16).

Proportion of COPD Patients Expressing Willingness to Use MV. Several studies elicited COPD patient preferences for MV (Table 2). The proportion of COPD patients who indicated that they would opt for MV varies widely across studies from approximately 22–77 percent (14;15;17;20–23). Note, however, that the respondents were asked very different preference-related questions.

Two studies used a comprehensive decision aid to elicit patient preferences concerning MV and intubation. They found that a majority of respondents were willing to forgo the intervention (60 percent to 74 percent) (12;24).

Proportion of COPD Patients Expressing Willingness to Use NIV. Only two studies on NIV preferences among COPD patients were identified for this review. A UK study found that approximately 67 percent of their respondents were willing to receive NIV if "admitted to hospital with worsening of their condition" (15). Another UK study in a hospital setting reported that 76 percent to 96 percent of COPD patients were willing to receive NIV if offered in the acute setting (14).

Predicting Preferences for MV and NIV among COPD Patients. Six studies tested associations between patient preferences for ventilation and covariates (11;12;14;15;21;23). None of the variables that were tested showed a consistent association with preferences for MV across all or even most studies. No associations were found between patient preferences for NIV and any measured covariate (14;15) One study, albeit with a small sample size (n = 19) compared patient preferences with the ventilation decisions that would be made by the patient's physician or family member (24). Agreement between patient preferences and physician was low (65 percent) and even lower between patient and next of kin (0 percent).

Patient Preferences Regarding MV for Serious Illness Situations. An American study explored COPD patient preferences for intubation and MV under different hypothetical health states (11). Only a minority of COPD patients (31 percent) stated that they would reject intubation and MV in their current health state. However, when asked about intubation and MV in situations of permanent coma, dementia or bed-bound, the percentage of participants rejecting the intervention was: 94 percent, 85 percent, and 84 percent, respectively.

The Issue of "Framing" and Patient Preferences for IMV and NPPV. One study elicited preferences – a multi-stage interview (14). They found that following a verbal description, 96 percent of study participants expressed a willingness to receive NIV. However, when shown a photograph, the percentage dropped to 76 percent and after receiving a demonstration, it was 84 percent. In terms of IMV, 60 percent were willing to receive the intervention following a verbal description. This decreased to 58 percent when patients were given an explanation of alternative treatments to IMV.

Decision Aids: Stressful for COPD Patients?. One study provided greater detail about the decision-making process by asking participants

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whether working through a decision aid was stressful (12). Although 55 percent of respondents found that "working through the decision aid" was "not at all upsetting," some patients found it "a little" (18 percent), "quite" (9 percent) or "very upsetting" (18 percent). In addition, five individuals (15 percent) completed the first interview but declined to take part in the follow-up, and of these, three "specifically indicated that they had been upset by the content of the decision aid".

COPD Patient Satisfaction with MV, NIPPV in the home. Two studies explored the experiences of COPD patients postventilation (18;19). Unfortunately, these two studies have considerable limitations: small sample sizes ($n = 9$ and $n = 11$, respectively) and the absence of a comparison group.

Brief Summary and Synthesis

These results indicate that: (i) a substantial proportion of COPD patients are willing to forgo a life-saving procedure (e.g., MV); (ii) expressed preferences among COPD patients for ventilation are not always fixed but can be contingent on the hypothetical situation that is presented to them and on how the intervention is described; (iii) a majority of COPD patients are willing to try NIV; (iv) it is difficult to predict those COPD patients who are willing to receive either IMV or NPPV based on demographic or clinical data, or by speaking to their physician or next of kin; and (v) not all patients want to discuss MV.

Additional insights can be obtained by briefly synthesizing some key themes across studies. Careful scrutiny of the preference-related question indicates that studies that asked patients about MV as "indefinite life-support" (23) or for "all the time" (17;20) had few responding affirmatively (approximately 22–40 percent), whereas studies that asked respondents whether they would be "willing to receive" the intervention (14;15) or willing to be on a machine "for a few days" (21) had approximately 58–77 percent responding affirmatively. Although these comparisons are across studies and with different individuals, this suggests that for many patients this unpleasant procedure may be tolerated for a short period of time but would not be chosen if the procedure is indefinite. The results suggest that health care professionals need to be mindful about how they pose these questions to their patients, because framing these questions in different ways may result in patients offering different responses. Patient preferences may be best elicited through a conversation in which a few different hypothetical situations can be described with patients, and in which possible time frames for the procedure are carefully described.

The results of these studies indicate that some patients do not want to discuss sensitive issues such as preferences for MV or ADCs. Furthermore, for some individuals working through a decision aid was upsetting. However, it is interesting that the one study in which preferences were elicited from a respiratory nurse who was responsible for coordinating the patients' care needs had 98 percent of respondents who thought that the sensitive issues of preferences for NIV, IMV, and CPR should be discussed. This suggests that preference-related discussions might be best addressed within the context of an established health care relationship.

