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How long do patients with chronic disease expect to live? A systematic review of the literature

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Title

How long do patients with chronic disease expect to live?

A systematic review of the literature

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Abstract

Objectives: To systematically identify and summarise the literature on self-estimated life expectancy by individuals with non-cancer life-limiting illnesses

Setting: Published and grey literature from 1985 to 2015 where adults with non-cancer chronic disease were asked to estimate their own life expectancy.

Participants: From 2356 titles six studies were identified that met pre-specified criteria for inclusion. Studies came from the UK, Netherlands and USA. A total of 545 subjects were included (heart failure 389; chronic obstructive pulmonary disease 89; end stage renal failure 62; chronic kidney disease 5). No papers reporting on other lung diseases, neurodegenerative disease or cirrhosis were found.

Primary and secondary outcome measures: All measures of self-estimated life expectancy were accepted. Self-estimated life expectancy was compared, where available, with observed survival, physician-estimated life expectancy and model-estimated life expectancy. Meta-analysis was not conducted due to the heterogeneity of the patient groups and study methodologies.

Results: Amongst patients with heart failure, median self-estimated life expectancy was 40% longer than predicted by a validated model. Outpatients receiving haemodialysis were more optimistic about prognosis than their nephrologists and overestimated their chances of surviving five years. Patients with heart failure and COPD were approximately three times more likely to die in the next year than they predicted. Data available for patients with chronic kidney disease were of insufficient quality to draw conclusions.

Conclusions: Individuals with chronic disease may have unrealistically optimistic expectations of their prognosis. More research is needed to understand how self-estimated life-expectancy affects behaviour. Meanwhile, clinicians should attempt to identify each patient's prognostic preferences and provide information in a way that they can understand and use to inform their decisions.

Trial registration: Prospero registration number: CRD42015020732

Strengths and limitations of this study

Strengths

- This is the first review of self-estimated life expectancy amongst patients with chronic non-cancer disease
- The findings build on and reproduce the oncology literature showing patients with cancer have a tendency to overestimate their life expectancy and chances of cure

Limitations

- The findings of this review are based on the small number of studies that have been conducted on this subject
- Literature was only available for patients with heart failure, renal failure and COPD

INTRODUCTION

An individual's health expectations reflect both how well they understand their disease and the medical profession's ability to prognosticate for and communicate with them. Prognostic forecasts may affect a variety of outcomes, including healthcare choices[1, 2]. Where decisions are affected by life expectancy, patients can only be considered fully-informed if they have an understanding of their prognosis, and the effects available treatments might have upon it.

Prognosis communication has been widely studied in malignancy. A systematic review found the majority of people with cancer want detailed prognostic information, presented honestly and openly[3]. Despite this, many patients – including those with advanced disease – report never discussing prognosis, or misunderstand their treatment aim and chance of survival.

Life expectancy for patients with advanced chronic disease; including chronic obstructive pulmonary disease (COPD), heart failure (HF) and end-stage renal failure (ESRF) can be as poor as that seen in incurable cancer[4-6]. However, the cultural meaning and clinical course of cancer and non-cancer diseases differ significantly, limiting extrapolation of findings from the oncology literature[7]. A systematic review of studies reporting self-estimates of life expectancy by patients with non-cancer life-limiting disease was conducted.

METHODS

A systematic search was performed of Medline, Embase, PsychINFO and the Cochrane Library. Unpublished works were searched using ProQuest dissertations and theses search, the Networked Digital Library of Theses and Dissertations Global ETD search and the System for Grey Literature in Europe. Search terms relating to 'life expectancy' and 'self-estimated' were used (see Appendix A). Search results were limited to publications from 1985 to November 2015 and English Language. Literature predating 1985 was deemed unlikely to inform understanding of current practice.

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3 Non-cancer life-limiting disease was defined as HF; chronic kidney disease stage five (CKD5); ESRF
4 receiving dialysis or conservative care; COPD, interstitial lung disease, neurodegenerative disease
5 and liver cirrhosis. Studies were included where adults (≥ 18 years of age) with these conditions were
6 asked to estimate their life expectancy. All measurements of life expectancy were accepted,
7 including those in terms of duration (e.g. *“How long do you expect to live”*), and chance (e.g. *“What*
8 *is the chance you will be alive in five years”*). Studies were excluded where only self-estimated
9 probability of ‘cure’ was determined, where the only option for survival duration was less than six
10 months and where subjects were asked to consider only hypothetical situations (e.g. *“How long do*
11 *you think you would live if you had a kidney transplant”*). Studies reporting only on subjects with
12 cancer, HIV/AIDS, congenital heart disease, cystic fibrosis and organ transplant were excluded. In all
13 these conditions the situation, illness culture or advances in treatment may have affected how
14 generalisable findings were to the larger chronic disease population. At the title and abstract
15 searching phase, papers assessing prognosis in excluded diagnoses were not rejected, so that
16 reference list searches could be performed from these papers. Where studies reported a mixture of
17 included and excluded diagnoses, they were incorporated if the data on individual diseases were
18 reported separately. Where data were not separately reported, authors were contacted to request
19 supplementary files.

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Titles were independently examined by two reviewers (BH and JS) according to the above criteria,
and a Kappa statistic calculated to assess agreement. Abstracts from titles accepted by either one or
both reviewers were collected and assessed independently, using the same criteria, and included if
both recommended inclusion. Where only one reviewer recommended inclusion, a consensus
decision was made after discussion. Full text articles were requested and read and reference lists
examined with additional papers included by the same criteria. At this point, papers reporting
excluded disease groups were rejected. Papers included for review were assessed using a purpose-
developed assessment tool (Appendix B) and evidence graded as low, medium or high quality. The
study was registered with the PROSPERO database, registration number CRD42015020732.

RESULTS

The initial search provided 2356 titles after removal of duplicates. 116 abstracts were selected for review by either one or both authors (agree to exclude, 2240; agree include, 68; disagree, 48; Kappa 0.73). 26 papers were collected and reference list searching provided an additional six. After full text examination of 32 papers, seven papers from six studies were included in the review (Figure 1). A complete list of papers including reasons for inclusion/rejection is available (Appendix C). Evidence was graded as medium in four and low in three of the included papers (Table 1).

Studies came from the UK,[8] Netherlands [9] and USA[10-14]. A total of 545 subjects were included (HF, 389; COPD, 89; ESRF, 62; CKD5, 5). Four papers reported on a single medical disease; HF[9, 11] and ESRF.[10] Others reported on a mixture of conditions; HF and COPD[12, 13] and HF, CKD5 and COPD[8]. No papers reporting on non-COPD lung disease, neurodegenerative disease or cirrhosis were found.

The mean age of study participants ranged from 58 to 75. In the study by Fried *et al.* only individuals over 60 years of age were recruited[12, 13] and only those over 50 in the study by Kraai *et al.*[9] No minimum age was set in the other studies. Two studies (accounting for 222 of the 389 subjects with HF) did not include selection criteria for disease severity.[9, 11] In the other studies criteria were used to select for patients with advanced disease. Patients with ESRF were all receiving outpatient haemodialysis.[10] Reported levels of comorbidity were high. The mean Charlson Comorbidity index for patients with ESRF was 5.8 (SD 1.6).[10] Amongst US patients with heart-failure in one study 82% had hypertension, 54% diabetes and 29% COPD.[11] Amongst patients with heart failure from the Netherlands, 57% had hypertension, 30% had diabetes, 24% had COPD and 11% had had a stroke.[9]

One study used a written questionnaire to measure self-estimated life expectancy.[14] All other studies used interviews. Participants with ESRF were asked about their chances of being alive at different time points.[10] In the other studies, participants were asked to indicate how long they

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3 expected to live by selecting from vignette answers,[8] giving a verbal response[11-13] and/or by
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5 using a visual analogue scale.[9, 11] In one study it was not possible to ascertain how the question
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7 had been posed or answered.[14] For studies where data were available, 168 of 541 (31%) initially
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9 eligible patients were excluded from the studies, largely on the grounds of language skills or
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11 cognitive impairment. 105 of 408 interviewed patients (26%) were unwilling or unable to estimate
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13 their own life expectancy.
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16 Self-estimates of life expectancy were compared with predictions from clinical risk calculators,[11]
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18 clinician-estimated life expectancy,[8, 10, 12, 13] observed survival[8, 10-13] or presented without
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Table A: Summary of included papers

Authors	Year	Country	Quality	Design	Patients included	Measures used	Results	Summary	Pros + and cons –
Allen et al.	2008	USA	Medium	Cross-sectional interviewer-administered questionnaire in a single centre outpatient heart-failure service.	122 sequentially recruited subjects with heart failure (NYHA-IV) Mean age 61 (IQR 53-74) 62% male 47% African American	1) Patients were asked “If you had to guess, how much longer do you think you will live?” and completed a) Multi-choice answers ranging from <3 months to >10 years, and b) A visual analogue scale, marking their estimated age at death 2) Model-predicted life expectancy using the Seattle Heart Failure Model 3) Observed survival over median follow-up of three years	Median self-estimated life-expectancy was 13 years (IQR 8-21; range 1-54 years) Median model-predicted life-expectancy was 10 years (IQR 7.2-13.3; range 2.0-25 years) 66% of patients overestimated their survival compared with the model by 30% or more The median overestimate was 40% 29% of patients died within three years.	Self-estimated-life expectancy was on average significantly greater than that predicted by a validated model Younger age, greater disease severity and measures of less depression were independently associated with overestimation of survival	+ Efforts made to improve and check patient understanding of question – 26 of 148 enrolled participants felt unable/unwilling to estimate survival – Only 35 of 122 patients were followed up until their death – Only 9 of 122 patients had NYHA IV heart failure – No index group without chronic disease was included
Fried et al.	2003	USA	Medium	Cross-sectional interview survey administered to patients registered at community practices and outpatient clinics of two hospitals, and inpatients of three hospitals. Same patient	135 patients with COPD or HF, aged 60 and older, meeting criteria for limited life expectancy and requiring assistance with daily living COPD – 79 patients Mean age 72 (SD 7) 51% Male 92% White HF – 56 patients Mean age 75 (SD 8) 70% Male 88% White	Patients and clinicians were asked how long they thought the patient would live and answered using multi-choice options ranging from <1 month to >10 years	Only 9 of 135 patients expected to live less than one year, but 38 patients died over this period. 58 of 79 patients who responded to being asked to estimate their own life expectancy expected to live two years or more Of the 65 available patient-clinician pairs who both responded, 34 agreed the prognosis was two years or more, 9 agreed the prognosis was two years or less, 7 clinicians	Patient expectations of one year mortality are higher than observed. Agreement between patients and their clinicians about likely prognosis is poor.	– 56 of 135 patients were unable or unwilling to estimate their life expectancy – No index group without chronic disease was included

				group as <i>Fried et al.</i> 2006		thought the patient would live two years or more when the patient did not expect to live this long and 15 patients expected to live two years or more when their clinician was less optimistic.			
							Kappa was 0.22 suggesting very poor agreement		
Fried et al.	2006	USA	Medium	Serial interview survey administered to patients registered at community practices and outpatient clinics of two hospitals, and inpatients of three hospitals. Same patient group as <i>Fried et al.</i> 2003	135 patients with COPD or HF, aged 60 and older, meeting criteria for limited life expectancy and requiring assistance with daily living COPD – 79 patients Mean age 72 (SD 7) 51% Male 92% White HF – 56 patients Mean age 75 (SD 8) 70% Male 88% White	Patients were asked how long they thought the patient would live and answered using multi-choice options ranging from <1 month to >10 years	9 of 59 patients who responded expected to live less than one year at their first interview. 5 of 59 expected to live less than one year at their final interview. 38 of 135 patients died over this period.	Patient expectations of one year mortality are higher than observed. The majority of patients (both those who were alive and dead at the end of the year-long study) made no adjustment to their self-estimated life expectancy.	– 56 of 135 patients were unable or unwilling to estimate their life expectancy – No index group without chronic disease was included
Kraai et al.	2013	The Netherlands	Low	Cross-sectional questionnaire administered in outpatient setting in one heart failure clinic. Sub-component of time trade-off study.	100 patients with heart failure (NYHA I-IV) all over 50 years of age. Mean age 70 (SD 9.4) 71% male	Visual analogue scale from 50 to 100 years of age; patients were asked to indicate the most accurate estimation of their life expectancy.	Mean life expectancy indicated by patients was 82 (SD 8.6) years. No difference in self-estimated life expectancy was found between patients unwilling vs. willing to trade time	Self-estimated life expectancy probably exceeds likely outcomes, but no comparator data was available. Despite patients with more advanced or symptomatic heart failure being more willing to trade time, no difference was found between the groups in terms of expected	– No comparator prediction or measurement of survival used – Only 2 of 100 patients had NYHA IV heart failure – No index group without chronic disease was included

								longevity.	
Shah et al.	2006	UK	Low	Cross-sectional interviewer-administered questionnaire in outpatient and inpatient settings at one acute NHS Trust and a neighbouring Hospice.	20 patients in total meeting criteria for limited life expectancy: 6 HF (NYHA III/IV) 9 COPD 5 CKD Median age 72 50% male 85% white	Patients and physicians chose one of seven short prognosis statements that most accurately predicted how their illness might affect their life expectancy.	7 of 20 (35%) patients estimated their prognosis to be <1 year 13/17 physicians (76%) estimated their patient's prognosis to be < 1 year	Exploratory study, no firm conclusions available	– Very small numbers – Sample poorly representative of a general outpatient population – No index group without chronic disease was included
Stewart et al.	2010	USA	Low	Cross-sectional written questionnaire with both inpatients and outpatients from two heart failure centres. Sub-component of time trade-off study.	105 patients with left ventricular ejection fraction (LVEF) <35% and symptomatic heart failure Mean age 58 (SD 13) 70% Male	Methodology for collecting self-estimated life expectancy not described	65% thought they would live more than 10 years and 34% believed they would be alive for at least 20 years. Patients willing to trade more time expected shorter survival than those unwilling to trade time. 46% of the patients willing to trade away at least 12 months anticipated that they would not survive 5 years. No difference was found in self-estimated survival between inpatients and outpatients (data not provided)	Self-estimated life expectancy probably exceeds likely outcomes, but no comparator data was available. Willingness to trade time is associated with shorter self-estimated life expectancy.	– No comparator prediction or measurement of survival – Only 3 of 105 patients had NYHA IV heart failure – Study methodology and tool not described – No index group without chronic disease was included
Wachterman et al.	2013	USA	Medium	Cross-sectional interviewer-administered questionnaire in two community-based haemodialysis	62 patients receiving maintenance haemodialysis with 20% or greater predicted risk of dying in the next year. Mean age 68 (SD 10) 42% Male	1) Patients asked what they thought their chance was of being alive at 1 and 5 years (>=90%, about 75%, about 50%, about 25%, <=10%, don't know). 2) Nephrologist in charge of care asked to estimate each	For 1 year survival prediction, patients were more optimistic in 64% of patient-nephrologist pairs, whereas nephrologists were more optimistic in only 10%. For 5 year survival	Patient expectations of five year mortality are higher than observed. Patients were significantly more optimistic about their survival than their nephrologists.	– 88 of 150 eligible patients were excluded or refused to participate – No index group without chronic disease was included

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s units.	52% Black	patients' chance of being alive at 1 and 5 years on a continuous scale of 0% to 100%.	prediction, patients were more optimistic in 69% patient-nephrologist pairs, whereas nephrologists were more optimistic in only 2%	Patients' 1 year survival expectations were more consistent with actual survival than clinician estimates.
		3) Survival data with follow up of 23 months	Only 6% of patients thought they had a less than 50% chance of being alive at 5 years, whereas actual survival at 23 months was only 56%.	Patients who expected to live longer were more likely to opt for life-extending treatments

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3 comparator data.[9, 14] Follow up periods ranged from one to three years and the majority of
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5 patients (range 56-73%) were alive at the end of the studies. Analysis was performed in one study to
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7 characterise factors associated with overestimation of survival.[11] In three papers patients were
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9 asked about their preferences around treatment aims, and analyses performed looking at how these
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11 responses correlated with self-estimated life expectancy.[9, 10, 14] One paper used repeat measures
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13 to examine how self-estimated life expectancy changed with disease course.[13]
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15 16 17 **Self-estimated life expectancy compared with observed survival**

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19 In general, self-estimated life expectancy exceeded observed survival. The only example of self-
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21 estimated life expectancy consistent with survival was one-year mortality in patients with ESRF.[10]
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23 81% of patients thought they had a better than 90% chance of being alive at one year. Observed
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25 survival was 93%. In comparison, 96% of patients believed they had a better than 50% chance of
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27 being alive at five years, but 44% had died within just 23 months. In one study only 5% of patients
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29 with HF estimated their life expectancy to be three years or less, but observed mortality was 29%
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31 after a median follow-up of 3.1 years.[11] Amongst patients with advanced HF, 3 out of 56 (5%)
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33 patients expected to live less than one year, but 17 (30%) were dead in this period.[12] 6 out of 79
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35 (8%) patients with COPD in the same study predicted their life expectancy to be less than one year;
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37 21 (27%) died. When interviewed within the 90 days before they died, only 2 out of 16 patients
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39 predicted their life expectancy to be less than a year.[13] Patient numbers were too low in one study
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41 to draw conclusions from observed survival.[8]
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46 47 **Self-estimated life expectancy compared with model-predictions of survival**

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49 In the only study that used a validated model[15] to predict survival, self-estimated life expectancy
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51 exceeded model predictions.[11] Median self-estimated life expectancy for 122 patients with HF was
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53 13 years and median model-predicted life expectancy 10 years. There was no significant relationship
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55 between self and model-predicted life expectancy. The median ratio between self-estimated and
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3 model-estimated life expectancy was 1.4; indicating a 40% overestimation. Self-estimates of life
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5 expectancy were more similar to model predictions based on age and gender alone, than to
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7 predictions taking heart disease into account.
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10 **Self-estimated life expectancy compared with clinician-estimated survival**

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12 Agreement between patient and physician predictions of life expectancy was poor. Patients tended
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14 to be more optimistic about life expectancy than their clinicians. Estimating one year survival;
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16 patients with ESRF on dialysis were significantly more optimistic than their nephrologist in 64% of
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18 patient-clinician pairs, whereas nephrologists were more optimistic in only 10%. Estimating five year
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20 survival, patients were significantly more optimistic in 69% of patient-nephrologist pairs, whereas
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22 nephrologists were more optimistic in only 2%.^[10] Amongst patients with COPD and HF, agreement
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24 between patient and clinician about whether the patient would survive two years was poor, with a
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26 Kappa statistic of 0.22.^[12] Numbers of patients in one study were too small for any conclusions to
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28 be drawn.^[8]
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33 **Other findings**

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36 Younger age, greater disease severity and lower levels of depression were independently associated
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38 with self-estimated life expectancy exceeding model predictions amongst patients with heart
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40 failure.^[11] Patients receiving haemodialysis who thought they had a $\geq 90\%$ chance of being alive in 1
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42 year were significantly more likely to choose life-extending therapy (44%) than patients who
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44 reported a $< 90\%$ chance (9%).^[10] Patients with advanced COPD and HF serially interviewed over
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46 one year showed no evidence of adjusting their self-estimated life expectancy with disease
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48 progression.^[13] Only one patient of 135 revised their estimate from greater than one year to less
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50 than one year. Mortality was 28% over this period. Two studies found that patients with heart failure
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52 make estimates of their life expectancy that are likely to be optimistic, but did not provide any other
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54 prediction or measure of survival. One found patients who anticipated shorter survival to be more
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3 willing to trade longevity for improved quality of life than those who predicted longer lives.[14] The
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5 other study did not demonstrate this.[9]
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8 **DISCUSSION**

