Defining priorities in prognostication research: results of a consensus workshop

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ABSTRACT

Purpose

To establish consensus among palliative care researchers on the priorities for prognostication research.

Methods

A nominal group technique was employed involving palliative care researchers attending a workshop within a scientific meeting on prognostication. Participants worked in small facilitated groups to generate future research questions which were amalgamated and rated according to importance.

Results

Twenty-five meeting delegates took part in the workshop including 10 palliative care physicians and four nurses, one dietician, and 10 academic researchers, all of whom had experience and/or interest in prognosis research. A total of 40 research questions were generated and after prioritisation ratings, the top five questions were: 1) How valid are prognostic tools? =2) Can we use prognostic criteria as entry criteria for research? =2) How do we judge the impact of a prognostic score in clinical practice? 4) What is the best way of presenting survival data to patients? 5) What is the most user-friendly validated tool?

Conclusions

Although a wide range of research questions relating to prognostication were identified, the strongest priority to emerge from the consensus data concerned the validity of prognostic tools. Further research to validate existing tools is essential to ensure their clinical value.

INTRODUCTION

Prognostication, the prediction of the course or outcome of a disease, has various purposes. In the clinical context it is used to inform individuals about the future course of illness and to guide treatment decision-making. At a personal level it aids patients in organising their affairs at the end of life. In policy terms it can determine eligibility for relevant benefits or access to resources. In research settings prognostic estimates may be used to select relevant patients for particular studies. Recently, we have also seen how prognostication can be involved in political decisions with the case of Abdelbaset Ali al-Megrahi. The convicted Lockerbie bomber was released from prison on compassionate grounds when estimated to have three months left to live.[1]

However, despite the wide use of prognostic estimates, accurate predictions are notoriously difficult. Subjective approaches based on doctors' clinical judgement have low correlations with actual outcomes.[2] A comparison of physician estimates and actual survival times for 300 terminally-ill patients with cancer showed a significant discrepancy (median of 75 days versus 26 days).[3] More objective approaches involve the use of actuarial data to make predictions from single variables (e.g. performance status, presence of symptoms, or laboratory parameters) or combinations of factors. Although numerous formal prediction tools have been developed, most are not adequately validated, and are unreliable in practice.[4] Given the important implications of prognostication for patient care and policy, there is a clear need for carefully designed research on this subject.

The Cancer Experiences Collaborative (CECo) Methodology theme organised a State of the Science meeting on prognostication within supportive and palliative care in order to bring together health professionals and researchers to discuss the current state of the evidence in prognostication and the future research agenda. An afternoon workshop was arranged using a consensus method with the aim of defining research priorities in the field.

METHOD

Participants

Twenty-five delegates took part in the workshop. These included ten palliative care physicians, four nurses, one dietician and ten academic researchers. All had involvement in palliative care research and experience and/or interest in the subject of prognostication.

Design

A nominal group technique was used for eliciting and ranking research questions in order to establish a level of consensus on future priorities. Nominal group techniques have a long history of use in health and medicine, providing a structured format for discussion and synthesis of information, with the aim of rapidly producing solutions or decisions.[5]

Procedure

The meeting involved presentations by five experienced researchers within the prognostication area (Table 1) each followed by an open discussion among delegates. Subsequently, workshop participants were allocated to four facilitated groups of 6-7 people. Facilitators asked participants to spend 10 minutes individually writing down potential research questions relating to prognostication based on what they had learnt from the presentations and their own experiences. These were reported one by one to the facilitator who wrote them all on a flipchart. Group discussion was then initiated to clarify and rephrase questions where necessary. The lists of questions from the four groups were amalgamated, duplicate items were removed, and the final list was presented to all individuals. Participants were asked to score up to ten items according to perceived importance with the highest priority scored 10, the second highest 9, and so on.

Analysis

Scores from all participants were summed for each question and totals sorted into descending order. The maximum possible score for questions was 250 (25 participants scoring 10). In three instances, two questions were deemed to address an identical topic. In the first two cases (Q16/Q28 and Q20/Q35), all participants had rated one or the other item, but not both, allowing the two totals to be summed. In the third case (Q23/Q40), the same procedure was used except with three participants who had rated both items and a mean of the two scores was used.

RESULTS

A total of forty research questions were generated between the four groups (Table 2) and were presented to all participants for ranking. Totals were calculated for all questions and the highest 15 are presented in Figure 1. Five questions emerged as having distinctly higher scores than the others and these priority questions are presented in Table 3.

DISCUSSION

The aim of the prioritisation exercise was to establish a consensus among a group of palliative care clinicians and researchers on the most important questions to be addressed relating to prognostication.

The highest priority question that emerged relates to the validity of prognostic tools. This is clearly vital with regard to the clinical value of such instruments. Although a particular model might successfully predict outcomes in one sample of patients, this is of little real use if it does not also do so in other samples.[6] There are many cases in the literature of prognostic tools that have been developed but not adequately validated.[7] However, there are also examples of prognostic indices that have demonstrated good generalisability such as the Palliative

Performance Scale,[8-10] and the Palliative Prognostic Score.[11-14] Nonetheless, the reviews of the state of the science of prognostication presented at the meeting, clearly revealed the need for further development and validation of tools to assist clinicians in their prognostic decision making.