DISCUSSION

Implications for Preference-Related Data within HTA

We found that it is indeed feasible to conduct a systematic review of quantitative preference-related evidence for an HTA topic and that it yields substantial insights beyond conventional evidence reviews and CEAs. Although effectiveness studies indicate whether an intervention is effective (or contributes favorably to QoL), they do not provide any indication of how many patients will opt for the procedure. By contrast, quantitative preference-related data are particularly useful for understanding the acceptability of an intervention among patients across a range of settings, countries, and based on a range of different questions. There are several other methods that can be used to understand patient preferences, such as patient representation on HTA committees, or qualitative research. However, quantitative research generates data with the most breadth, thus particularly useful for estimating the patient acceptability of an intervention across an entire population.

This evidence could be used to inform purchasing decisions. For instance, if one must decide between two technologies of equivalent effectiveness, then choosing the technology that more patients prefer can optimize patient outcomes.

This type of review has other advantages. It can sensitize readers to the importance of patient preference within the policy domain and at the level of the patient-physician interaction. For physicians who may read such evidence (within an HTA), it can reaffirm the importance of soliciting their patients' preferences and provide suggestions about how to pose these difficult questions to their patients. For patients, understanding the treatment choices made by others who share their illness, enhances patient knowledge and can thus be empowering.

The process of conducting this preference-related case study also revealed several methodological challenges. We found that the taxonomy was inconsistent across studies. Some researchers used the word "preferences," while others used terms such as "attitudes toward . . ." when referring to the same construct. Developing appropriate searches is further complicated by the highly varied study designs in this area. For instance, research using discrete choice experiments in which COPD patients are asked about their least and most favorite clinical and social outcomes clearly has implications for their preferences for ventilation, but this research was excluded for this review because it did not satisfy our inclusion criteria as ventilation was not specifically addressed. In addition, although we found many articles associated with patient preferences, our search revealed fewer articles that related to other types of patient perspective research. But "patient perspectives" is an amorphous area with unclear boundaries. Researchers choosing

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to expand the search on topics related to patient perspective could do so easily; for instance, by including research that investigates the social and psychological consequences of using the intervention.

In addition, we found that the studies in this review used nonstandardized questionnaires and posed different types of preference-related questions. Therefore for this type of evidence, a meta-analytic approach is not ideal. However, a narrative synthesis is appropriate, because in this case, these differences in questions and answers can be analyzed and interpreted, rather than simply ignored and averaged out (as in meta-analysis). Because the primary purpose of this study was to understand how this literature might inform the HTA process, we did not perform an extensive, critical appraisal of the research literature, However, to conduct a critical appraisal of this type of literature, a checklist of items would have to be developed (e.g., response/drop-out rates, validity of measures) and evidence would be judged based on the quality score of each research article. By contrast, systematic reviews of the effectiveness of a technology can typically exclude a large number of studies (e.g., those that are not RCTs). For all of these reasons, a systematic review of the preference literature is messier and likely more time consuming than a traditional, systematic review of effectiveness. Due to these challenges and disadvantages, we do not recommend a systematic review of preference-related data to be appropriate as a routine part of the HTA process. However, we do suggest that it is a promising addition to the HTA process, for preference-sensitive technological decisions and in particular, if some of these methodological challenges are sorted out.

In light of the considerations presented above, we suggest that the area of patient perspectives, preferences, attitudes, and values would benefit from further conceptual and taxonomic work that specifies the conceptual distinctions and commonalities of terms. There is a broad range of terms (preference, quality of life, satisfaction, values, choices, utility), which emanate from different academic disciplines (e.g., economics, psychology, nursing) and/or specific research methods (e.g., qualitative research, conjoint analysis). Identifying all of these terms and understanding the areas of difference and overlap will be a major undertaking, but will be extremely useful when seeking to integrate patients' perspectives into HTA.

SUPPLEMENTARY MATERIAL

Supplementary Table 1: www.journals.cambridge.org/thc2013119

CONTACT INFORMATION

Ann-Sylvia Brooker, PhD, MSc (as.brooker@utoronto.ca) THETA (Toronto Health Economics and Technology Assessment) collaborative, University of Toronto, Toronto, Ontario, Canada

Steven Carcone, MSc THETA collaborative, University of Toronto, Toronto, Ontario, Canada

William Witteman, BSc., MLIS THETA collaborative, University of Toronto, Toronto, Ontario, Canada

Murray Krahn, MD, MSc, FRCPC Director of THETA collaborative, F. Norman Hughes Chair in Pharmacoeconomics, Social and Administrative Pharmacy Division Head in the Faculty of Pharmacy, Professor at the University of Toronto, Scientist at Toronto General Research Institute; Adjunct Scientist at the Institute for Clinical Evaluative Sciences

CONFLICTS OF INTEREST

All authors report they have no potential conflicts of interest.

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