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10 Practice guidelines advocate considering prognosis when making decisions with patients who have
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12 chronic disease[16, 17] and promote sharing survival statistics with patients.[18, 19] There is
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14 evidence from both the cancer[13, 20, 21] and chronic disease[12, 22, 23] literature that patients
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16 with life limiting illness want open and honest communication about their prognosis. Where
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18 treatment options differ markedly in survival benefit, patients require an understanding of their life
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20 expectancy with each treatment to make fully-informed decisions between them. Hospitalised
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22 individuals are more likely to want cardiopulmonary resuscitation if they expect to survive their
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24 illness, even if these expectations are improbable.[2, 24] Patients with terminal cancer who are
25
26 optimistic about their prognosis are more interventional in their choice of medical therapy.[1] It is
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28 conceivable that behaviours as diverse as adherence to preventative drugs and deciding whether to
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30 make a will could be influenced by how long an individual expects to live.
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35 In this systematic review of self-estimated life expectancy in chronic disease, individuals' estimates
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37 exceeded nearly all predictions and measures of survival; including model-predicted and observed
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39 survival. Patients were more optimistic than their clinicians when estimating life expectancy. Only in
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41 one instance (one year survival in ESRF) were patients' estimations in keeping with actual survival,
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43 and more accurate than their physicians', but by two years this had reversed. Patients with HF and
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45 COPD were approximately three times more likely to be dead within the year than they predicted.
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47 Life expectancy was overestimated by a median of 40% by patients with heart failure, when
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49 compared with a validated model; equating to three years of life for the average patient. Self-
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51 estimates were more in keeping with the life expectancy of matched adults without chronic disease.
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54 There was evidence that individuals with the worst prognosis may be the most overoptimistic, and
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3 that no meaningful adjustment in expected survival is made by patients approaching the ends of
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5 their lives.
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8 If the findings of this review reflect pervasive overestimation of life expectancy by individuals with
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10 chronic disease, there are several possible explanations. Firstly, patients might never be informed
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12 that their condition could affect their life expectancy. Such individuals are likely to base survival
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14 expectations on familial and media exposure, influenced by hopefulness and 'fighting spirit'. Others
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16 might receive overoptimistic forecasts; either due to methods of estimation, or adjustment by the
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18 communicating clinician. Finally, patients might be provided with appropriate quantitative
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20 estimates, but instead, form more favourable personal predictions.
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24 These findings are compatible with the oncology literature. Most patients with cancer want to
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26 discuss life expectancy, although desire for quantitative estimation varies.[25] Despite this, many
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28 report not having discussed prognosis, or are found to misunderstand the status of their disease, the
29
30 aim of their treatment and their prognosis.[3] Overestimation of the chances of cure and survival is
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32 common, even if disease is advanced and where individuals report having discussed prognosis with
33
34 their clinician.[26] The prognosis in non-cancer disease can be equivalently poor to that seen in
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36 malignancy.[4-6] Care must be taken generalising findings, given the cultural and clinical differences
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38 between conditions.[7]
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42 End of life care discussions between patients with chronic disease and their clinicians appear not to
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44 be routine.[22, 27, 28] None of the patients with ESRF in this review recalled discussing life
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46 expectancy with their clinician; their nephrologists reported they had done so with only 3% of the
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48 patients.[10] 63% of patients with HF in one study did not recall having spoken with their physician
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50 about their prognosis following the diagnosis of heart failure and only 36% believed HF would
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52 shorten their life.[11] Only 22% of patients in one study with advanced COPD and HF recalled having
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54 been told that they could die of their disease and only 1% recalled having been given an estimate of
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56 how long they might live.[12]
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3 There are boundaries to clinicians initiating prognostic discussions, such as fear of causing anxiety or
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5 destroying hope;[29] uncertainty about the validity, accuracy or precision of estimates;[30] cultural
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7 differences; and lack of experience and training in communication skills.[31] Some patients will not
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9 feel able to discuss prognosis, so clinicians must take care to elucidate preferences for information.
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11 However clinicians should continue to provide opportunities for prognostic discussion, since
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13 preferences may change over time and with disease progression. In other diseases such as breast
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15 cancer, the use of prognostic models and decision tools has been shown to increase understanding
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17 of prognosis and treatment options, leading to higher degrees of satisfaction.[32] Validated tools to
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19 help predict survival in chronic disease are available,[15, 33-35] but there is no evidence that these
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21 are widely employed. Only a minority are provided with accessible calculators (Box A). Studies are
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23 needed to examine how prognostic tools can be used in the clinical setting.[36] It is possible that
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25 clinical practice has not kept pace with the paradigm shift towards information-sharing with
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27 patients. Even where prognostic discussions happen, survival statistics may be misrepresented or
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29 censored.[37] In one study included in this review, nephrologists provided estimates of life
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31 expectancy for 89% of the interviewed patients, but reported they would withhold over half of these
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33 estimates in clinical practice.[10]

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38 The ability to make firm conclusions from the literature was highly limited by the lack of available
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40 evidence. The literature comes largely from single centre cohorts and is of medium to low-quality.
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42 Data from diseases other than COPD, heart and kidney failure is extremely limited, and those with
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44 the most advanced disease were under-represented. Included studies are likely to have come from
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46 centres where prognostication is considered important. We excluded studies including only subjects
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48 with cancer, HIV/AIDS, congenital heart disease, cystic fibrosis and organ transplant. The cancer
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50 literature has been well summarised,[3] but it is possible that these excluded conditions could have
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52 provided additional insight. We are aware of only one paper that would have been included without
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54 this exclusion, showing that young adults with congenital heart disease expect to live almost as long
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56 as their healthy peers.[38]

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3 There is no standardised or validated method for assessing self-estimated life expectancy, and it is
4 likely that responses are influenced by methodology. Additionally, asking a patient how long they
5 expect to live facilitates a quantitative assessment of their understanding, but does not provide
6 information on how such perceptions are formed and influenced. Large numbers of patients were
7 excluded from the studies or were unable or unwilling to estimate their own life expectancy, with
8 the potential to introduce bias. In addition, many patients were excluded on grounds of language
9 skills or cognitive impairment. These excluded individuals are likely to find discussing and
10 understanding prognosis particularly challenging and this undermines the relevance of the included
11 studies to a population of patients with chronic disease, in whom cognitive impairment is common.
12 All the studies reporting actual survival were limited by short follow-up times and low numbers of
13 deaths in the cohorts. Hospitalised patients were underrepresented in the included studies. It is
14 feasible that survival expectations are different during periods of acute illness requiring admission;
15 the point at which critical decisions about healthcare are often made. There is evidence to suggest
16 that overestimation of survival persists in these situations however; both in malignant and non-
17 malignant disease.[2, 24, 26, 39]

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36 None of the included studies had a healthy reference group. Overestimation of life expectancy
37 cannot, therefore, be presumed a phenomenon limited to patients with disease. A recently
38 published prospective cohort study provides some evidence to suggest self-estimation of survival
39 might be different amongst individuals unselected for chronic disease. Approximately half of
40 participants made predictions of their life expectancy consistent with those from a statistical
41 model.[40] Where predictions were inaccurate, they were approximately three times more likely to
42 be under, than over-estimates. Overestimation increased with age, but it is unclear whether this
43 represented an independent effect of ageing on subjective life expectancy, or confounding by the
44 increased prevalence of disease. It is possible that general population studies of self-estimated life
45 expectancy could be analysed for differences between individuals with and without disease.
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CONCLUSION

Patients with non-cancer life-limiting illness may have survival expectations that markedly exceed outcomes. These expectations might lead some patients to make health decisions and life choices that they would not if their predictions were more realistic. A better understanding is needed of the interaction between survival expectations and behaviour in chronic disease. If compelling evidence is found showing overestimation of survival leads patients to make decisions out of keeping with their likely future, approaches to adjusting such expectations could be developed. Meanwhile, clinicians caring for patients with chronic disease must make attempts to elucidate what prognostic information each patient already knows, wants to know and might benefit from knowing. Appropriate information should then be shared in a form that the patient can use to inform their decisions.

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CONTRIBUTORSHIP STATEMENT

BH led on concept development, study design and manuscript preparation. BH and JS contributed equally to data collection and analysis. JS assisted in manuscript preparation.

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The corresponding author affirms that the manuscript is an honest, accurate, and transparent account of the study being reported; that no important aspects of the study have been omitted; and that any discrepancies from the study as planned (and, if relevant, registered) have been explained.

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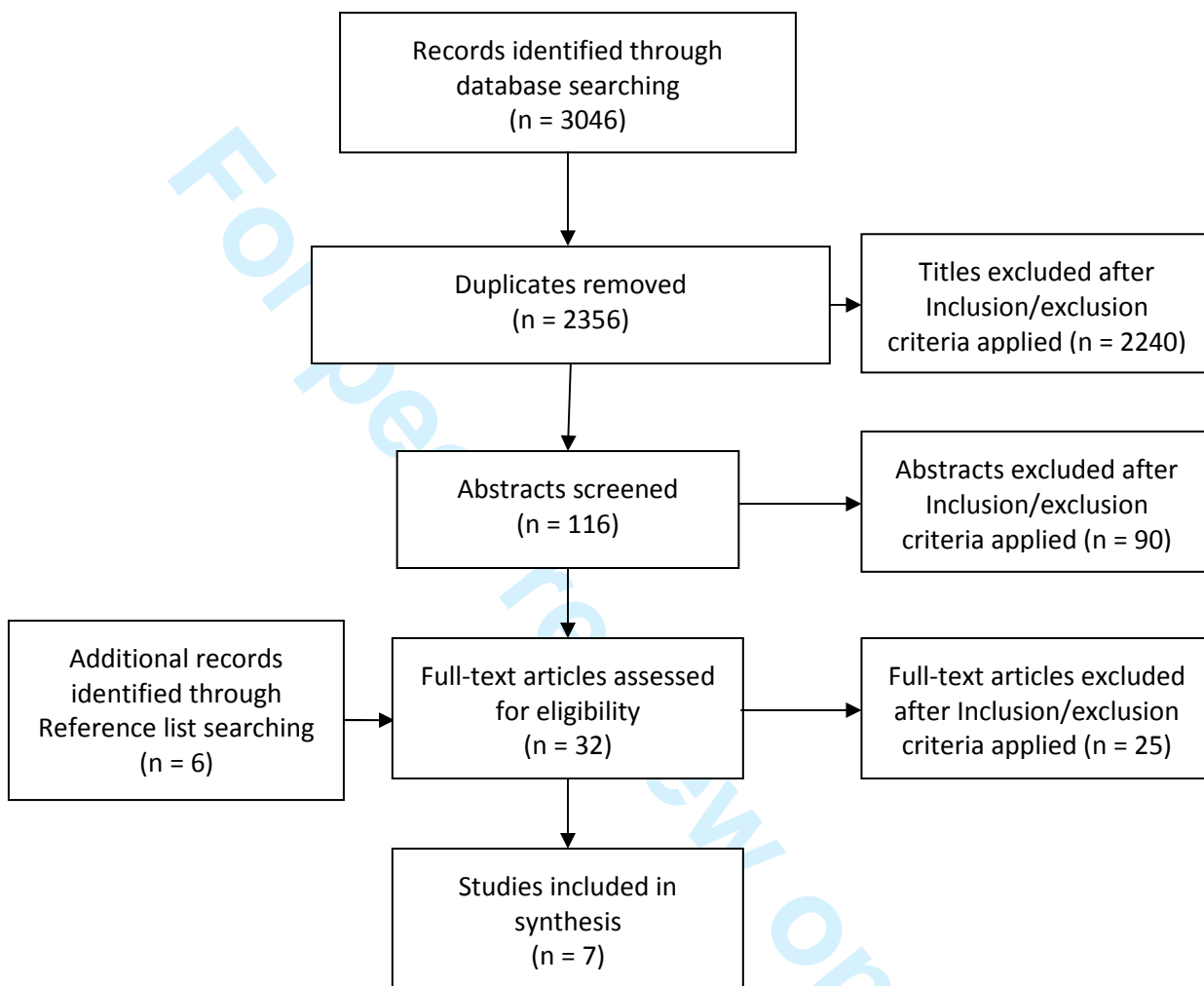
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Figure 1: PRISMA Flow Diagram



Box A – Online calculators available for predicting survival in chronic disease

The BODE Index: 4-year survival in COPD (Celli et al. 2004)

- <http://www.gxmd.com/calculate-online/respirology/bode-index;>

The Seattle Heart Failure Model: 1, 2 and 3-year survival in HF (Levy et al. 2006)

- <https://depts.washington.edu/shfm/>

Integrated Prognostic Model: 6-month mortality on haemodialysis (Cohen et al. 2010)

- <http://www.gxmd.com/calculate-online/nephrology/predicting-6-month-mortality-on-hemodialysis>

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Search strategy

Two separate searches of the published literature were performed and results combined:

1. **Combining terms for ‘life expectancy’ AND ‘patient-estimated’**
2. **Using terms for ‘prognostic understanding’**

- Medline 1950, Embase (including Cochrane) 1974, PsycINFO 1987 to present day (date of search 13th November 2015)
- Limited to English, humans, adults, 1985 to present
- Fingertip search of the reference lists of all included papers and reviews

Grey literature searching was performed using a ProQuest dissertations and theses search, the Networked Digital Library of Theses and Dissertations Global ETD search and the System for Grey Literature in Europe. The search terms ‘Life expectancy’, ‘survival’, ‘self-estimated’ and ‘patient-estimated’ were used.

Search 1: Terms for ‘Life expectancy’

Mesh

Exp Prognosis

Exp Life expectancy

Text word

Prognosis.ti,ab

Life expect\$.ti,ab

Life duration.ti,ab

Length of life.ti,ab

Duration of life.ti,ab

Days left.ti,ab

Weeks left.ti,ab

Months left.ti,ab

Years left.ti,ab

Survival benefit.ti,ab

Life left.ti,ab

Period of existence.ti,ab

Long term survival.ti,ab

Short term survival.ti,ab

Medium term survival.ti,ab

Life exten\$.ti,ab

Prognos\$ expect\$.ti,ab

Predict\$ surviv\$.ti,ab

“Within 5”

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3 (Chance\$ adj5 surviv\$).ti,ab
4 (Expect\$ adj5 alive).ti,ab
5
6 (Surviv\$ adj5 Estim\$).ti,ab
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8 (Surviv\$ adj5 probab\$).ti,ab
9
10 (Surviv\$ adj5 expect\$).ti,ab
11 (Surviv\$ adj5 Predict\$).ti,ab
12 (Estimat\$ adj5 prognosis).ti,ab
13
14 (Prognos\$ adj5 expect\$).ti,ab
15

16 17 18 **Search 1 : Terms for 'Self-estimated'**

19 **Text word**

20 Self estimat\$.ti,ab
21 Patient\$ estimat\$.ti,ab
22 Patient\$ predict\$.ti,ab
23 Patient expect\$.ti,ab
24 Self assess\$.ti,ab
25 Self forecast\$.ti,ab
26 Self generate\$.ti,ab
27 Self estimate\$.ti,ab
28 Patient\$ generat\$.ti,ab
29 Patient\$ forecast\$.ti,ab
30 Personal\$ estimat\$.ti,ab
31 Personal\$ forecast\$.ti,ab
32 Prognos\$ belie\$.ti,ab
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34 **"Within 5"**

35 (Own adj3 estimat\$).ti,ab
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37 38 39 **Search 2: Terms for 'prognostic understanding'**

40 (Prognos\$ adj5 disclos\$).mp.
41 (Perceiv\$ adj5 prognos\$).mp.
42 (Communicat\$ adj5 prognos\$).mp.
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Quality assessment tool:

- 1) Was the sample representative of patients in the general population with chronic life-limiting non-cancer disease?
 - a) Truly representative
 - b) Somewhat representative
 - c) Poorly representative or insufficient description of the of the group provided
- 2) Was the method by which the sample was identified, recruited and retained described?
 - a) Clear description/diagram illustrating recruitment, consent, exclusion, loss to follow up, death etc.
 - b) Unclear or incomplete description/diagram
 - c) Poor or no description of process provided
- 3) Were biases generated by the selection process; for example due to a very low participation rate, an all-volunteer sample or extremely restricted sampling?
 - a) Selection bias unlikely
 - b) Selection bias possible
 - c) Selection bias very likely
- 4) Was a control or comparison group available?
 - a) A well matched control/comparison group was available
 - b) A poorly matched control/comparison group was available
 - c) No control/comparison group was available
- 5) Were the measures used well-chosen to provide a serviceable assessment of self-estimation of life-expectancy?
 - a) Measures likely to provide a high quality assessment of self-estimated life expectancy
 - b) Measures moderately likely to provide a high quality assessment of self-estimated life expectancy
 - c) Measures unlikely to provide a high quality assessment of self-estimated life expectancy
- 6) Is comparator data available to provide a test of the accuracy of the patient's estimate?
 - a) Prospective collection of actual survival statistics
 - b) Use of physician estimates, predictive models, or equivalent
 - c) Disease standard survival only, or no comparator data used

Result:

For each question, A = 3, B = 2, C = 1. Mean score from reviewers. 6-9 = Low quality, 10-14= medium quality, 15-18 = high quality

Appendix B: Complete list of full papers considered

Author/Date	Title	Journal	Accepted/Rejected	Reasoning
(Allen et al. 2008)	Discordance between patient-predicted and model-predicted life expectancy among ambulatory patients with heart failure	Journal of the American Medical Association	Accepted	Meets criteria: Patients with heart failure asked how long they expect to live.
(Belkora et al. 2011)	Does use of the adjuvant! Model influence use of adjuvant therapy through better risk communication?	Journal of the National Comprehensive Cancer Network	Rejected	Patients with cancer only
(Brouwer and van Exel 2005)	Expectations regarding length and health related quality of life: some empirical findings	Social science and medicine	Rejected	Questionnaire applied to members of public, rather than individuals with chronic disease
(Chen et al. 2013)	Expectations about the effectiveness of radiation therapy among patients with incurable lung cancer	Journal of Clinical Oncology	Rejected	Patients with cancer only
(Christakis and Lamont 2000)	Extent and determinants of error in doctors' prognoses in terminally ill patients: Prospective cohort study	British Medical Journal	Rejected	Doctors, but not patients predicted life-expectancy
(Connors 1995)	A Controlled Trial to Improve Care for Seriously Ill Hospitalized Patients	Journal of the American Medical Association	Rejected	Patients only asked about the likelihood of being alive at two months
(Enzinger et al. 2013)	Outcomes of prognostic disclosure: Effects on advanced cancer patients'	Journal of Clinical Oncology	Rejected	Patients with cancer only

	prognostic understanding, mental health, and relationship with their oncologist			
(Fisher et al. 2015)	Patient characteristics associated with prognostic awareness: a study of a Canadian palliative care population using the InterRAI palliative care instrument	Journal of Pain and Symptom Management	Rejected	Whilst study reports on awareness of six month prognosis patients were not asked directly to estimate their life expectancy. Data gathered from interviewer subjective inference.
(Fried, Bradley, and O'Leary 2003)	Prognosis Communication in Serious Illness: Perceptions of Older Patients, Caregivers, and Clinicians	Journal of the American Geriatrics Society	Accepted	Meets criteria: Patients with advanced heart failure, COPD and cancer asked how long they expect to live. Authors provided additional data to permit analysis of non-cancer diagnoses alone.
(Fried, Bradley, and O'Leary 2006)	Changes in prognostic awareness among seriously ill older persons and their caregivers	Journal of Palliative Medicine	Accepted	Meets criteria: Same cohort as 2003 paper, interviewed sequentially. Authors provided additional data to permit analysis of non-cancer diagnoses alone.
(Gleason et al. 2009)	The influence of patient expectations regarding cure on treatment decisions	Patient Education & Counselling	Rejected	Patients with cancer only.
(Griffin, Loh, and Hesketh 2013)	A mental model of factors associated with subjective life expectancy	Social science and medicine	Rejected	Questionnaire applied to unselected members of the public, rather than individuals with chronic

				disease
(Gwilliam et al. 2013)	Prognosticating in patients with advanced cancer-observational study comparing the accuracy of clinicians' and patients' estimates of survival	Annals of Oncology	Rejected	Patients with cancer only
(Haidet et al. 1998)	Outcomes, preferences for resuscitation, and physician-patient communication among patients with metastatic colorectal cancer	American Journal of Medicine	Rejected	Patients with cancer only
(Kitko and Hupcey 2015)	Patients perceptions of illness severity in advanced heart failure	Heart Failure 2015 and the 2nd World Congress on Acute Heart Failure Seville Spain.	Rejected	Qualitative evidence only
(Kraai et al. 2013)	Preferences of heart failure patients in daily clinical practice: Quality of life or longevity?	European Journal of Heart Failure	Accepted	Meets criteria: Patients with advanced heart failure were asked to estimate their own life expectancy.
(Krumholz et al. 1998)	Resuscitation Preferences Among Patients With Severe Congestive Heart Failure : Results From the SUPPORT Project	Circulation	Rejected	Patients only asked about the likelihood of being alive at two months
(Le Blanc et al. 2014)	Acute myeloid leukemia (AML) patients' understanding of prognosis and treatment goals: A mixed-methods study	Journal of Clinical Oncology	Rejected	Patients with cancer only
(Lee et al. 2001)	Discrepancies between patient and physician	Journal of the American Medical Association	Rejected	Patients with cancer only

	estimates for the success of stem cell transplantation				
(Lipkus et al. 2010)	Breast cancer patients' treatment expectations after exposure to the decision aid program adjuvant online: the influence of numeracy	Medical decision making : an international journal of the Society for Medical Decision Making	Rejected		Patients with cancer only
(Lynn et al. 2000)	Living and dying with chronic obstructive pulmonary disease	Journal of the American Geriatrics Society	Rejected		Patients only asked about the likelihood of being alive at two months
(Phillips et al. 1996)	Choices of seriously ill patients about cardiopulmonary resuscitation: Correlates and outcomes	American Journal of Medicine	Rejected		Patients only asked about the likelihood of being alive at two months
(Reid et al. 2006)	Estimates of Life Expectancy by Adolescents and Young Adults With Congenital Heart Disease	Journal of the American College of Cardiology	Rejected		Patients with congenital disease only
(Sanchez-Menegay and Stalder 1994)	Do physicians take into account patients' expectations?	Journal of General Internal Medicine	Rejected		No quantitative assessment made of subjective life expectancy
(Schell et al. 2012)	Discussions of the kidney disease trajectory by elderly patients and nephrologists: a qualitative study	American Journal of Kidney Disease	Rejected		No quantitative assessment made of subjective life expectancy
(Sekeres et al. 2004)	Decision-making and quality of life in older adults with acute myeloid leukemia or advanced myelodysplastic syndrome	Leukemia	Rejected		Patients with cancer only
(Shah et al. 2006)	Estimating needs in life	Palliative medicine	Accepted		Meets criteria: Patients with

	threatening illness: A feasibility study to assess the views of patients and doctors				advanced chronic disease and cancer asked to estimate their life expectancy. Data reported separately.
(Sheldon, Fetting, and Siminoff 1993)	Offering the option of randomized clinical trials to cancer patients who overestimate their prognoses with standard therapies	Cancer Investigation		Rejected	Patients with cancer only
(Siegel, Bradley, and Kasl 2003)	Self-Rated Life Expectancy as a Predictor of Mortality: Evidence from the HRS and AHEAD Surveys	Gerontology		Rejected	Questionnaire applied to unselected members of public, rather than individuals with chronic disease
(Stewart et al. 2010)	Patient expectations from implantable defibrillators to prevent death in heart failure	Journal of Cardiac Failure		Accepted	Meets criteria: Patients with advanced heart failure asked to estimate their life expectancy.
(Wachterman et al. 2013)	Relationship between the prognostic expectations of seriously ill patients undergoing hemodialysis and their nephrologists	Journal of the American Medical Association		Accepted	Meets criteria: Patients receiving haemodialysis asked to estimate their life expectancy.
(Weeks et al. 1998)	Relationship between cancer patients' predictions of prognosis and their treatment preferences.	Journal of the American Medical Association		Rejected	Patients with cancer only

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PRISMA 2009 Checklist

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Section/topic	#	Checklist item	Reported on page #
TITLE			
Title	1	Identify the report as a systematic review, meta-analysis, or both.	1
ABSTRACT			
Structured summary	2	Provide a structured summary including, as applicable: background; objectives; data sources; study eligibility criteria, participants, and interventions; study appraisal and synthesis methods; results; limitations; conclusions and implications of key findings; systematic review registration number.	2
INTRODUCTION			
Rationale	3	Describe the rationale for the review in the context of what is already known.	4
Objectives	4	Provide an explicit statement of questions being addressed with reference to participants, interventions, comparisons, outcomes, and study design (PICOS).	4-5
METHODS			
Protocol and registration	5	Indicate if a review protocol exists, if and where it can be accessed (e.g., Web address), and, if available, provide registration information including registration number.	5
Eligibility criteria	6	Specify study characteristics (e.g., PICOS, length of follow-up) and report characteristics (e.g., years considered, language, publication status) used as criteria for eligibility, giving rationale.	4-5
Information sources	7	Describe all information sources (e.g., databases with dates of coverage, contact with study authors to identify additional studies) in the search and date last searched.	5
Search	8	Present full electronic search strategy for at least one database, including any limits used, such that it could be repeated.	Appendix A
Study selection	9	State the process for selecting studies (i.e., screening, eligibility, included in systematic review, and, if applicable, included in the meta-analysis).	5
Data collection process	10	Describe method of data extraction from reports (e.g., piloted forms, independently, in duplicate) and any processes for obtaining and confirming data from investigators.	5
Data items	11	List and define all variables for which data were sought (e.g., PICOS, funding sources) and any assumptions and simplifications made.	5
Risk of bias in individual studies	12	Describe methods used for assessing risk of bias of individual studies (including specification of whether this was done at the study or outcome level), and how this information is to be used in any data synthesis.	5 and Appendix B
Summary measures	13	State the principal summary measures (e.g., risk ratio, difference in means).	N/a – no summary

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			made
Synthesis of results	14	Describe the methods of handling data and combining results of studies, if done, including measures of consistency (e.g., I^2) for each meta-analysis.	N/a – no summary made

Page 1 of 2

Section/topic	#	Checklist item	Reported on page #
Risk of bias across studies	15	Specify any assessment of risk of bias that may affect the cumulative evidence (e.g., publication bias, selective reporting within studies).	6 and Table 1
Additional analyses	16	Describe methods of additional analyses (e.g., sensitivity or subgroup analyses, meta-regression), if done, indicating which were pre-specified.	N/a – not done
RESULTS			
Study selection	17	Give numbers of studies screened, assessed for eligibility, and included in the review, with reasons for exclusions at each stage, ideally with a flow diagram.	6 and Figure 1
Study characteristics	18	For each study, present characteristics for which data were extracted (e.g., study size, PICOS, follow-up period) and provide the citations.	Table 1
Risk of bias within studies	19	Present data on risk of bias of each study and, if available, any outcome level assessment (see item 12).	Table 1
Results of individual studies	20	For all outcomes considered (benefits or harms), present, for each study: (a) simple summary data for each intervention group (b) effect estimates and confidence intervals, ideally with a forest plot.	N/a – no intervention
Synthesis of results	21	Present results of each meta-analysis done, including confidence intervals and measures of consistency.	N/a – no meta-analysis
Risk of bias across studies	22	Present results of any assessment of risk of bias across studies (see Item 15).	N/a – not done
Additional analysis	23	Give results of additional analyses, if done (e.g., sensitivity or subgroup analyses, meta-regression [see Item 16]).	N/a – not done
DISCUSSION			
Summary of evidence	24	Summarize the main findings including the strength of evidence for each main outcome; consider their relevance to key groups (e.g., healthcare providers, users, and policy makers).	12-13
Limitations	25	Discuss limitations at study and outcome level (e.g., risk of bias), and at review-level (e.g., incomplete retrieval of identified research, reporting bias).	16

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Conclusions	26	Provide a general interpretation of the results in the context of other evidence, and implications for future research.	18
FUNDING			
Funding	27	Describe sources of funding for the systematic review and other support (e.g., supply of data); role of funders for the systematic review.	19

From: Moher D, Liberati A, Tetzlaff J, Altman DG, The PRISMA Group (2009). Preferred Reporting Items for Systematic Reviews and Meta-Analyses: The PRISMA Statement. PLoS Med 6(6): e1000097. doi:10.1371/journal.pmed1000097

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BMJ Open

How long do patients with chronic disease expect to live? A systematic review of the literature

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Manuscript ID	bmjopen-2016-012248.R1
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Primary Subject Heading:	Patient-centred medicine
Secondary Subject Heading:	Communication
Keywords:	Chronic airways disease < THORACIC MEDICINE, Heart failure < CARDIOLOGY, chronic disease, prognosis, life expectancy, chronic kidney disease

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Title

How long do patients with chronic disease expect to live?

A systematic review of the literature

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Keywords (MESH headings)

Prognosis, life expectancy, Chronic Disease, chronic kidney disease, heart failure, chronic airways disease

Word count

3,967

Abstract

Objectives: To systematically identify and summarise the literature on perceived life expectancy amongst individuals with non-cancer chronic disease.

Setting: Published and grey literature from 1985 to 2015 where adults with non-cancer chronic disease were asked to estimate their own life expectancy.

Participants: From 2472 screened titles six studies were identified that met pre-specified criteria for inclusion. Studies came from the UK, Netherlands and USA. A total of 545 subjects were included (heart failure 389; chronic obstructive pulmonary disease 89; end stage renal failure 62; chronic kidney disease 5). No papers reporting on other lung diseases, neurodegenerative disease or cirrhosis were found.

Primary and secondary outcome measures: All measures of self-estimated life expectancy were accepted. Self-estimated life expectancy was compared, where available, with observed survival, physician-estimated life expectancy and model-estimated life expectancy. Meta-analysis was not conducted due to the heterogeneity of the patient groups and study methodologies.

Results: Amongst patients with heart failure, median self-estimated life expectancy was 40% longer than predicted by a validated model. Outpatients receiving haemodialysis were more optimistic about prognosis than their nephrologists and overestimated their chances of surviving five years. Patients with heart failure and COPD were approximately three times more likely to die in the next year than they predicted. Data available for patients with chronic kidney disease were of insufficient quality to draw conclusions.

Conclusions: Individuals with chronic disease may have unrealistically optimistic expectations of their prognosis. More research is needed to understand how perceived life-expectancy affects behaviour. Meanwhile, clinicians should attempt to identify each patient's prognostic preferences and provide information in a way that they can understand and use to inform their decisions.

Trial registration: Prospero registration number: CRD42015020732

Strengths and limitations of this study

Strengths

- This is the first review of perceived life expectancy amongst patients with chronic non-cancer disease
- The findings build on and reproduce the oncology literature showing patients with cancer have a tendency to overestimate their life expectancy and chances of cure

Limitations

- The findings of this review are based on the small number of studies that have been conducted on this subject
- Literature was only available for patients with heart failure, end stage renal failure and COPD

INTRODUCTION

Chronic non-communicable disease causes more deaths worldwide than all other causes combined, with 78% due to non-cancerous conditions. Cardiovascular disease is the biggest killer and accounted for 17.5 million deaths in 2012[1]. Conditions such as heart failure and chronic kidney disease have become prevalent in higher-income countries, although there is evidence that incidence and mortality may have plateaued.[2, 3] Almost 2.3 million people in the United Kingdom (UK) have a diagnosis of coronary heart disease and over half a million have heart failure (HF).[2] An estimated 1.2 million people have a diagnosis of chronic obstructive pulmonary disease (COPD)[4] and almost 60,000 receive renal replacement therapy for end stage renal failure (ESRF)[5]. Life expectancy for patients with chronic disease; including advanced COPD, HF and ESRF can be as poor as that seen in incurable cancer.[6-8]

How long an individual expects to live – their perceived life expectancy – reflects their disease understanding and the medical profession’s ability to prognosticate for and communicate with them. Perceived life expectancy may affect a variety of outcomes, including healthcare choices. Patients with incurable lung and colon cancer who thought they were going to live for at least six months were more likely to favour life-extending therapy over comfort care compared with patients who thought there was at least a 10% chance that they would not live six months.[9] Critically unwell inpatients who do not expect to live two months are less likely to opt for cardiopulmonary resuscitation in the event of sudden death than individuals who perceive their prognosis to be better.[10]

Lately there has been a practice shift away from paternalistic medicine. Shared decision making empowers individuals and their carers to make choices about what care they want based on honest, open disclosure of the known benefits and risks of proposed treatment options.[11] So called ‘minimally disruptive medicine’ advocates a pragmatic approach to therapy in an effort to minimise

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3 treatment-burden.[12] Decisions to accept treatment with invasive therapies such as ventilation,
4
5 dialysis and implanted cardiac defibrillator placement may be influenced by how long individuals
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7 expect to live. Patients facing such decisions can only be considered fully-informed if they have an
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9 understanding of their prognosis and the effects available treatments might have upon it. Up to 38%
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11 of patients near the end of life receive treatment administered with little or no hope of it having any
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13 effect, largely because of the underlying state of the patient's health and the known or expected
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15 poor prognosis regardless of treatment.[13] In the UK almost half of adults die in a hospital bed[14]
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17 and about one in five Americans die during a hospitalisation including intensive care[15]. Quality of
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19 end-of-life care is significantly better for patients with cancer than for patients with ESRF or HF,
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21 largely due to higher rates of palliative care review and lower rates of intensive care admission and
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23 cardiopulmonary resuscitation amongst individuals with malignancy.[16] It is possible that
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25 suboptimal end of life treatment is partly driven by unrealistic expectations of prognosis.
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30 Prognosis communication has been widely studied in malignancy and a systematic review found the
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32 majority of people with cancer want detailed prognostic information, presented honestly and
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34 openly.[17] Despite this, many patients, including those with incurable malignancy, report never
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36 discussing prognosis with their healthcare team, misunderstand whether their condition is curable
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38 and overestimate their expected survival.[17] No systematic analysis of perceived life expectancy
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40 amongst individuals with non-cancer chronic disease has been performed. This review was
41
42 conducted to evaluate what is known about how long patients with non-cancer chronic disease
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44 expect to live and how these estimates compare with other methods of predicting survival and
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46 measured outcomes.
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49 **METHODS**

50 **Search strategy**

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3 A systematic search was performed of Medline, Embase, PsychINFO and the Cochrane Library.
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5 Abstracts of unpublished works were searched using ProQuest dissertations and theses search and
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7 the Networked Digital Library of Theses and Dissertations Global ETD search. Search terms relating
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9 to 'life expectancy' and 'self-estimated' were used (see Appendix A). Given the rapid changes in the
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11 demographic of patients with chronic disease and shift in attitudes and practices around sharing of
12
13 health information over the past thirty years, literature predating 1985 was deemed unlikely to
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15 inform understanding of current practice. Search results were limited to publications from 1985 to
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17 November 2015 and English Language.
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20 21 **Inclusion and exclusion criteria**

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24 Non-cancer chronic disease was defined as any long-term illness that is associated with reduced life
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26 expectancy, but not caused by cancer or infection. Conditions included were HF; chronic kidney
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28 disease stage five (CKD); ESRF receiving dialysis or conservative care; diabetes mellitus; COPD;
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30 interstitial lung disease; neurodegenerative disease and liver cirrhosis. Studies were included where
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32 adults (≥ 18 years of age) with these conditions were asked to estimate their life expectancy. All
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34 measurements of life expectancy were accepted, including those in terms of duration (e.g. "*How*
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36 *long do you expect to live*"), and chance (e.g. "*What is the chance you will be alive in five years*").
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38 Studies were excluded where only self-estimated probability of 'cure' was determined, where the
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40 only option for survival duration was less than six months and where subjects were asked to
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42 consider only hypothetical situations (e.g. "*How long do you think you would live if you had a kidney*
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44 *transplant*"). Studies reporting only on subjects with cancer, HIV/AIDS, congenital heart disease,
45
46 cystic fibrosis and organ transplant were excluded. In all these conditions the situation, illness
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48 culture or advances in treatment may have affected how generalisable findings were to the larger
49
50 chronic disease population. At the title and abstract searching phase, papers assessing prognosis in
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52 excluded diagnoses were not rejected, so that reference list searches could be performed from
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54 these papers. Where studies reported a mixture of included and excluded diagnoses, they were
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3 incorporated if the data on individual diseases were reported separately. Where data were not
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5 separately reported, authors were contacted to request supplementary files. Data were extracted
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7 from figures and tables in papers, where needed.
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10 **Study selection process**

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12 Titles were independently examined by two reviewers (BH and JS) according to the above criteria,
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14 and a Kappa statistic calculated to assess agreement. Abstracts from titles accepted by either one or
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16 both reviewers were collected and assessed independently, using the same criteria, and included if
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18 both recommended inclusion. Where only one reviewer recommended inclusion, a consensus
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20 decision was made after discussion. Full text articles were requested and read and reference lists
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22 examined with additional papers included by the same criteria. At this point, papers reporting
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24 excluded disease groups were rejected. Disagreement between authors was addressed by discussion
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26 and a consensus decision reached in all cases.
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31 **Quality assessment**

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33 No suitable tool to grade the quality of included literature could be found. A quality assessment tool
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35 (Appendix B) was developed by the authors to assess and grade the quality of available literature
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37 based on semi-objective assessment of factors influencing the generalisability, risk of bias and
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39 reporting quality of included literature. This tool has not been previously validated. Papers included
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41 for review were independently graded by the authors and a mean score taken to categorise each as
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43 low, medium or high quality. The study was registered with the PROSPERO database, registration
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45 number CRD42015020732.
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49 **RESULTS**

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51 The initial search provided 2472 titles after removal of duplicates. 116 abstracts were selected for
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53 review by either one or both authors (agree to exclude, 2356; agree include, 68; disagree, 48; Kappa
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55 0.73). 26 papers were collected and reference list searching provided an additional six. After full text
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3 examination of 32 papers, seven papers from six studies were included in the review (Figure 1). No
4 unpublished works met the inclusion criteria. A complete list of papers including reasons for
5 inclusion/rejection is available (Appendix C). Evidence was graded as medium in four and low in
6 three of the included papers (Table 1).
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11
12 Studies came from the UK[18], Netherlands[19] and USA[20-24]. A total of 545 subjects were
13 included (HF, 389; COPD, 89; ESRF, 62; CKD, 5) with study sizes ranging from 20 to 135 patients (see
14 Table 1). Four papers reported on a single medical disease; HF[19, 21, 24] and ESRF[20]. Others
15 reported on a mixture of conditions; HF and COPD[22, 23] and HF, CKD and COPD[18]. No papers
16 reporting on non-COPD lung disease, neurodegenerative disease or cirrhosis were found.
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24 The mean age of study participants ranged from 58 to 75. In the study by Fried *et al.* only individuals
25 over 60 years of age were recruited[22, 23] and only those over 50 in the study by Kraai *et al.*[19]. No
26 minimum age was set in the other studies. Two studies did not include selection criteria for disease
27 severity.[19, 21] In the other studies criteria were used to select for patients with advanced disease.
28 Patients with ESRF were all receiving outpatient haemodialysis.[20] Reported levels of comorbidity
29 were high. The mean Charlson Comorbidity index for patients with ESRF was 5.8 (SD 1.6).[20]
30 Amongst US patients with heart-failure in one study 82% had hypertension, 54% diabetes and 29%
31 COPD.[21] Amongst patients with heart failure from the Netherlands, 57% had hypertension, 30%
32 had diabetes, 24% had COPD and 11% had had a stroke.[19]
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44 One study used a written questionnaire to measure self-estimated life expectancy.[24] All other
45 studies used interviews. Participants with ESRF were asked about their chances of being alive at
46 different time points.[20] In the other studies, participants were asked to indicate how long they
47 expected to live by selecting from vignette answers,[18] giving a verbal response[21-23] and/or by
48 using a visual analogue scale.[19, 21] In one study it was not possible to ascertain how the question
49 had been posed or answered.[24] For studies where data were available, large numbers of initially
50 eligible patients were excluded from the studies, largely on the grounds of language skills or
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3 cognitive impairment (range: 88/150 (59%)[20]; 82/238 (34%)[19]; 82/361 (23%)[22, 23]; 4/44
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5 (9%)[18]), Some participants were unable or unwilling to provide a self-estimate of life expectancy
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7 (range: 56/135 (41%)[22, 23]; 26/148 (18%)[25]; 3/62 (5%)[20, 22, 23]; 0/40 (0%)[18]).
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10 Self-estimates of life expectancy were compared with predictions from clinical risk calculators[21],
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12 clinician-estimated life expectancy[18, 20, 22, 23], observed survival[18, 20-23] or presented without
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14 comparator data[19, 24]. Follow up periods ranged from one to three years and the majority of
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16 patients (range 56-73%) were alive at the end of the studies. Analysis was performed in one study to
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18 characterise factors associated with overestimation of survival.[21] In three papers patients were
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20 asked about their preferences around treatment aims, and analyses performed looking at how these
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22 responses correlated with self-estimated life expectancy.[19, 20, 24] One paper used repeat
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24 measures to examine how self-estimated life expectancy changed with disease course.[22]
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Table 1: Summary of included papers

Authors and Condition(s)	Year	Country	Quality	Design	Patients included	Measures used	Results	Summary	Pros + and cons –
Allen et al.[21] <i>Heart failure</i>	2008	USA	Medium	Cross-sectional interviewer-administered questionnaire in a single centre outpatient heart-failure service.	122 sequentially recruited subjects with heart failure (NYHA-IV) Mean age 61 (IQR 53-74) 62% male 47% African American	1) Patients were asked “If you had to guess, how much longer do you think you will live?” and completed a) Multi-choice answers ranging from <3 months to >10 years, and b) A visual analogue scale, marking their estimated age at death 2) Model-predicted life expectancy using the Seattle Heart Failure Model 3) Observed survival over median follow-up of three years	Median self-estimated life-expectancy was 13 years (IQR 8-21; range 1-54 years) Median model-predicted life-expectancy was 10 years (IQR 7.2-13.3; range 2.0-25 years) 66% of patients overestimated their survival compared with the model by 30% or more The median overestimate was 40% 29% of patients died within three years.	Self-estimated-life expectancy was on average significantly greater than that predicted by a validated model Younger age, greater disease severity and measures of less depression were independently associated with overestimation of survival	+ Efforts made to improve and check patient understanding of question – 26 of 148 enrolled participants felt unable/unwilling to estimate survival – Only 35 of 122 patients were followed up until their death – Only 9 of 122 patients had NYHA IV heart failure – No index group without chronic disease was included
Fried et al.[23] <i>COPD</i> <i>Heart failure</i>	2003	USA	Medium	Cross-sectional interview survey administered to patients registered at community practices and outpatient clinics of two hospitals, and inpatients of three hospitals. Same patient group as	135 patients with COPD or HF, aged 60 and older, meeting criteria for limited life expectancy and requiring assistance with daily living COPD – 79 patients Mean age 72 (SD 7) 51% Male 92% White HF – 56 patients Mean age 75 (SD 8) 70% Male 88% White	Patients and clinicians were asked how long they thought the patient would live and answered using multi-choice options ranging from <1 month to >10 years	Only 9 of 135 patients expected to live less than one year, but 38 patients died over this period. 58 of 79 patients who responded to being asked to estimate their own life expectancy expected to live two years or more Of the 65 available patient-clinician pairs who both responded, 34 agreed the prognosis was two years or more, 9 agreed the prognosis was two years or less, 7 clinicians thought the patient would	Patient expectations of one year mortality are higher than observed. Agreement between patients and their clinicians about likely prognosis is poor.	– 56 of 135 patients were unable or unwilling to estimate their life expectancy – No index group without chronic disease was included

				Fried <i>et al.</i> 2006			live two years or more when the patient did not expect to live this long and 15 patients expected to live two years or more when their clinician was less optimistic.			
							Kappa was 0.22 suggesting very poor agreement			
	Fried <i>et al.</i> [22]	2006	USA	Medium	Serial interview survey administered to patients registered at community practices and outpatient clinics of two hospitals, and inpatients of three hospitals.	135 patients with COPD or HF, aged 60 and older, meeting criteria for limited life expectancy and requiring assistance with daily living	Patients were asked how long they thought the patient would live and answered using multi-choice options ranging from <1 month to >10 years	9 of 59 patients who responded expected to live less than one year at their first interview. 5 of 59 expected to live less than one year at their final interview.	Patient expectations of one year mortality are higher than observed.	– 56 of 135 patients were unable or unwilling to estimate their life expectancy
	COPD					COPD – 79 patients Mean age 72 (SD 7) 51% Male 92% White		38 of 135 patients died over this period.	The majority of patients (both those who were alive and dead at the end of the year-long study) made no adjustment to their self-estimated life expectancy.	– No index group without chronic disease was included
	Heart failure				Same patient group as Fried <i>et al.</i> 2003	HF – 56 patients Mean age 75 (SD 8) 70% Male 88% White				
	Kraai <i>et al.</i> [19]	2013	The Netherlands	Low	Cross-sectional questionnaire administered in outpatient setting in one heart failure clinic.	100 patients with heart failure (NYHA I-IV) all over 50 years of age. Mean age 70 (SD 9.4) 71% male	Visual analogue scale from 50 to 100 years of age; patients were asked to indicate the most accurate estimation of their life expectancy.	Mean life expectancy indicated by patients was 82 (SD 8.6) years.	Self-estimated life expectancy probably exceeds likely outcomes, but no comparator data was available.	– No comparator prediction or measurement of survival used
	Heart failure				Sub-component of time trade-off study.			No difference in self-estimated life expectancy was found between patients unwilling vs. willing to trade time	Despite patients with more advanced or symptomatic heart failure being more willing to trade time, no difference was found between the groups in terms of expected longevity.	– Only 2 of 100 patients had NYHA IV heart failure – No index group without chronic disease was included

1 2 3 4 5 6 7 8 9 10 11 12 13 14 15 16 17 18 19 20 21 22 23 24 25 26 27 28 29 30 31	<p>Shah et al.[18]</p> <p>Heart failure</p> <p>COPD</p> <p>Chronic kidney disease</p>	2006	UK	Low	<p>Cross-sectional interviewer-administered questionnaire in outpatient and inpatient settings at one acute NHS Trust and a neighbouring Hospice.</p>	<p>20 patients in total meeting criteria for limited life expectancy: 6 HF (NYHA III/IV) 9 COPD 5 CKD</p> <p>Median age 72 50% male 85% white</p>	<p>Patients and physicians chose one of seven short prognosis statements that most accurately predicted how their illness might affect their life expectancy.</p>	<p>7 of 20 (35%) patients estimated their prognosis to be <1 year</p> <p>13/17 physicians (76%) estimated their patient's prognosis to be < 1 year</p>	<p>Exploratory study, no firm conclusions available</p>	<p>– Very small numbers</p> <p>– Sample poorly representative of a general outpatient population</p> <p>– No index group without chronic disease was included</p>
32 33 34 35 36 37 38 39 40 41 42 43 44 45 46 47 48 49	<p>Stewart et al.[24]</p> <p>Heart failure</p>	2010	USA	Low	<p>Cross-sectional written questionnaire with both inpatients and outpatients from two heart failure centres.</p> <p>Sub-component of time trade-off study.</p>	<p>105 patients with left ventricular ejection fraction (LVEF) <35% and symptomatic heart failure</p> <p>Mean age 58 (SD 13) 70% Male</p>	<p>Methodology for collecting self-estimated life expectancy not described</p>	<p>65% thought they would live more than 10 years and 34% believed they would be alive for at least 20 years.</p> <p>Patients willing to trade more time expected shorter survival than those unwilling to trade time. 46% of the patients willing to trade away at least 12 months anticipated that they would not survive 5 years.</p> <p>No difference was found in self-estimated survival between inpatients and outpatients (data not provided)</p>	<p>Self-estimated life expectancy probably exceeds likely outcomes, but no comparator data was available.</p> <p>Willingness to trade time is associated with shorter self-estimated life expectancy.</p>	<p>– No comparator prediction or measurement of survival</p> <p>– Only 3 of 105 patients had NYHA IV heart failure</p> <p>– Study methodology and tool not described</p> <p>– No index group without chronic disease was included</p>
	<p>Wachterman et al.[20]</p> <p>End stage renal failure</p>	2013	USA	Medium	<p>Cross-sectional interviewer-administered questionnaire in two community-based haemodialysis units.</p>	<p>62 patients receiving maintenance haemodialysis with 20% or greater predicted risk of dying in the next year.</p> <p>Mean age 68 (SD 10)</p>	<p>1) Patients asked what they thought their chance was of being alive at 1 and 5 years (>=90%, about 75%, about 50%, about 25%, <=10%, don't know).</p> <p>2) Nephrologist in charge of care asked to estimate each patients' chance of being alive at 1 and 5 years on a</p>	<p>For 1 year survival prediction, patients were more optimistic in 64% of patient-nephrologist pairs, whereas nephrologists were more optimistic in only 10%.</p> <p>For 5 year survival prediction, patients were more optimistic in 69%</p>	<p>Patient expectations of five year mortality are higher than observed.</p> <p>Patients were significantly more optimistic about their survival than their nephrologists.</p> <p>Patients' 1 year survival</p>	<p>– 88 of 150 eligible patients were excluded or refused to participate</p> <p>– No index group without chronic disease was included</p>

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	42% Male 52% Black	continuous scale of 0% to 100%.	patient-nephrologist pairs, whereas nephrologists were more optimistic in only 2%	expectations were more consistent with actual survival than clinician estimates.
		3) Survival data with follow up of 23 months	Only 6% of patients thought they had a less than 50% chance of being alive at 5 years, whereas actual survival at 23 months was only 56%.	Patients who expected to live longer were more likely to opt for life-extending treatments

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Self-estimated life expectancy compared with observed survival

Comparisons of self-estimated life expectancy and observed survival were reported in five papers from four studies.[18, 20-23] In general, self-estimated life expectancy exceeded observed survival. The only example of self-estimated life expectancy consistent with survival was one-year mortality in patients with ESRF.[20] 81% of patients thought they had a better than 90% chance of being alive at one year. Observed survival was 93%. In comparison, 96% of patients believed they had a better than 50% chance of being alive at five years, but 44% had died within just 23 months. In one study only 5% of patients with HF estimated their life expectancy to be three years or less, but observed mortality was 29% after a median follow-up of 3.1 years.[21] Amongst patients with advanced HF, 3 out of 56 (5%) patients expected to live less than one year, but 17 (30%) were dead in this period.[23] 6 out of 79 (8%) patients with COPD in the same study predicted their life expectancy to be less than one year; 21 (27%) died. When interviewed within the 90 days before they died, only 2 out of 16 patients predicted their life expectancy to be less than a year.[22] Patient numbers were too low in one study to draw conclusions from observed survival.[18]

Self-estimated life expectancy compared with model-predictions of survival

In the only study that used a validated model[26] to predict survival, self-estimated life expectancy exceeded model predictions.[21] Median self-estimated life expectancy for 122 patients with HF was 13 years and median model-predicted life expectancy 10 years. There was no significant relationship between self and model-predicted life expectancy. The median ratio between self-estimated and model-estimated life expectancy was 1.4; indicating a 40% overestimation. Self-estimates of life expectancy were more similar to model predictions based on age and gender alone, than to predictions taking heart disease into account.

Self-estimated compared with clinician-estimated life expectancy

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3 Four papers from three studies reported comparisons of self-estimated and clinician-estimated life
4 expectancy.[18, 20, 22, 23] Estimates agreed poorly, with a tendency for patients to be more
5
6 optimistic about life expectancy than their clinicians. Estimating one and five year survival, patients
7
8 with ESRF on dialysis were significantly more optimistic than their nephrologists.[20] Amongst
9
10 patients with COPD and HF, agreement between patients and their clinicians about whether the
11
12 patient would survive two years was poor, with a Kappa statistic of 0.22.[23] Numbers of patients in
13
14 one study were too small for any conclusions to be drawn.[18]
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17 18 19 **Other findings**

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22 Younger age, greater disease severity and lower levels of depression were independently associated
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24 with self-estimated life expectancy exceeding model predictions amongst patients with heart
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26 failure.[21] Patients receiving haemodialysis who thought they had a $\geq 90\%$ chance of being alive in 1
27
28 year were significantly more likely to choose life-extending therapy (44%) than patients who
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30 reported a $< 90\%$ chance (9%).[20] Patients with advanced COPD and HF serially interviewed over
31
32 one year showed no evidence of adjusting their self-estimated life expectancy with disease
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34 progression.[22] Only one patient of 135 revised their estimate from greater than one year to less
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36 than one year, whilst mortality was 28% over this period. Two studies found that patients with heart
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38 failure make estimates of their life expectancy that are likely to be optimistic, but did not provide
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40 any other prediction or measure of survival.[19, 24] One found patients who anticipated shorter
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42 survival to be more willing to trade longevity for improved quality of life than those who predicted
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44 longer lives.[24] The other study did not demonstrate this.[19]
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48 49 **DISCUSSION**

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52 Practice guidelines advocate considering prognosis when making decisions with patients who have
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54 chronic disease[27, 28] and promote sharing survival statistics with patients[29, 30]. There is
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56 evidence from both the cancer[22, 31, 32] and non-cancer[23, 33, 34] literature that patients with
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3 life limiting illness want open and honest communication about their prognosis. Where treatment
4 options differ markedly in survival benefit, patients require an understanding of their life expectancy
5 with each treatment to make fully-informed decisions between them. Hospitalised individuals are
6 more likely to want cardiopulmonary resuscitation if they expect to survive their illness, even if these
7 expectations are improbable.[10, 35] Patients with terminal cancer who are optimistic about their
8 prognosis are more interventional in their choice of medical therapy.[9] It is conceivable that
9 behaviours as diverse as adherence to preventative drugs and deciding whether to make a will could
10 be influenced by how long an individual expects to live.

11
12 In this systematic review of self-estimated life expectancy in chronic disease, individuals' estimates
13 exceeded nearly all predictions and measures of survival; including model-predicted and observed
14 survival. Patients with non-cancer chronic disease may have survival expectations that markedly
15 exceed outcomes. These expectations might lead some patients to make health decisions and life
16 choices that they would not if their predictions were more realistic. Patients were more optimistic
17 than their clinicians when estimating life expectancy. Only in one instance (one year survival in ESRF)
18 were patients' estimations in keeping with actual survival, and more accurate than their physicians',
19 but by two years this had reversed.[20] Whether this time-based effect represents a reproducible
20 feature of perceived vs. clinician-predicted life expectancy would require replication in other disease
21 groups. Patients with HF and COPD were approximately three times more likely to be dead within
22 the year than they predicted.[23] Life expectancy was overestimated by a median of 40% by patients
23 with heart failure, when compared with a validated model; equating to three years of life for the
24 average patient.[21] Self-estimates were more in keeping with the life expectancy of matched adults
25 without chronic disease.[21] There was evidence that no meaningful adjustment in expected survival
26 is made by patients approaching the ends of their lives.[22]

27
28 If the findings of this review reflect pervasive overestimation of life expectancy by individuals with
29 chronic disease, there are several possible explanations. Firstly, patients might never be informed

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3 that their condition could affect their life expectancy. Such individuals are likely to base survival
4 expectations on familial and media exposure, influenced by hopefulness and 'fighting spirit'. Others
5 might receive overoptimistic forecasts; either due to methods of estimation, or adjustment by the
6 communicating clinician. Finally, patients might be provided with appropriate quantitative
7 estimates, but instead, form more favourable personal predictions.
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14 These findings are compatible with the oncology literature. Most patients with cancer want to
15 discuss life expectancy, although desire for quantitative estimation varies.[36] Despite this, many
16 report not having discussed prognosis, or are found to misunderstand the status of their disease, the
17 aim of their treatment and their prognosis.[17] Overestimation of the chances of cure and survival is
18 common, even if disease is incurable and where individuals report having discussed prognosis with
19 their clinician.[37] The prognosis in non-cancer disease can be equivalently poor to that seen in
20 malignancy.[6-8] End of life care differs by diagnosis, so caution must be taken when generalising
21 findings from cancer to non-cancer disease settings.[16, 38]
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33 None of the patients with ESRF in this review recalled discussing life expectancy with their clinician;
34 their nephrologists reported they had done so with only 3% of the patients.[20] 63% of patients with
35 HF in one study did not recall having spoken with their physician about their prognosis following the
36 diagnosis of heart failure and only 36% believed HF would shorten their life.[21] Only 22% of
37 patients in one study with advanced COPD and HF recalled having been told that they could die of
38 their disease and only 1% recalled having been given an estimate of how long they might live.[23]
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45 Prognostic discussions between patients with non-cancer chronic disease and their clinicians may be
46 infrequent. In a systematic review of the literature it was found that most patients with COPD report
47 that they have never had an end of life care discussion with a healthcare provider.[39] Interviews
48 with individuals with ESRF suggest that whilst early information is beneficial, the daily focus on
49 clinical care and a reliance on clinicians to initiate end of life care discussions act as barriers to
50 advance planning.[34] Interviews with patients with ESRF and their clinicians suggest that
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3 nephrologists tend to avoid discussions about the future.[40] The evidence for prognostic
4
5 discussions between patients with cancer and their clinicians is varied.[17] Discussions are more
6
7 likely to be triggered by the clinician than the patient, and are probably infrequent amongst
8
9 individuals with advanced malignancy.[17] Where discussion occur, they are often unclear and both
10
11 parties tend to avoid acknowledging or discussing prognosis.[41] There are boundaries to clinicians
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13 initiating prognostic discussions, such as fear of causing anxiety or destroying hope[42]; uncertainty
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15 about the validity, accuracy or precision of estimates[43]; and lack of experience and training in
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17 communication skills[44].

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21 A better understanding is needed of the interaction between survival expectations and behaviour in
22
23 chronic disease. If compelling evidence is found showing overestimation of survival leads patients to
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25 make decisions out of keeping with their likely future, approaches to adjusting such expectations
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27 could be developed. Inclusion of validated methods for estimating and communicating prognosis in
28
29 decision support materials may be one way of increasing the frequency of prognostic discussions.
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31 Research into the acceptability and best methodology for facilitating these discussions should be a
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33 research priority. Some patients will not feel able to discuss prognosis, so clinicians must take care to
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35 elucidate preferences for information. However clinicians should continue to provide opportunities
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37 for prognostic discussion, since preferences may change over time and with disease progression. In
38
39 other diseases such as breast cancer, the use of prognostic models and decision tools has been
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41 shown to increase understanding of prognosis and treatment options, leading to higher degrees of
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43 satisfaction.[45] Validated tools to help predict survival in chronic disease are available[26, 46-48],
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45 but there is no evidence that these are widely employed. Only a minority are provided with
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47 accessible calculators (Box A). Studies are needed to examine how prognostic tools can be used in
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49 the clinical setting.[49] It is possible that clinical practice has not kept pace with the paradigm shift
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51 towards information-sharing with patients. Even where prognostic discussions happen, survival
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53 statistics may be misrepresented or censored.[50] In one study included in this review, nephrologists
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provided estimates of life expectancy for 89% of the interviewed patients, but reported they would withhold over half of these estimates in clinical practice.[20]

Box A – Online calculators available for predicting survival in chronic disease

The BODE Index: 4-year survival in COPD[46]

<http://www.qxmd.com/calculate-online/respirology/bode-index>

The Seattle Heart Failure Model: 1, 2 and 3-year survival in HF[26]

<https://depts.washington.edu/shfm/>

Integrated Prognostic Model: 6-month mortality on haemodialysis[47]

<http://www.qxmd.com/calculate-online/nephrology/predicting-6-month-mortality-on-hemodialysis>

The ability to make firm conclusions from the literature was highly limited by the lack of available evidence. The literature comes largely from single centre cohorts and is of medium to low-quality. Data from diseases other than COPD, heart and kidney failure is extremely limited, and those with the most advanced disease were under-represented. Included studies are likely to have come from centres where prognostication is considered important. We excluded studies including only subjects with cancer, HIV/AIDS, congenital heart disease, cystic fibrosis and organ transplant. The cancer literature has been well summarised[17], but it is possible that these excluded conditions could have provided additional insight. We are aware of only one paper that would have been included without this exclusion, showing that young adults with congenital heart disease expect to live almost as long as their healthy peers.[51]

There is no standardised or validated method for assessing self-estimated life expectancy, and it is likely that responses are influenced by methodology. Additionally, asking a patient how long they expect to live facilitates a quantitative assessment of their understanding, but does not provide information on how such perceptions are formed and influenced. Large numbers of patients were excluded from the studies or were unable or unwilling to estimate their own life expectancy, with the potential to introduce bias. In addition, many patients were excluded on grounds of language skills or cognitive impairment. These excluded individuals are likely to find discussing and understanding prognosis particularly challenging and this undermines the relevance of the included studies to a population of patients with chronic disease, in whom cognitive impairment is common.

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3 All the studies reporting actual survival were limited by short follow-up times and low numbers of
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5 deaths in the cohorts. Hospitalised patients were underrepresented in the included studies. It is
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7 feasible that survival expectations are different during periods of acute illness requiring admission;
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9 the point at which critical decisions about healthcare are often made. There is evidence to suggest
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11 that overestimation of survival persists in these situations however; both in malignant and non-
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13 malignant disease.[10, 35, 37, 52]
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16 None of the included studies had a healthy reference group. Overestimation of life expectancy
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18 cannot, therefore, be presumed a phenomenon limited to patients with disease. A prospective
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20 cohort study provides some evidence to suggest self-estimation of survival might be different
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22 amongst individuals unselected for chronic disease. Approximately half of participants made
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24 predictions of their life expectancy consistent with those from a statistical model.[53] Where
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26 predictions were inaccurate, they were approximately three times more likely to be under, than
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28 over-estimates. Overestimation increased with age, but it is unclear whether this represented an
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30 independent effect of ageing on subjective life expectancy, or confounding by the increased
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32 prevalence of disease. It is possible that general population studies of self-estimated life expectancy
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34 could be analysed for differences between individuals with and without disease.
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39 **CONCLUSION**

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42 Patients with non-cancer chronic disease may have survival expectations that markedly exceed
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44 outcomes. These expectations might lead some patients to make health decisions and life choices
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46 that they would not if their predictions were more realistic. A better understanding is needed of the
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48 interaction between survival expectations and behaviour in chronic disease. If compelling evidence is
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50 found showing overestimation of survival leads patients to make decisions out of keeping with their
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52 likely future, approaches to adjusting such expectations could be developed. Meanwhile, clinicians
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54 caring for patients with chronic disease must make attempts to elucidate what prognostic
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56 information each patient already knows, wants to know and might benefit from knowing.
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3 Appropriate information should then be shared in a form that the patient can use to inform their
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5 decisions.
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9
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11

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13
14 BH led on concept development, study design and manuscript preparation. BH and JS contributed
15 equally to data collection and analysis. JS assisted in manuscript preparation.
16

17
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19 behalf of all authors, a worldwide licence
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34 The corresponding author affirms that the manuscript is an honest, accurate, and transparent
35 account of the study being reported; that no important aspects of the study have been omitted; and
36 that any discrepancies from the study as planned (and, if relevant, registered) have been explained.
37

38 No ethical approval was required or sought for this literature review.
39

40 **COMPETING INTERESTS**

41
42 The authors declare no support from any organisation for the submitted work; no financial
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45 submitted work.
46

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50

51 **DATA SHARING STATEMENT**

52
53 All data used in the preparation of this manuscript come from published studies. No additional data
54 are available.
55

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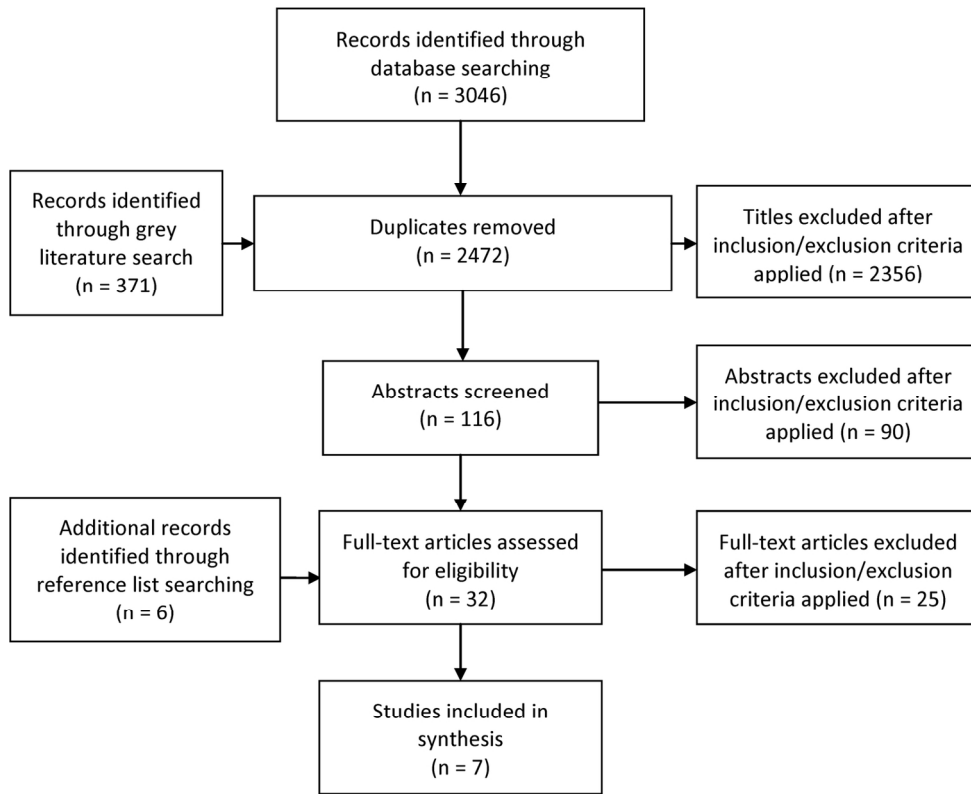


Figure 1 PRISMA Diagram
Figure 1
143x122mm (300 x 300 DPI)

Search plan

Two separate searches of published literature were performed and results combined:

1. **Combining terms for 'life expectancy' AND 'patient-estimated'**
2. **Terms for 'prognostic understanding'**

- Medline 1950, Embase (including Cochrane) 1974, PsycINFO 1987 to present day (date of search 13th November 2015)
- Limited to English, humans, adults, 1985 to present
- Fingertip search of the reference lists of all included papers and reviews on the subject

Search 1: Terms for 'Life expectancy'

Mesh

Exp Prognosis

Exp Life expectancy

Text word

Prognosis.ti,ab

Life expect\$.ti,ab

Life duration.ti,ab

Length of life.ti,ab

Duration of life.ti,ab

Days left.ti,ab

Weeks left.ti,ab

Months left.ti,ab

Years left.ti,ab

Survival benefit.ti,ab

Life left.ti,ab

Period of existence.ti,ab

Long term survival.ti,ab

Short term survival.ti,ab

Medium term survival.ti,ab

Life exten\$.ti,ab

Prognos\$ expect\$.ti,ab

Predict\$ surviv\$.ti,ab

"Within 5"

(Chance\$ adj5 surviv\$.ti,ab

(Expect\$ adj5 alive).ti,ab

(Surviv\$ adj5 Estimati\$.ti,ab

(Surviv\$ adj5 probab\$.ti,ab

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3 (Surviv\$ adj5 expect\$).ti,ab
4 (Surviv\$ adj5 Predict\$).ti,ab
5 (Estimat\$ adj5 prognosis).ti,ab
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7 (Prognos\$ adj5 expect\$).ti,ab
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10 11 **Search 1 : Terms for ‘Patient Estimated’**

12 **Text word**

13
14 Patient\$ estimat\$.ti,ab
15 Self estimat\$.ti,ab
16 Patient\$ predict\$.ti,ab
17 Patient expect\$.ti,ab
18 Self assess\$.ti,ab
19 Self forecast\$.ti,ab
20 Self generate\$.ti,ab
21 Self estimate\$.ti,ab
22 Patient\$ generat\$.ti,ab
23 Patient\$ forcast\$.ti,ab
24 Personal\$ estimat\$.ti,ab
25 Personal\$ forecast\$.ti,ab
26 Prognos\$ belie\$.ti,ab
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41 **“Within 5”**

42 (Own adj3 estimat\$).ti,ab
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50 **Search 2: Terms for ‘prognostic expectations’**

51 (Prognos\$ adj5 disclos\$).mp.
52 (Perceiv\$ adj5 prognos\$).mp.
53 (Communicat\$ adj5 prognos\$).mp.
54 (Understand\$ adj5 prognos\$).mp.
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61 **Grey literature:**

62 The grey literature was searched using ProQuest dissertations and theses search and the
63 Networked Digital Library of Theses and Dissertations Global ETD search. Databases
64 were searched from 1985-13th November 2015 for English language manuscripts where
65 the abstract contained the terms “life expectancy” and “perceived” or “self-estimated”.

Quality assessment tool:

- 1) Was the sample representative of patients in the general population with chronic life-limiting non-cancer disease?
 - a) Truly representative
 - b) Somewhat representative
 - c) Poorly representative or insufficient description of the of the group provided
- 2) Was the method by which the sample was identified, recruited and retained described?
 - a) Clear description/diagram illustrating recruitment, consent, exclusion, loss to follow up, death etc.
 - b) Unclear or incomplete description/diagram
 - c) Poor or no description of process provided
- 3) Were biases generated by the selection process; for example due to a very low participation rate, an all-volunteer sample or extremely restricted sampling?
 - a) Selection bias unlikely
 - b) Selection bias possible
 - c) Selection bias very likely
- 4) Was a control or comparison group available?
 - a) A well matched control/comparison group was available
 - b) A poorly matched control/comparison group was available
 - c) No control/comparison group was available
- 5) Were the measures used well-chosen to provide a serviceable assessment of self-estimation of life-expectancy?
 - a) Measures likely to provide a high quality assessment of self-estimated life expectancy
 - b) Measures moderately likely to provide a high quality assessment of self-estimated life expectancy
 - c) Measures unlikely to provide a high quality assessment of self-estimated life expectancy
- 6) Is comparator data available to provide a test of the accuracy of the patient's estimate?
 - a) Prospective collection of actual survival statistics
 - b) Use of physician estimates, predictive models, or equivalent
 - c) Disease standard survival only, or no comparator data used

Result:

For each question, A = 3, B = 2, C = 1. Mean score from reviewers. 6-9 = Low quality, 10-14= medium quality, 15-18 = high quality

Appendix B: Complete list of full papers considered

Author/Date	Title	Journal	Accepted/Rejected	Reasoning
(Allen et al. 2008)	Discordance between patient-predicted and model-predicted life expectancy among ambulatory patients with heart failure	Journal of the American Medical Association	Accepted	Meets criteria: Patients with heart failure asked how long they expect to live.
(Belkora et al. 2011)	Does use of the adjuvant! Model influence use of adjuvant therapy through better risk communication?	Journal of the National Comprehensive Cancer Network	Rejected	Patients with cancer only
(Brouwer and van Exel 2005)	Expectations regarding length and health related quality of life: some empirical findings	Social science and medicine	Rejected	Questionnaire applied to members of public, rather than individuals with chronic disease
(Chen et al. 2013)	Expectations about the effectiveness of radiation therapy among patients with incurable lung cancer	Journal of Clinical Oncology	Rejected	Patients with cancer only
(Christakis and Lamont 2000)	Extent and determinants of error in doctors' prognoses in terminally ill patients: Prospective cohort study	British Medical Journal	Rejected	Doctors, but not patients predicted life-expectancy
(Connors 1995)	A Controlled Trial to Improve Care for Seriously Ill Hospitalized Patients	Journal of the American Medical Association	Rejected	Patients only asked about the likelihood of being alive at two months
(Enzinger et al. 2013)	Outcomes of prognostic disclosure: Effects on advanced cancer patients'	Journal of Clinical Oncology	Rejected	Patients with cancer only

	prognostic understanding, mental health, and relationship with their oncologist				
(Fisher et al. 2015)	Patient characteristics associated with prognostic awareness: a study of a Canadian palliative care population using the InterRAI palliative care instrument	Journal of Pain and Symptom Management	Rejected		Whilst study reports on awareness of six month prognosis patients were not asked directly to estimate their life expectancy. Data gathered from interviewer subjective inference.
(Fried, Bradley, and O'Leary 2003)	Prognosis Communication in Serious Illness: Perceptions of Older Patients, Caregivers, and Clinicians	Journal of the American Geriatrics Society	Accepted		Meets criteria: Patients with advanced heart failure, COPD and cancer asked how long they expect to live. Authors provided additional data to permit analysis of non-cancer diagnoses alone.
(Fried, Bradley, and O'Leary 2006)	Changes in prognostic awareness among seriously ill older persons and their caregivers	Journal of Palliative Medicine	Accepted		Meets criteria: Same cohort as 2003 paper, interviewed sequentially. Authors provided additional data to permit analysis of non-cancer diagnoses alone.
(Gleason et al. 2009)	The influence of patient expectations regarding cure on treatment decisions	Patient Education & Counselling	Rejected		Patients with cancer only.
(Griffin, Loh, and Hesketh 2013)	A mental model of factors associated with subjective life expectancy	Social science and medicine	Rejected		Questionnaire applied to unselected members of the public, rather than individuals with chronic

				disease
(Gwilliam et al. 2013)	Prognosticating in patients with advanced cancer-observational study comparing the accuracy of clinicians' and patients' estimates of survival	Annals of Oncology	Rejected	Patients with cancer only
(Haidet et al. 1998)	Outcomes, preferences for resuscitation, and physician-patient communication among patients with metastatic colorectal cancer	American Journal of Medicine	Rejected	Patients with cancer only
(Kitko and Hupcey 2015)	Patients perceptions of illness severity in advanced heart failure	Heart Failure 2015 and the 2nd World Congress on Acute Heart Failure Seville Spain.	Rejected	Qualitative evidence only
(Kraai et al. 2013)	Preferences of heart failure patients in daily clinical practice: Quality of life or longevity?	European Journal of Heart Failure	Accepted	Meets criteria: Patients with advanced heart failure were asked to estimate their own life expectancy.
(Krumholz et al. 1998)	Resuscitation Preferences Among Patients With Severe Congestive Heart Failure : Results From the SUPPORT Project	Circulation	Rejected	Patients only asked about the likelihood of being alive at two months
(Le Blanc et al. 2014)	Acute myeloid leukemia (AML) patients' understanding of prognosis and treatment goals: A mixed-methods study	Journal of Clinical Oncology	Rejected	Patients with cancer only
(Lee et al. 2001)	Discrepancies between patient and physician	Journal of the American Medical Association	Rejected	Patients with cancer only

	estimates for the success of stem cell transplantation				
(Lipkus et al. 2010)	Breast cancer patients' treatment expectations after exposure to the decision aid program adjuvant online: the influence of numeracy	Medical decision making : an international journal of the Society for Medical Decision Making	Rejected		Patients with cancer only
(Lynn et al. 2000)	Living and dying with chronic obstructive pulmonary disease	Journal of the American Geriatrics Society	Rejected		Patients only asked about the likelihood of being alive at two months
(Phillips et al. 1996)	Choices of seriously ill patients about cardiopulmonary resuscitation: Correlates and outcomes	American Journal of Medicine	Rejected		Patients only asked about the likelihood of being alive at two months
(Reid et al. 2006)	Estimates of Life Expectancy by Adolescents and Young Adults With Congenital Heart Disease	Journal of the American College of Cardiology	Rejected		Patients with congenital disease only
(Sanchez-Menegay and Stalder 1994)	Do physicians take into account patients' expectations?	Journal of General Internal Medicine	Rejected		No quantitative assessment made of subjective life expectancy
(Schell et al. 2012)	Discussions of the kidney disease trajectory by elderly patients and nephrologists: a qualitative study	American Journal of Kidney Disease	Rejected		No quantitative assessment made of subjective life expectancy
(Sekeres et al. 2004)	Decision-making and quality of life in older adults with acute myeloid leukemia or advanced myelodysplastic syndrome	Leukemia	Rejected		Patients with cancer only
(Shah et al. 2006)	Estimating needs in life	Palliative medicine	Accepted		Meets criteria: Patients with

	threatening illness: A feasibility study to assess the views of patients and doctors				advanced chronic disease and cancer asked to estimate their life expectancy. Data reported separately.
(Sheldon, Fetting, and Siminoff 1993)	Offering the option of randomized clinical trials to cancer patients who overestimate their prognoses with standard therapies	Cancer Investigation		Rejected	Patients with cancer only
(Siegel, Bradley, and Kasl 2003)	Self-Rated Life Expectancy as a Predictor of Mortality: Evidence from the HRS and AHEAD Surveys	Gerontology		Rejected	Questionnaire applied to unselected members of public, rather than individuals with chronic disease
(Stewart et al. 2010)	Patient expectations from implantable defibrillators to prevent death in heart failure	Journal of Cardiac Failure		Accepted	Meets criteria: Patients with advanced heart failure asked to estimate their life expectancy.
(Wachterman et al. 2013)	Relationship between the prognostic expectations of seriously ill patients undergoing hemodialysis and their nephrologists	Journal of the American Medical Association		Accepted	Meets criteria: Patients receiving haemodialysis asked to estimate their life expectancy.
(Weeks et al. 1998)	Relationship between cancer patients' predictions of prognosis and their treatment preferences.	Journal of the American Medical Association		Rejected	Patients with cancer only

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PRISMA 2009 Checklist

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Section/topic	#	Checklist item	Reported on page #
TITLE			
Title	1	Identify the report as a systematic review, meta-analysis, or both.	1
ABSTRACT			
Structured summary	2	Provide a structured summary including, as applicable: background; objectives; data sources; study eligibility criteria, participants, and interventions; study appraisal and synthesis methods; results; limitations; conclusions and implications of key findings; systematic review registration number.	2
INTRODUCTION			
Rationale	3	Describe the rationale for the review in the context of what is already known.	4
Objectives	4	Provide an explicit statement of questions being addressed with reference to participants, interventions, comparisons, outcomes, and study design (PICOS).	4-5
METHODS			
Protocol and registration	5	Indicate if a review protocol exists, if and where it can be accessed (e.g., Web address), and, if available, provide registration information including registration number.	5
Eligibility criteria	6	Specify study characteristics (e.g., PICOS, length of follow-up) and report characteristics (e.g., years considered, language, publication status) used as criteria for eligibility, giving rationale.	4-5
Information sources	7	Describe all information sources (e.g., databases with dates of coverage, contact with study authors to identify additional studies) in the search and date last searched.	5
Search	8	Present full electronic search strategy for at least one database, including any limits used, such that it could be repeated.	Appendix A
Study selection	9	State the process for selecting studies (i.e., screening, eligibility, included in systematic review, and, if applicable, included in the meta-analysis).	5
Data collection process	10	Describe method of data extraction from reports (e.g., piloted forms, independently, in duplicate) and any processes for obtaining and confirming data from investigators.	5
Data items	11	List and define all variables for which data were sought (e.g., PICOS, funding sources) and any assumptions and simplifications made.	5
Risk of bias in individual studies	12	Describe methods used for assessing risk of bias of individual studies (including specification of whether this was done at the study or outcome level), and how this information is to be used in any data synthesis.	5 and Appendix B
Summary measures	13	State the principal summary measures (e.g., risk ratio, difference in means).	N/a – no summary

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PRISMA 2009 Checklist

			made
Synthesis of results	14	Describe the methods of handling data and combining results of studies, if done, including measures of consistency (e.g., I^2) for each meta-analysis.	N/a – no summary made

Page 1 of 2

Section/topic	#	Checklist item	Reported on page #
Risk of bias across studies	15	Specify any assessment of risk of bias that may affect the cumulative evidence (e.g., publication bias, selective reporting within studies).	6 and Table 1
Additional analyses	16	Describe methods of additional analyses (e.g., sensitivity or subgroup analyses, meta-regression), if done, indicating which were pre-specified.	N/a – not done
RESULTS			
Study selection	17	Give numbers of studies screened, assessed for eligibility, and included in the review, with reasons for exclusions at each stage, ideally with a flow diagram.	6 and Figure 1
Study characteristics	18	For each study, present characteristics for which data were extracted (e.g., study size, PICOS, follow-up period) and provide the citations.	Table 1
Risk of bias within studies	19	Present data on risk of bias of each study and, if available, any outcome level assessment (see item 12).	Table 1
Results of individual studies	20	For all outcomes considered (benefits or harms), present, for each study: (a) simple summary data for each intervention group (b) effect estimates and confidence intervals, ideally with a forest plot.	N/a – no intervention
Synthesis of results	21	Present results of each meta-analysis done, including confidence intervals and measures of consistency.	N/a – no meta-analysis
Risk of bias across studies	22	Present results of any assessment of risk of bias across studies (see Item 15).	N/a – not done
Additional analysis	23	Give results of additional analyses, if done (e.g., sensitivity or subgroup analyses, meta-regression [see Item 16]).	N/a – not done
DISCUSSION			
Summary of evidence	24	Summarize the main findings including the strength of evidence for each main outcome; consider their relevance to key groups (e.g., healthcare providers, users, and policy makers).	12-13
Limitations	25	Discuss limitations at study and outcome level (e.g., risk of bias), and at review-level (e.g., incomplete retrieval of identified research, reporting bias).	16

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PRISMA 2009 Checklist

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Conclusions	26	Provide a general interpretation of the results in the context of other evidence, and implications for future research.	18
FUNDING			
Funding	27	Describe sources of funding for the systematic review and other support (e.g., supply of data); role of funders for the systematic review.	19

From: Moher D, Liberati A, Tetzlaff J, Altman DG, The PRISMA Group (2009). Preferred Reporting Items for Systematic Reviews and Meta-Analyses: The PRISMA Statement. PLoS Med 6(6): e1000097. doi:10.1371/journal.pmed1000097

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BMJ Open

How long do patients with chronic disease expect to live? A systematic review of the literature

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Secondary Subject Heading:	Communication
Keywords:	Chronic airways disease < THORACIC MEDICINE, Heart failure < CARDIOLOGY, chronic disease, prognosis, life expectancy, chronic kidney disease

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Title

How long do patients with chronic disease expect to live?

A systematic review of the literature

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Prognosis, life expectancy, Chronic Disease, chronic kidney disease, heart failure, chronic airways disease

Word count

4,040

Abstract

Objective: To systematically identify and summarise the literature on perceived life expectancy amongst individuals with non-cancer chronic disease.

Setting: Published and grey literature up to and including September 2016 where adults with non-cancer chronic disease were asked to estimate their own life expectancy.

Participants: From 6837 screened titles, nine articles were identified that met pre-specified criteria for inclusion. Studies came from the UK, Netherlands and USA. A total of 729 subjects were included (heart failure 573; chronic obstructive pulmonary disease 89; end stage renal failure 62; chronic kidney disease 5). No papers reporting on other lung diseases, neurodegenerative disease or cirrhosis were found.

Primary and secondary outcome measures: All measures of self-estimated life expectancy were accepted. Self-estimated life expectancy was compared, where available, with observed survival, physician-estimated life expectancy and model-estimated life expectancy. Meta-analysis was not conducted due to the heterogeneity of the patient groups and study methodologies.

Results: Amongst patients with heart failure, median self-estimated life expectancy was 40% longer than predicted by a validated model. Outpatients receiving haemodialysis were more optimistic about prognosis than their nephrologists and overestimated their chances of surviving five years. Patients with heart failure and COPD were approximately three times more likely to die in the next year than they predicted. Data available for patients with chronic kidney disease were of insufficient quality to draw conclusions.

Conclusions: Individuals with chronic disease may have unrealistically optimistic expectations of their prognosis. More research is needed to understand how perceived life-expectancy affects behaviour. Meanwhile, clinicians should attempt to identify each patient's prognostic preferences and provide information in a way that they can understand and use to inform their decisions.

Trial registration: Prospero registration number: CRD42015020732

Strengths and limitations of this study

Strengths

- This is the first review of perceived life expectancy amongst patients with chronic non-cancer disease
- The findings build on and reproduce the oncology literature showing patients with cancer have a tendency to overestimate their life expectancy and chances of cure

Limitations

- The findings of this review are based on the small number of studies that have been conducted on this subject
- Literature was only available for patients with heart failure, end stage renal failure and COPD

INTRODUCTION

How long an individual expects to live – their perceived life expectancy – reflects their disease understanding and the medical profession’s ability to prognosticate for and communicate with them. Perceived life expectancy may affect a variety of outcomes, including healthcare choices. Patients with incurable lung and colon cancer who thought they were going to live for at least six months were more likely to favour life-extending therapy over comfort care compared with patients who thought there was at least a 10% chance that they would not live six months.[1] Critically unwell inpatients who do not expect to live two months are less likely to opt for cardiopulmonary resuscitation in the event of sudden death than individuals who perceive their prognosis to be better.[2]

Prognosis communication has been widely studied in oncology and the majority of people with cancer want detailed prognostic information, presented honestly and openly.[3] However, non-cancer chronic disease causes more deaths than cancer worldwide , with cardiovascular disease being the biggest killer.[4] Almost 2.3 million people in the United Kingdom (UK) have a diagnosis of coronary heart disease and over half a million have heart failure (HF).[5] An estimated 1.2 million people have a diagnosis of chronic obstructive pulmonary disease (COPD)[6] and almost 60,000 receive renal replacement therapy for end stage renal failure (ESRF)[7]. Life expectancy for patients with chronic disease; including advanced COPD, HF and ESRF can be as poor as that seen in incurable cancer.[8-10]

Lately there has been a practice shift away from paternalistic medicine. Shared decision making empowers individuals and their carers to make choices about what care they want based on honest, open disclosure of the known benefits and risks of proposed treatment options.[11] Decisions to accept treatment with invasive therapies such as ventilation, dialysis and implanted cardiac defibrillator placement may be influenced by how long individuals expect to live. Patients facing such

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2
3 decisions can only be considered fully-informed if they have an understanding of their prognosis and
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5 the effects available treatments might have upon it. Up to 38% of patients near the end of life
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7 receive treatment administered with little or no hope of it having any effect, largely because of the
8
9 underlying state of the patient's health and the known or expected poor prognosis regardless of
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11 treatment.[12] Quality of end-of-life care is significantly better for patients with cancer than for
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13 patients with ESRF or HF, largely due to higher rates of palliative care review and lower rates of
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15 intensive care admission and cardiopulmonary resuscitation amongst individuals with
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17 malignancy.[13] It is possible that suboptimal end of life treatment is partly driven by unrealistic
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19 expectations of prognosis.
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23 Many patients with cancer, including those with incurable disease, report never discussing prognosis
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25 with their healthcare team, misunderstand whether their condition is curable and overestimate their
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27 expected survival.[3] No systematic analysis of perceived life expectancy amongst individuals with
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29 non-cancer chronic disease has been performed. This review was conducted to evaluate what is
30
31 known about how long patients with non-cancer chronic disease expect to live and how these
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33 estimates compare with other methods of predicting survival and measured outcomes.
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36 37 **METHODS**

38 39 **Search strategy**

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42 A systematic search of Medline, Embase, PsychINFO and the Cochrane Library was conducted up to
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44 and including September 2016. Abstracts of unpublished works were searched using ProQuest
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46 dissertations and theses search and the Networked Digital Library of Theses and Dissertations Global
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48 ETD search. Search terms relating to 'life expectancy' and 'self-estimated' were used (see Appendix
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50 A). Search results were limited to humans and English language.
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53 54 **Inclusion and exclusion criteria**

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3 Non-cancer chronic disease was defined as any long-term illness that is associated with reduced life
4 expectancy, but not caused by cancer or infection. Conditions included were HF; chronic kidney
5 disease stage five (CKD); ESRF receiving dialysis or conservative care; diabetes mellitus; COPD;
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7 interstitial lung disease; neurodegenerative disease and liver cirrhosis. Studies were included where
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9 adults (≥ 18 years of age) with these conditions were asked to estimate their life expectancy. All
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11 measurements of life expectancy were accepted, including those in terms of duration (e.g. *“How*
12
13 *long do you expect to live”*), and chance (e.g. *“What is the chance you will be alive in five years”*).
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15 Studies were excluded where only self-estimated probability of ‘cure’ was determined, where the
16
17 only option for survival duration was less than six months and where subjects were asked to
18
19 consider only hypothetical situations (e.g. *“How long do you think you would live if you had a kidney*
20
21 *transplant”*). Studies reporting only on subjects with cancer, HIV/AIDS, congenital heart disease,
22
23 cystic fibrosis and organ transplant were excluded. In all these conditions the situation, illness
24
25 culture or advances in treatment may have affected how generalisable findings were to the larger
26
27 chronic disease population. At the title and abstract searching phase, articles assessing prognosis in
28
29 excluded diagnoses were not rejected, so that reference list searches could be performed from
30
31 these papers. Where studies reported a mixture of included and excluded diagnoses, they were
32
33 incorporated if the data on individual diseases were reported separately. Where data were not
34
35 separately reported, authors were contacted to request supplementary files. Data were extracted
36
37 from figures and tables in papers, where needed.
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45 **Study selection process**

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47 Titles were independently examined by two reviewers (BH and JS) according to the above criteria,
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49 and a Kappa statistic calculated to assess agreement. Abstracts from titles accepted by either one or
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51 both reviewers were collected and assessed independently, using the same criteria, and included if
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53 both recommended inclusion. Where only one reviewer recommended inclusion, a consensus
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55 decision was made after discussion. Full text articles were requested and read and reference lists
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3 examined with additional papers included by the same criteria. At this point, papers reporting
4
5 excluded disease groups were rejected. Disagreement between authors was addressed by discussion
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7 and a consensus decision reached in all cases.
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10 **Quality assessment**

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12 No suitable tool to grade the quality of included literature could be found. A quality assessment tool
13
14 (Appendix B) was developed by the authors to assess and grade the quality of available literature
15
16 based on semi-objective assessment of factors influencing the generalisability, risk of bias and
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18 reporting quality of included literature. This tool has not been previously validated. Papers included
19
20 for review were independently graded by the authors and a mean score taken to categorise each as
21
22 low, medium or high quality. The study was registered with the PROSPERO database, registration
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24 number CRD42015020732.
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28 **RESULTS**

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30 The initial search provided 6837 titles after removal of duplicates. 249 abstracts were selected for
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32 review by either one or both authors (agree to exclude, 6588; agree include, 158; disagree, 91;
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34 Kappa 0.77). Thirty-one articles were collected and reference list searching provided an additional
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36 eight. After full text examination of 39 articles, seven papers and two conference abstracts were
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38 included in the review (Figure 1). No unpublished works met the inclusion criteria. Two of the
39
40 included papers originate from a single study.[14, 15] A complete list of papers including reasons for
41
42 inclusion/rejection is available (Appendix C). Evidence was graded as medium-quality in four and
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44 low-quality in three of the included papers (Table 1). No articles were graded as high-quality. The
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46 two abstracts were not quality-assessed as insufficient information was available.
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52 **INSERT FIGURE 1 HERE**

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55 Studies came from the UK[16], Netherlands[17] and USA[14, 18-23]. A total of 729 subjects were
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57 included (HF, 573; COPD, 89; ESRF, 62; CKD, 5) with study sizes ranging from 20 to 135 patients (see
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3 Table 1). Four papers reported on a single medical disease; HF[17, 19, 21-23] and ESRF[18]. Others
4
5 reported on a mixture of conditions; HF and COPD[14, 20] and HF, CKD and COPD[16]. No papers
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7 reporting on non-COPD lung disease, neurodegenerative disease or cirrhosis were found.
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10 The mean age of study participants ranged from 58 to 75. In the study by Fried *et al.* only individuals
11
12 over 60 years of age were recruited[14, 20] and only those over 50 in the study by Kraai *et al*[17]. No
13
14 minimum age was set in the other studies. Two studies did not include selection criteria for disease
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16 severity[17, 19] and selection criteria were unreported in one study[22]. In all other studies criteria
17
18 were used to select for patients with advanced disease. Patients with ESRF were all receiving
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20 outpatient haemodialysis.[18] Reported levels of comorbidity were high. The mean Charlson
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22 Comorbidity index for patients with ESRF was 5.8 (SD 1.6).[18] Amongst US patients with heart-
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24 failure in one study 82% had hypertension, 54% diabetes and 29% COPD.[19] Amongst patients with
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26 heart failure from the Netherlands, 57% had hypertension, 30% had diabetes, 24% had COPD and
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28 11% had had a stroke.[17]
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32 One study used a written questionnaire to measure self-estimated life expectancy.[21] Methodology
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34 was unreported in two studies.[22, 23] All other studies used interviews. Participants with ESRF were
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36 asked about their chances of being alive at different time points.[18] In the other studies,
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38 participants were asked to indicate how long they expected to live by selecting from vignette
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40 answers,[16] giving a verbal response[14, 19, 20] and/or by using a visual analogue scale.[17, 19] In
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42 one study it was not possible to ascertain how the question had been posed or answered.[21] For
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44 studies where data were available, large numbers of initially eligible patients were excluded from
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46 the studies, largely on the grounds of language skills or cognitive impairment (range: 88/150
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48 (59%)[18]; 82/238 (34%)[17]; 82/361 (23%)[14, 20]; 4/44 (9%)[16]), Some participants were unable
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50 or unwilling to provide a self-estimate of life expectancy (range: 56/135 (41%)[14, 20]; 26/148
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52 (18%)[19]; 3/62 (5%)[14, 18, 20]; 0/40 (0%)[16]).
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3 Self-estimates of life expectancy were compared with predictions from clinical risk calculators[19],
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5 clinician-estimated life expectancy[14, 16, 18, 20], observed survival[14, 16, 18-20, 22] or presented
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7 without comparator data[17, 21, 23]. Follow up periods ranged from one to three years and the
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9 majority of patients (range 56-73%) were alive at the end of the studies. Analysis was performed in
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11 one study to characterise factors associated with overestimation of survival.[19] In three papers
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13 patients were asked about their preferences around treatment aims, and analyses performed
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15 looking at how these responses correlated with self-estimated life expectancy.[17, 18, 21] One paper
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17 used repeat measures to examine how self-estimated life expectancy changed with disease
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19 course.[14]
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Table 1: Summary of included articles

Reference	Conditions	Origin	Quality	Design	Patients included	Measures used	Results	Summary	Pros + and cons –
Allen et al. 2008 [19]	Heart failure	USA	Medium	Cross-sectional interviewer-administered questionnaire in a single centre outpatient heart-failure service.	122 sequentially recruited subjects with heart failure (NYHA-IV) Mean age 61 (IQR 53-74) 62% male 47% African American	1) Patients were asked “If you had to guess, how much longer do you think you will live?” and completed a) Multi-choice answers ranging from <3 months to >10 years, and b) A visual analogue scale, marking their estimated age at death 2) Model-predicted life expectancy using the Seattle Heart Failure Model 3) Observed survival over median follow-up of three years	Median self-estimated life-expectancy was 13 years (IQR 8-21; range 1-54 years) Median model-predicted life-expectancy was 10 years (IQR 7.2-13.3; range 2.0-25 years) 66% of patients overestimated their survival compared with the model by 30% or more The median overestimate was 40% 29% of patients died within three years.	Self-estimated-life expectancy was on average significantly greater than that predicted by a validated model Younger age, greater disease severity and measures of less depression were independently associated with overestimation of survival	+ Efforts made to improve and check patient understanding of question – 26 of 148 enrolled participants felt unable/unwilling to estimate survival – Only 35 of 122 patients were followed up until their death – Only 9 of 122 patients had NYHA IV heart failure – No index group without chronic disease was included
Fried et al. 2003 [20]	COPD Heart failure	USA	Medium	Cross-sectional interview and survey administered to patients registered at community practices and outpatient clinics of two hospitals, and inpatients of three hospitals. Same patient group as	135 patients with COPD or HF, aged 60 and older, meeting criteria for limited life expectancy and requiring assistance with daily living COPD – 79 patients Mean age 72 (SD 7) 51% Male 92% White HF – 56 patients	Patients and clinicians were asked how long they thought the patient would live and answered using multi-choice options ranging from <1 month to >10 years	Only 9 of 135 patients expected to live less than one year, but 38 patients died over this period. 58 of 79 patients who responded to being asked to estimate their own life expectancy expected to live two years or more Of the 65 available patient-clinician pairs who both responded, 34 agreed the prognosis was two years or more, 9 agreed the prognosis was two years or less, 7 clinicians thought the patient would	Patient expectations of one year mortality are higher than observed. Agreement between patients and their clinicians about likely prognosis is poor.	– 56 of 135 patients were unable or unwilling to estimate their life expectancy – No index group without chronic disease was included

				Fried et al. 2006	Mean age 75 (SD 8) 70% Male 88% White		live two years or more when the patient did not expect to live this long and 15 patients expected to live two years or more when their clinician was less optimistic.		
							Kappa was 0.22 suggesting very poor agreement		
Fried et al. 2006 [14]	COPD Heart failure	USA	Medium	Serial interview survey administered to patients registered at community practices and outpatient clinics of two hospitals, and inpatients of three hospitals.	135 patients with COPD or HF, aged 60 and older, meeting criteria for limited life expectancy and requiring assistance with daily living COPD – 79 patients Mean age 72 (SD 7) 51% Male 92% White HF – 56 patients Mean age 75 (SD 8) 70% Male 88% White	Patients were asked how long they thought the patient would live and answered using multi-choice options ranging from <1 month to >10 years	9 of 59 patients who responded expected to live less than one year at their first interview. 5 of 59 expected to live less than one year at their final interview. 38 of 135 patients died over this period.	Patient expectations of one year mortality are higher than observed. The majority of patients (both those who were alive and dead at the end of the year-long study) made no adjustment to their self-estimated life expectancy.	– 56 of 135 patients were unable or unwilling to estimate their life expectancy – No index group without chronic disease was included
Kraai et al. 2013 [17]	Heart failure	The Netherlands	Low	Cross-sectional questionnaire administered in outpatient setting in one heart failure clinic. Sub-component of time trade-	100 patients with heart failure (NYHA I-IV) all over 50 years of age. Mean age 70 (SD 9.4) 71% male	Visual analogue scale from 50 to 100 years of age; patients were asked to indicate the most accurate estimation of their life expectancy.	Mean life expectancy indicated by patients was 82 (SD 8.6) years. No difference in self-estimated life expectancy was found between patients unwilling vs. willing to trade time	Self-estimated life expectancy probably exceeds likely outcomes, but no comparator data was available. Despite patients with more advanced or symptomatic heart failure being more willing to trade time, no difference was found	– No comparator prediction or measurement of survival used – Only 2 of 100 patients had NYHA IV heart failure – No index group without chronic disease was included

				off study.				between the groups in terms of expected longevity.	
Shah et al. 2006 [16]	Heart failure COPD Chronic kidney disease	UK	Low	Cross-sectional interviewer-administered questionnaire in outpatient and inpatient settings at one acute NHS Trust and a neighbouring Hospice.	20 patients in total meeting criteria for limited life expectancy: 6 HF (NYHA III/IV) 9 COPD 5 CKD Median age 72 50% male 85% white	Patients and physicians chose one of seven short prognosis statements that most accurately predicted how their illness might affect their life expectancy.	7 of 20 (35%) patients estimated their prognosis to be <1 year 13/17 physicians (76%) estimated their patient's prognosis to be < 1 year	Exploratory study, no firm conclusions available	– Very small numbers – Sample poorly representative of a general outpatient population – No index group without chronic disease was included
Stewart et al. 2010 [21]	Heart failure	USA	Low	Cross-sectional written questionnaire with both inpatients and outpatients from two heart failure centres. Sub-component of time trade-off study.	105 patients with left ventricular ejection fraction (LVEF) <35% and symptomatic heart failure Mean age 58 (SD 13) 70% Male	Methodology for collecting self-estimated life expectancy not described	65% thought they would live more than 10 years and 34% believed they would be alive for at least 20 years. Patients willing to trade more time expected shorter survival than those unwilling to trade time. 46% of the patients willing to trade away at least 12 months anticipated that they would not survive 5 years. No difference was found in self-estimated survival between inpatients and outpatients (data not provided)	Self-estimated life expectancy probably exceeds likely outcomes, but no comparator data was available. Willingness to trade time is associated with shorter self-estimated life expectancy.	– No comparator prediction or measurement of survival – Only 3 of 105 patients had NYHA IV heart failure – Study methodology and tool not described – No index group without chronic disease was included
Wachterman et al. 2013 [18]	End stage renal failure	USA	Medium	Cross-sectional interviewer-administered questionnaire in two community-	62 patients receiving maintenance haemodialysis with 20% or greater predicted risk of	1) Patients asked what they thought their chance was of being alive at 1 and 5 years (>=90%, about 75%, about 50%, about 25%, <=10%, don't know).	For 1 year survival prediction, patients were more optimistic in 64% of patient-nephrologist pairs, whereas nephrologists were more optimistic in only 10%.	Patient expectations of five year mortality are higher than observed. Patients were significantly more optimistic about their	– 88 of 150 eligible patients were excluded or refused to participate – No index group without chronic

				based haemodialysis units.	dying in the next year. Mean age 68 (SD 10) 42% Male 52% Black	2) Nephrologist in charge of care asked to estimate each patients' chance of being alive at 1 and 5 years on a continuous scale of 0% to 100%. 3) Survival data with follow up of 23 months	For 5 year survival prediction, patients were more optimistic in 69% patient-nephrologist pairs, whereas nephrologists were more optimistic in only 2% Only 6% of patients thought they had a less than 50% chance of being alive at 5 years, whereas actual survival at 23 months was only 56%.	survival than their nephrologists. Patients' 1 year survival expectations were more consistent with actual survival than clinician estimates. Patients who expected to live longer were more likely to opt for life-extending treatments	disease was included
Ambardekar et al. 2016 [22] (abstract only)	Heart failure	USA	Not rated	Cross-sectional report of self-estimated life expectancy. Methodology not reported. Sub-component of multi-centre prospective cohort study.	161 ambulatory patients with advanced HF from 10 American centres	1) Patient self-assessment of life expectancy 2) Outcomes at mean follow-up of 13 months Methodology for data collection not described	64% of patients identified by a physician to have 'high-risk' heart failure estimated a life expectancy of greater than two years. 40% died, were transplanted or required a mechanical left-ventricular assist device over a mean follow-up of 13 months	Patients expectations of outcome were optimistic compared with physician-predicted or observed outcomes	+ Multicentre prospective cohort - Abstract only at time of review - No index group without chronic disease was included
O'Donnell et al. 2015 [23] (abstract only)	Heart failure	USA	Not rated	Self-assessment of prognosis in single centre cohort of hospitalised patients with HF. Methodology incompletely reported.	23 subjects Mean age 73 66% Male 77% White	Patient self-assessment of life expectancy	70% of patients estimated a life expectancy of greater than 5 years 43% of patients estimated a life expectancy of greater than 10 years	Self-estimated life expectancy probably exceeds likely outcomes, but no comparator data was available. Patients who did not want to discuss prognosis all expected to live >10 years	- Very small numbers - Abstract only at time of review

Self-estimated life expectancy compared with observed survival

Comparisons of self-estimated life expectancy and observed survival were reported in five papers from four studies[14, 16, 18-20] and one abstract[22]. In general, self-estimated life expectancy exceeded observed survival. The only example of self-estimated life expectancy consistent with survival was one-year mortality in patients with ESRF.[18] 81% of patients thought they had a better than 90% chance of being alive at one year. Observed survival was 93%. In comparison, 96% of patients believed they had a better than 50% chance of being alive at five years, but 44% had died within just 23 months. In one study only 5% of patients with HF estimated their life expectancy to be three years or less, but observed mortality was 29% after a median follow-up of 3.1 years.[19] Amongst patients with advanced HF, 3 out of 56 (5%) patients expected to live less than one year, but 17 (30%) were dead in this period.[20] 6 out of 79 (8%) patients with COPD in the same study predicted their life expectancy to be less than one year; 21 (27%) died. When interviewed within the 90 days before they died, only 2 out of 16 patients predicted their life expectancy to be less than a year.[14] In the study published only as an abstract, 64% of patients with heart failure expected to live for longer than two years, but at a mean follow-up of 13 months 40% had died, been transplanted or required a left-ventricular assist device.[22] Patient numbers were too low in one study to draw conclusions from observed survival.[16]

Self-estimated life expectancy compared with model-predictions of survival

In the only study that used a validated model[24] to predict survival, self-estimated life expectancy exceeded model predictions.[19] Median self-estimated life expectancy for 122 patients with HF was 13 years and median model-predicted life expectancy 10 years. There was no significant relationship between self and model-predicted life expectancy. The median ratio between self-estimated and model-estimated life expectancy was 1.4; indicating a 40% overestimation. Self-estimates of life expectancy were more similar to model predictions based on age and gender alone, than to predictions taking heart disease into account.

Self-estimated life expectancy compared with clinician-estimated life expectancy

Four papers from three studies reported comparisons of self-estimated and clinician-estimated life expectancy.[14, 16, 18, 20] Estimates agreed poorly, with a tendency for patients to be more optimistic about life expectancy than their clinicians. Estimating one and five year survival, patients with ESRF on dialysis were significantly more optimistic than their nephrologists.[18] Amongst patients with COPD and HF, agreement between patients and their clinicians about whether the patient would survive two years was poor, with a Kappa statistic of 0.22.[20] Numbers of patients in one study were too small for any conclusions to be drawn.[16]

Other findings

Younger age, greater disease severity and lower levels of depression were independently associated with self-estimated life expectancy exceeding model predictions amongst patients with heart failure.[19] Patients receiving haemodialysis who thought they had a $\geq 90\%$ chance of being alive in 1 year were significantly more likely to choose life-extending therapy (44%) than patients who reported a $< 90\%$ chance (9%).[18] Patients with advanced COPD and HF serially interviewed over one year showed no evidence of adjusting their self-estimated life expectancy with disease progression.[14] Only one patient of 135 revised their estimate from greater than one year to less than one year, whilst mortality was 28% over this period. Three studies found that patients with heart failure make estimates of their life expectancy that are likely to be optimistic, but did not present any other validated prediction or measure of survival.[17, 21, 23] One found patients who anticipated shorter survival to be more willing to trade longevity for improved quality of life than those who predicted longer lives.[21] The other study did not demonstrate this.[17] One study was published only as an abstract and had insufficient numbers of patients to draw conclusions.[23]

DISCUSSION

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3 Practice guidelines advocate considering prognosis when making decisions with patients who have
4 chronic disease[25, 26] and promote sharing survival statistics with patients[27, 28]. There is
5 evidence from both the cancer[14, 29, 30] and non-cancer[20, 31, 32] literature that patients with
6 life limiting illness want open and honest communication about their prognosis. Where treatment
7 options differ markedly in survival benefit, patients require an understanding of their life expectancy
8 with each treatment to make fully-informed decisions between them. Hospitalised individuals are
9 more likely to want cardiopulmonary resuscitation if they expect to survive their illness, even if these
10 expectations are improbable.[2, 33] Patients with terminal cancer who are optimistic about their
11 prognosis are more interventional in their choice of medical therapy.[1] It is conceivable that
12 behaviours as diverse as adherence to preventative drugs and deciding whether to make a will could
13 be influenced by how long an individual expects to live.

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28 In this systematic review of self-estimated life expectancy in chronic disease, individuals' estimates
29 exceeded nearly all predictions and measures of survival; including model-predicted and observed
30 survival. Patients with non-cancer chronic disease may have survival expectations that markedly
31 exceed outcomes. These expectations might lead some patients to make health decisions and life
32 choices that they would not if their predictions were more realistic. Patients were more optimistic
33 than their clinicians when estimating life expectancy. Only in one instance (one year survival in ESRF)
34 were patients' estimations in keeping with actual survival, and more accurate than their physicians',
35 but by two years this had reversed.[18] Whether this time-based effect represents a reproducible
36 feature of perceived vs. clinician-predicted life expectancy would require replication in other disease
37 groups. Patients with HF and COPD were approximately three times more likely to be dead within
38 the year than they predicted.[20] Life expectancy was overestimated by a median of 40% by patients
39 with heart failure, when compared with a validated model; equating to three years of life for the
40 average patient.[19] Self-estimates were more in keeping with the life expectancy of matched adults
41 without chronic disease.[19] There was evidence that no meaningful adjustment in expected survival
42 is made by patients approaching the ends of their lives.[14]

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3 If the findings of this review reflect pervasive overestimation of life expectancy by individuals with
4 chronic disease, there are several possible explanations. Firstly, patients might never be informed
5 that their condition could affect their life expectancy. Such individuals are likely to base survival
6 expectations on familial and media exposure, influenced by hopefulness and 'fighting spirit'. Others
7 might receive overoptimistic forecasts; either due to methods of estimation, or adjustment by the
8 communicating clinician. Finally, patients might be provided with appropriate quantitative
9 estimates, but instead, form more favourable personal predictions.
10

11
12 These findings are compatible with the oncology literature. Most patients with cancer want to
13 discuss life expectancy, although desire for quantitative estimation varies.[34] Despite this, many
14 report not having discussed prognosis, or are found to misunderstand the status of their disease, the
15 aim of their treatment and their prognosis.[3] Overestimation of the chances of cure and survival is
16 common, even if disease is incurable and where individuals report having discussed prognosis with
17 their clinician.[35] The prognosis in non-cancer disease can be equivalently poor to that seen in
18 malignancy.[8-10] End of life care differs by diagnosis, so caution must be taken when generalising
19 findings from cancer to non-cancer disease settings.[13, 36]
20

21
22 None of the patients with ESRF in this review recalled discussing life expectancy with their clinician;
23 their nephrologists reported they had done so with only 3% of the patients.[18] 63% of patients with
24 HF in one study did not recall having spoken with their physician about their prognosis following the
25 diagnosis of heart failure and only 36% believed HF would shorten their life.[19] Only 22% of
26 patients in one study with advanced COPD and HF recalled having been told that they could die of
27 their disease and only 1% recalled having been given an estimate of how long they might live.[20]
28

29
30 Prognostic discussions between patients with non-cancer chronic disease and their clinicians may be
31 infrequent. In a systematic review of the literature it was found that most patients with COPD report
32 that they have never had an end of life care discussion with a healthcare provider.[37] Interviews
33 with individuals with ESRF suggest that whilst early information is beneficial, the daily focus on
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3 clinical care and a reliance on clinicians to initiate end of life care discussions act as barriers to
4
5 advance planning.[32] Interviews with patients with ESRF and their clinicians suggest that
6
7 nephrologists tend to avoid discussions about the future.[38] The evidence for prognostic
8
9 discussions between patients with cancer and their clinicians is varied.[3] Discussions are more likely
10
11 to be triggered by the clinician than the patient, and are probably infrequent amongst individuals
12
13 with advanced malignancy.[3] Where discussion occur, they are often unclear and both parties tend
14
15 to avoid acknowledging or discussing prognosis.[39] There are boundaries to clinicians initiating
16
17 prognostic discussions, such as fear of causing anxiety or destroying hope[40]; uncertainty about the
18
19 validity, accuracy or precision of estimates[41]; and lack of experience and training in
20
21 communication skills[42].
22
23

24
25 A better understanding is needed of the interaction between survival expectations and behaviour in
26
27 chronic disease. If compelling evidence is found showing overestimation of survival leads patients to
28
29 make decisions out of keeping with their likely future, approaches to adjusting such expectations
30
31 could be developed. Inclusion of validated methods for estimating and communicating prognosis in
32
33 decision support materials may be one way of increasing the frequency of prognostic discussions.
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35 Research into the acceptability and best methodology for facilitating these discussions should be a
36
37 research priority. Some patients will not feel able to discuss prognosis, so clinicians must take care to
38
39 elucidate preferences for information. However clinicians should continue to provide opportunities
40
41 for prognostic discussion, since preferences may change over time and with disease progression. In
42
43 other diseases such as breast cancer, the use of prognostic models and decision tools has been
44
45 shown to increase understanding of prognosis and treatment options, leading to higher degrees of
46
47 satisfaction.[43] Validated tools to help predict survival in chronic disease are available[24, 44-46],
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49 but there is no evidence that these are widely employed. Only a minority are provided with
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51 accessible calculators (Box A). Studies are needed to examine how prognostic tools can be used in
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53 the clinical setting.[47] It is possible that clinical practice has not kept pace with the paradigm shift
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55 towards information-sharing with patients. Even where prognostic discussions happen, survival
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3 statistics may be misrepresented or censored.[48] In one study included in this review, nephrologists
4 provided estimates of life expectancy for 89% of the interviewed patients, but reported they would
5 withhold over half of these estimates in clinical practice.[18]
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10 **BOX A INSERTED HERE**

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12 The ability to make firm conclusions from the literature was highly limited by the lack of available
13 evidence. The literature comes largely from single centre cohorts and is of medium to low-quality.
14
15 Data from diseases other than heart failure is extremely limited, and those with the most advanced
16 disease were under-represented. Included studies are likely to have come from centres where
17 prognostication is considered important. We excluded studies including only subjects with cancer,
18 HIV/AIDS, congenital heart disease, cystic fibrosis and organ transplant. The cancer literature has
19 been well summarised[3], but it is possible that these excluded conditions could have provided
20 additional insight. We are aware of only one paper that would have been included without this
21 exclusion, showing that young adults with congenital heart disease expect to live almost as long as
22 their healthy peers.[49]
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35 There is no standardised or validated method for assessing self-estimated life expectancy, and it is
36 likely that responses are influenced by methodology. Additionally, asking a patient how long they
37 expect to live facilitates a quantitative assessment of their understanding, but does not provide
38 information on how such perceptions are formed and influenced. Large numbers of patients were
39 excluded from the studies or were unable or unwilling to estimate their own life expectancy, with
40 the potential to introduce bias. In addition, many patients were excluded on grounds of language
41 skills or cognitive impairment. These excluded individuals are likely to find discussing and
42 understanding prognosis particularly challenging and this undermines the relevance of the included
43 studies to a population of patients with chronic disease, in whom cognitive impairment is common.
44
45 All the studies reporting actual survival were limited by short follow-up times and low numbers of
46 deaths in the cohorts. Hospitalised patients were underrepresented in the included studies. It is
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feasible that survival expectations are different during periods of acute illness requiring admission; the point at which critical decisions about healthcare are often made. There is evidence to suggest that overestimation of survival persists in these situations however; both in malignant and non-malignant disease.[2, 33, 35, 50]

None of the included studies had a healthy reference group. Overestimation of life expectancy cannot, therefore, be presumed a phenomenon limited to patients with disease. A recently published prospective cohort study provides some evidence to suggest self-estimation of survival might be different amongst individuals unselected for chronic disease. Approximately half of participants made predictions of their life expectancy consistent with those from a statistical model.[51] Where predictions were inaccurate, they were approximately three times more likely to be under, than over-estimates. Overestimation increased with age, but it is unclear whether this represented an independent effect of ageing on subjective life expectancy, or confounding by the increased prevalence of disease. It is possible that general population studies of self-estimated life expectancy could be analysed for differences between individuals with and without disease.

CONCLUSION

Patients with non-cancer chronic disease may have survival expectations that markedly exceed outcomes. These expectations might lead some patients to make health decisions and life choices that they would not if their predictions were more realistic. A better understanding is needed of the interaction between survival expectations and behaviour in chronic disease. If compelling evidence is found showing overestimation of survival leads patients to make decisions out of keeping with their likely future, approaches to adjusting such expectations could be developed. Meanwhile, clinicians caring for patients with chronic disease must make attempts to elucidate what prognostic information each patient already knows, wants to know and might benefit from knowing. Appropriate information should then be shared in a form that the patient can use to inform their decisions.

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3 **INSERT BOX B HERE**
4

5
6 **ACKNOWLEDGEMENTS**
7

8 Dr T Fried and team for sharing detailed data from their studies.
9

10 **CONTRIBUTORSHIP STATEMENT**
11

12 BH led on concept development, study design and manuscript preparation. BH and JS contributed
13 equally to data collection and analysis. JS assisted in manuscript preparation.
14

15 The Corresponding Author (BH) has the right to grant on behalf of all authors and does grant on
16 behalf of all authors, a worldwide licence
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26 located; and, vi) licence any third party to do any or all of the above.
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31 The corresponding author affirms that the manuscript is an honest, accurate, and transparent
32 account of the study being reported; that no important aspects of the study have been omitted; and
33 that any discrepancies from the study as planned (and, if relevant, registered) have been explained.
34

35 No ethical approval was required or sought for this literature review.
36

37 **COMPETING INTERESTS**
38

39 The authors declare no support from any organisation for the submitted work; no financial
40 relationships with any organisations that might have an interest in the submitted work in the
41 previous three years, no other relationships or activities that could appear to have influenced the
42 submitted work.
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44

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46

47 Unfunded research
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49 **DATA SHARING STATEMENT**
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51 All data used in the preparation of this manuscript come from published studies. No additional data
52 are available.
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55 **REFERENCE LIST**
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Figure 1: PRISMA Diagram

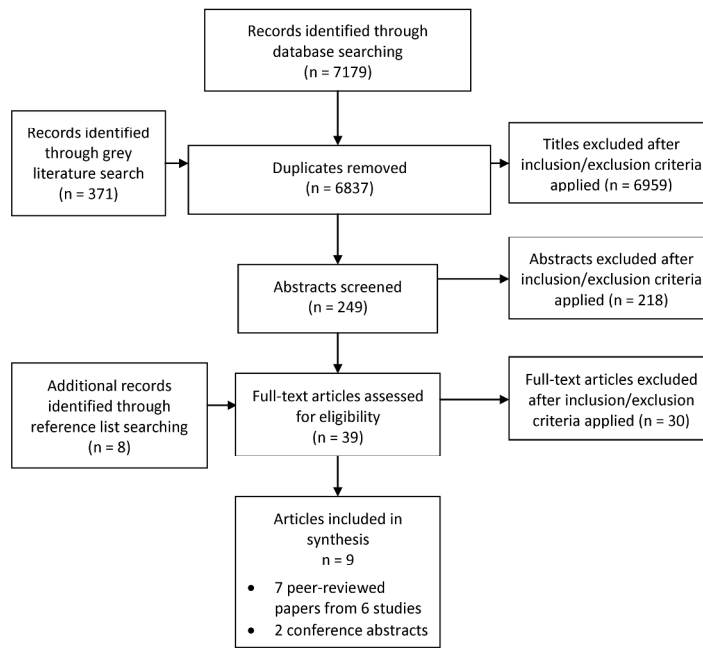


Figure 1: PRISMA diagram
Figure 1
279x361mm (300 x 300 DPI)

Box A – Online calculators available for predicting survival in chronic disease

The BODE Index: 4-year survival in COPD (Celli et al. 2004)

- <http://www.gxmd.com/calculate-online/respirology/bode-index;>

The Seattle Heart Failure Model: 1, 2 and 3-year survival in HF (Levy et al. 2006)

- <https://depts.washington.edu/shfm/>

Integrated Prognostic Model: 6-month mortality on haemodialysis (Cohen et al. 2010)

- <http://www.gxmd.com/calculate-online/nephrology/predicting-6-month-mortality-on-hemodialysis>

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What we know

- The majority of patients want prognostic information, presented honestly and openly, but such conversations do not happen routinely
- How long someone expects to live can affect their healthcare decisions
- Individuals with cancer have a tendency to overestimate their prognosis and misunderstand their chances of cure

What this paper adds

- Published data on survival expectations of individuals with non-cancer chronic disease are limited
- Patients with non-cancer chronic disease may have unrealistically optimistic expectations of their prognosis
- Individuals who overestimate their prognosis may make decisions that they would not if their expectations were more realistic

Search plan

Combining terms for 'life expectancy' AND 'self-estimated'

- Medline 1946, Embase (including Cochrane) 1974, PsycINFO 1806 to present day (date of search 3rd October 2016)
- Limited to English, humans
- Fingertip search of the reference lists of all included papers and reviews on the subject

Terms for 'life expectancy'

Mesh

Exp Prognosis

Exp Life expectancy

Text word

Prognosis.ti,ab

Life expect\$.ti,ab

Life duration.ti,ab

Length of life.ti,ab

Duration of life.ti,ab

Days left.ti,ab

Weeks left.ti,ab

Months left.ti,ab

Years left.ti,ab

Survival benefit.ti,ab

Life left.ti,ab

Period of existence.ti,ab

Long term survival.ti,ab

Short term survival.ti,ab

Medium term survival.ti,ab

Life exten\$.ti,ab

Prognos\$ expect\$.ti,ab

Predict\$ surviv\$.ti,ab

"Within 5"

(Chance\$ adj5 surviv\$.ti,ab

(Expect\$ adj5 alive).ti,ab

(Surviv\$ adj5 Estim\$.ti,ab

(Surviv\$ adj5 probab\$.ti,ab

(Surviv\$ adj5 expect\$.ti,ab

(Surviv\$ adj5 Predict\$.ti,ab

(Estimat\$ adj5 prognosis).ti,ab

(Prognos\$ adj5 expect\$).ti,ab

Terms for 'self-estimated'

Text word

Patient\$ estimat\$.ti,ab

Self estimat\$.ti,ab

Patient\$ predict\$.ti,ab

Patient expect\$.ti,ab

Self assess\$.ti,ab

Self forecast\$.ti,ab

Self generate\$.ti,ab

Self estimate\$.ti,ab

Patient\$ generat\$.ti,ab

Patient\$ forecast\$.ti,ab

Personal\$ estimat\$.ti,ab

Personal\$ forecast\$.ti,ab

Prognos\$ belie\$.ti,ab

(Prognos\$ adj5 disclos\$).mp.

(Perceiv\$ adj5 prognos\$).mp.

(Communicat\$ adj5 prognos\$).mp.

(Understand\$ adj5 prognos\$).mp.

"Within 5"

(Own adj3 estimat\$).ti,ab

Grey literature:

The grey literature was searched using ProQuest dissertations and theses search and the Networked Digital Library of Theses and Dissertations Global ETD search. Databases were searched for English language manuscripts where the abstract contained the terms "life expectancy" and "perceived" or "self-estimated".

Quality assessment tool:

- 1) Was the sample representative of patients in the general population with chronic life-limiting non-cancer disease?
 - a) Truly representative
 - b) Somewhat representative
 - c) Poorly representative or insufficient description of the of the group provided
- 2) Was the method by which the sample was identified, recruited and retained described?
 - a) Clear description/diagram illustrating recruitment, consent, exclusion, loss to follow up, death etc.
 - b) Unclear or incomplete description/diagram
 - c) Poor or no description of process provided
- 3) Were biases generated by the selection process; for example due to a very low participation rate, an all-volunteer sample or extremely restricted sampling?
 - a) Selection bias unlikely
 - b) Selection bias possible
 - c) Selection bias very likely
- 4) Was a control or comparison group available?
 - a) A well matched control/comparison group was available
 - b) A poorly matched control/comparison group was available
 - c) No control/comparison group was available
- 5) Were the measures used well-chosen to provide a serviceable assessment of self-estimation of life-expectancy?
 - a) Measures likely to provide a high quality assessment of self-estimated life expectancy
 - b) Measures moderately likely to provide a high quality assessment of self-estimated life expectancy
 - c) Measures unlikely to provide a high quality assessment of self-estimated life expectancy
- 6) Is comparator data available to provide a test of the accuracy of the patient's estimate?
 - a) Prospective collection of actual survival statistics
 - b) Use of physician estimates, predictive models, or equivalent
 - c) Disease standard survival only, or no comparator data used

Result:

For each question, A = 3, B = 2, C = 1. Mean score from reviewers. 6-9 = Low quality, 10-14= medium quality, 15-18 = high quality

Appendix B: Complete list of full papers considered

Author/Date	Title	Journal	Accepted/Rejected	Reasoning
(Allen et al. 2008)	Discordance between patient-predicted and model-predicted life expectancy among ambulatory patients with heart failure	Journal of the American Medical Association	Accepted	Meets criteria: Patients with heart failure were asked how long they expect to live.
(Ambardekar et al. 2016)	Conflicting Perceptions of Prognosis and Treatment Options between Physicians and Patients with Advanced Heart Failure: Results From the Medical Arm of Mechanically Assisted Circulatory Support (Medamacs) Registry	Journal of Cardiac Failure	Accepted, abstract only	Meets criteria: Patients with heart failure were asked how long they expect to live.
(Belkora et al. 2011)	Does use of the adjuvant! Model influence use of adjuvant therapy through better risk communication?	Journal of the National Comprehensive Cancer Network	Rejected	Patients with cancer only
(Brouwer and van Exel 2005)	Expectations regarding length and health related quality of life: some empirical findings	Social science and medicine	Rejected	Questionnaire applied to members of public, rather than individuals with chronic disease
(Chen et al. 2013)	Expectations about the effectiveness of radiation therapy among patients with incurable lung cancer	Journal of Clinical Oncology	Rejected	Patients with cancer only
(Christakis and Lamont 2000)	Extent and determinants of error in doctors' prognoses	British Medical Journal	Rejected	Doctors, but not patients predicted life-expectancy

	in terminally ill patients: Prospective cohort study				
(Connors 1995)	A Controlled Trial to Improve Care for Seriously Ill Hospitalized Patients	Journal of the American Medical Association	Rejected		Patients only asked about the likelihood of being alive at two months
(Edwards and Baharani 2015)	Beyond Believing- Thoughts on End of Life from Haemodialysis Patients at the End of Life	Nephrology Dialysis Transplantation	Rejected		No quantitative data for self-estimated life expectancy made
(Enzinger et al. 2013)	Outcomes of prognostic disclosure: Effects on advanced cancer patients' prognostic understanding, mental health, and relationship with their oncologist	Journal of Clinical Oncology	Rejected		Patients with cancer only
(Fisher et al. 2015)	Patient characteristics associated with prognostic awareness: a study of a Canadian palliative care population using the InterRAI palliative care instrument	Journal of Pain and Symptom Management	Rejected		Whilst study reports on awareness of six month prognosis patients were not asked directly to estimate their life expectancy. Data gathered from interviewer subjective inference.
(Fried, Bradley, and O'Leary 2003)	Prognosis Communication in Serious Illness: Perceptions of Older Patients, Caregivers, and Clinicians	Journal of the American Geriatrics Society	Accepted		Meets criteria: Patients with advanced heart failure, COPD and cancer asked how long they expect to live. Authors provided additional data to permit analysis of non-cancer diagnoses alone.

(Fried, Bradley, and O'Leary 2006)	Changes in prognostic awareness among seriously ill older persons and their caregivers	Journal of Palliative Medicine	Accepted	Meets criteria: Same cohort as 2003 paper, interviewed sequentially. Authors provided additional data to permit analysis of non-cancer diagnoses alone.
(Gleason et al. 2009)	The influence of patient expectations regarding cure on treatment decisions	Patient Education & Counselling	Rejected	Patients with cancer only.
(Griffin, Loh, and Hesketh 2013)	A mental model of factors associated with subjective life expectancy	Social science and medicine	Rejected	Questionnaire applied to unselected members of the public, rather than individuals with chronic disease
(Gwilliam et al. 2013)	Prognosticating in patients with advanced cancer-observational study comparing the accuracy of clinicians' and patients' estimates of survival	Annals of Oncology	Rejected	Patients with cancer only
(Haidet et al. 1998)	Outcomes, preferences for resuscitation, and physician-patient communication among patients with metastatic colorectal cancer	American Journal of Medicine	Rejected	Patients with cancer only
(Kitko and Hupcey 2015)	Patients perceptions of illness severity in advanced heart failure	Heart Failure 2015 and the 2nd World Congress on Acute Heart Failure Seville Spain.	Rejected	Qualitative evidence only
(Kraai et al. 2013)	Preferences of heart failure patients in daily clinical practice: Quality of life or	European Journal of Heart Failure	Accepted	Meets criteria: Patients with advanced heart failure were asked to estimate their own

	longevity?				life expectancy.
(Krumholz et al. 1998)	Resuscitation Preferences Among Patients With Severe Congestive Heart Failure : Results From the SUPPORT Project	Circulation	Rejected		Patients only asked about the likelihood of being alive at two months
(Le Blanc et al. 2014)	Acute myeloid leukemia (AML) patients' understanding of prognosis and treatment goals: A mixed-methods study	Journal of Clinical Oncology	Rejected		Patients with cancer only
(Lee et al. 2001)	Discrepancies between patient and physician estimates for the success of stem cell transplantation	Journal of the American Medical Association	Rejected		Patients with cancer only
(Lipkus et al. 2010)	Breast cancer patients' treatment expectations after exposure to the decision aid program adjuvant online: the influence of numeracy	Medical decision making : an international journal of the Society for Medical Decision Making	Rejected		Patients with cancer only
(Lynn et al. 2000)	Living and dying with chronic obstructive pulmonary disease	Journal of the American Geriatrics Society	Rejected		Patients only asked about the likelihood of being alive at two months
(O'Donnell et al. 2015)	Need to Elicit Patient Preferences for Information About Limited Prognosis in Heart Failure	Journal of Cardiac Failure	Accepted, abstract only		Meets criteria: Patients with advanced heart failure were asked to estimate how long they expect to live
(O'Donnell et al. 2003)	Preferences for cardiopulmonary resuscitation among patients 80 years or older: The views of patients and their	Journal of the American Medical Directors Association	Rejected		Patients only asked about the likelihood of being alive at two months

	physicians				
(Phillips et al. 1996)	Choices of seriously ill patients about cardiopulmonary resuscitation: Correlates and outcomes	American Journal of Medicine	Rejected – note multiple sub-studies of the SUPPORT study were rejected during title/abstract searching phase		In the SUPPORT study patients were only asked about the likelihood of being alive at two months
(Reid et al. 2006)	Estimates of Life Expectancy by Adolescents and Young Adults With Congenital Heart Disease	Journal of the American College of Cardiology	Rejected		Patients with congenital disease only
(Sanchez-Menegay and Stalder 1994)	Do physicians take into account patients' expectations?	Journal of General Internal Medicine	Rejected		No quantitative assessment made of subjective life expectancy
(Schell et al. 2012)	Discussions of the kidney disease trajectory by elderly patients and nephrologists: a qualitative study	American Journal of Kidney Disease	Rejected		No quantitative assessment made of subjective life expectancy
(Sekeres et al. 2004)	Decision-making and quality of life in older adults with acute myeloid leukemia or advanced myelodysplastic syndrome	Leukemia	Rejected		Patients with cancer only
(Shah et al. 2006)	Estimating needs in life threatening illness: A feasibility study to assess the views of patients and doctors	Palliative medicine	Accepted		Meets criteria: Patients with advanced chronic disease and cancer asked to estimate their life expectancy. Data reported separately.
(Sheldon, Fetting, and Siminoff 1993)	Offering the option of randomized clinical trials to cancer patients who overestimate their prognoses with standard	Cancer Investigation	Rejected		Patients with cancer only

	therapies			
(Siegel, Bradley, and Kasl 2003)	Self-Rated Life Expectancy as a Predictor of Mortality: Evidence from the HRS and AHEAD Surveys	Gerontology	Rejected	Questionnaire applied to unselected members of public, rather than individuals with chronic disease
(Stewart et al. 2010)	Patient expectations from implantable defibrillators to prevent death in heart failure	Journal of Cardiac Failure	Accepted	Meets criteria: Patients with advanced heart failure asked to estimate their life expectancy.
(Van Der Wal et al. 2016)	Heart failure patients' future expectations and their association with disease severity, quality of life, depressive symptoms and clinical outcomes	International Journal of Clinical Practice	Rejected	Qualitative data only – patients were not directly asked to quantitatively estimate their own survival
(Wachterman et al. 2013)	Relationship between the prognostic expectations of seriously ill patients undergoing hemodialysis and their nephrologists	Journal of the American Medical Association	Accepted	Meets criteria: Patients receiving haemodialysis asked to estimate their life expectancy.
(Weeks et al. 1998)	Relationship between cancer patients' predictions of prognosis and their treatment preferences.	Journal of the American Medical Association	Rejected	Patients with cancer only

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Section/topic	#	Checklist item	Reported on page #
TITLE			
Title	1	Identify the report as a systematic review, meta-analysis, or both.	1
ABSTRACT			
Structured summary	2	Provide a structured summary including, as applicable: background; objectives; data sources; study eligibility criteria, participants, and interventions; study appraisal and synthesis methods; results; limitations; conclusions and implications of key findings; systematic review registration number.	2
INTRODUCTION			
Rationale	3	Describe the rationale for the review in the context of what is already known.	4
Objectives	4	Provide an explicit statement of questions being addressed with reference to participants, interventions, comparisons, outcomes, and study design (PICOS).	4-5
METHODS			
Protocol and registration	5	Indicate if a review protocol exists, if and where it can be accessed (e.g., Web address), and, if available, provide registration information including registration number.	5
Eligibility criteria	6	Specify study characteristics (e.g., PICOS, length of follow-up) and report characteristics (e.g., years considered, language, publication status) used as criteria for eligibility, giving rationale.	4-5
Information sources	7	Describe all information sources (e.g., databases with dates of coverage, contact with study authors to identify additional studies) in the search and date last searched.	5
Search	8	Present full electronic search strategy for at least one database, including any limits used, such that it could be repeated.	Appendix A
Study selection	9	State the process for selecting studies (i.e., screening, eligibility, included in systematic review, and, if applicable, included in the meta-analysis).	5
Data collection process	10	Describe method of data extraction from reports (e.g., piloted forms, independently, in duplicate) and any processes for obtaining and confirming data from investigators.	5
Data items	11	List and define all variables for which data were sought (e.g., PICOS, funding sources) and any assumptions and simplifications made.	5
Risk of bias in individual studies	12	Describe methods used for assessing risk of bias of individual studies (including specification of whether this was done at the study or outcome level), and how this information is to be used in any data synthesis.	5 and Appendix B
Summary measures	13	State the principal summary measures (e.g., risk ratio, difference in means).	N/a – no summary

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			made
Synthesis of results	14	Describe the methods of handling data and combining results of studies, if done, including measures of consistency (e.g., I^2) for each meta-analysis.	N/a – no summary made

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Section/topic	#	Checklist item	Reported on page #
Risk of bias across studies	15	Specify any assessment of risk of bias that may affect the cumulative evidence (e.g., publication bias, selective reporting within studies).	6 and Table 1
Additional analyses	16	Describe methods of additional analyses (e.g., sensitivity or subgroup analyses, meta-regression), if done, indicating which were pre-specified.	N/a – not done
RESULTS			
Study selection	17	Give numbers of studies screened, assessed for eligibility, and included in the review, with reasons for exclusions at each stage, ideally with a flow diagram.	6 and Figure 1
Study characteristics	18	For each study, present characteristics for which data were extracted (e.g., study size, PICOS, follow-up period) and provide the citations.	Table 1
Risk of bias within studies	19	Present data on risk of bias of each study and, if available, any outcome level assessment (see item 12).	Table 1
Results of individual studies	20	For all outcomes considered (benefits or harms), present, for each study: (a) simple summary data for each intervention group (b) effect estimates and confidence intervals, ideally with a forest plot.	N/a – no intervention
Synthesis of results	21	Present results of each meta-analysis done, including confidence intervals and measures of consistency.	N/a – no meta-analysis
Risk of bias across studies	22	Present results of any assessment of risk of bias across studies (see Item 15).	N/a – not done
Additional analysis	23	Give results of additional analyses, if done (e.g., sensitivity or subgroup analyses, meta-regression [see Item 16]).	N/a – not done
DISCUSSION			
Summary of evidence	24	Summarize the main findings including the strength of evidence for each main outcome; consider their relevance to key groups (e.g., healthcare providers, users, and policy makers).	12-13
Limitations	25	Discuss limitations at study and outcome level (e.g., risk of bias), and at review-level (e.g., incomplete retrieval of identified research, reporting bias).	16

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Conclusions	26	Provide a general interpretation of the results in the context of other evidence, and implications for future research.	18
FUNDING			
Funding	27	Describe sources of funding for the systematic review and other support (e.g., supply of data); role of funders for the systematic review.	19

From: Moher D, Liberati A, Tetzlaff J, Altman DG, The PRISMA Group (2009). Preferred Reporting Items for Systematic Reviews and Meta-Analyses: The PRISMA Statement. PLoS Med 6(6): e1000097. doi:10.1371/journal.pmed1000097

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