One example of ongoing research to develop and validate a prognostic tool is the Prognosis in Palliative Care Study (PIPS).[15] PIPS aims to develop a new prognostic index to improve, or possibly replace, clinician estimates of survival for palliative care patients. PIPS also aims to determine whether repeated prognostic assessments are more accurate than single assessments and to determine whether self-reported symptom severity provides better prognostic accuracy than observer-rated symptom severity in competent palliative care patients. PIPS is a prospective multicentre observational study of consecutive eligible referrals to participating palliative care units and collects a range of socio-demographic, clinical, performance and self-report measures that have been demonstrated to be predictors or possible predictors of survival. Recruitment closed at the end of July 2009, and analysis is underway at time of writing. Ultimately it is envisaged that the resulting instrument will be used in clinical practice to aid decision-making.

The next two highest ranked research questions received equal scores. One was directly research oriented (use of prognostic scores as eligibility criteria for research) and the other an applied issue (the impact of a prognostic score on clinical practice).

Obtaining a well-defined sample is one of the challenges of research in palliative care. A comparison of hospital doctors and general practitioners indicated a discrepancy in 20% of cases regarding the palliative status of patients.[16] Many studies involving patients at the end of life rely on physician judgement for determining eligibility. The inherent variability in the accuracy of these judgements can lead to participants dying earlier than expected and not completing the study, or in other cases surviving longer than expected and retrospectively proving ineligible.[17] If the use of more objective criteria (i.e. a prognostic score from a validated tool) provides more reliable entry criteria it would lead to an enhancement of the overall quality of palliative care studies.

Evaluating the impact of a prognostic score on clinical practice is of clear importance since there would be little value in developing and validating tools if they do not lead to improvements in practice. Impact may be measured in terms of physician decision-making, patient outcome, and cost-effectiveness.[18] The potential value of using a predictive tool depends on how its predictions are translated into decisions that are applied to patient care. Despite the large number of predictive tools developed within different healthcare domains, few have been formally evaluated in impact studies and implemented in daily practice. Recent guidance on the process and methodological considerations for impact analysis[18 19] should help in the development and assessment of prognostic tools that will ultimately lead to greater quality and cost-effectiveness of care.

The fourth priority concerned the best way of presenting survival data to patients. Existing research indicates that patients vary in the degree of detail desired from their doctors regarding their prognosis, and do not always fully comprehend the information given.[20 21] The risk is that incomplete understanding can lead to sub-optimal decision-making by patients and subsequently poor outcomes. In one study, patients who believed they had at least a 90% chance of living for ≥6 months were more than twice as likely to favour life-extending therapy over palliative care than those who thought they were less likely to live that long. However, adjusted analyses revealed no difference in 6-month survival between the two groups, while those choosing life-extending therapy were more likely to experience adverse events (e.g. readmission, resuscitation or death while receiving ventilatory assistance).[22] These findings suggest that improving patient understanding of their prognosis may help avoid unnecessary suffering at the end of life.

A study of the information preferences of patients with metastatic cancer found that information presented as words and numbers were favoured over charts or graphs, with over 80% wanting to know statistical information (e.g. longest survival time with treatment, 5-year survival rates, and average survival).[23] The optimal ways of communicating such information have not been determined. There is a body of research exploring the effects of information framing on patient decisions. One widely cited study found that participants were more likely to opt (hypothetically) for a riskier treatment option (surgery over radiation) for lung cancer when presented in a positive frame (e.g. 90% chance of survival) than a negative frame (e.g. 10% chance of mortality).[24] However, a review of similar studies found no consistent pattern of patient decision-making based on how information was framed.[25] In an interview study with health professionals, doctors reported that they tried to provide survival estimates as ranges or proportions in lay language to avoid patients focussing on absolute figures and to maintain the right mix of realism and hope.[26] Clinical practice guidelines for prognostic and end-of-life communication have been published based on existing evidence.[27] However, the reliance on expert opinion in places where research was lacking highlights the need for investigation of this important subject.

The fifth priority identified relates to identifying the most user-friendly validated tool. This links directly with the first priority of validating tools. Many simple predictive indices or nomograms exist that are undoubtedly user-friendly, but may not have been formally validated. Equally, a model with high reliability and validity may not be useful in practice if it is difficult or time-consuming to administer or interpret by busy clinicians. A systematic review of ten prognostic tools in palliative care found the validity data promising for several instruments.[28] However some tools were burdensome to use due to the large number of variables to take into account or the need for laboratory tests. Furthermore, continued research to refine tools may identify additional variables or suggest that repeated measures are necessary to improve predictive accuracy which would further increase the complexity of the task. One of the challenges in developing and validating predictive tools is ensuring that they are also sufficiently user-friendly to be of practical value.

Establishing consensus among a particular group of individuals on an issue is not an attempt to provide a ‘correct’ answer. Rather it is a systematic way of identifying current opinion and areas of disagreement.[29] The twenty-five delegates taking part in the current task are not necessarily representative of the nation’s experts in this area, and it is important to recognise that a different group of prognostication researchers may have produced an alternative set of priorities. However, the participants did represent a broad range of disciplines, levels of research and clinical experience and seniority, and a broad geographical spread. The presentations and discussions which preceded the consensus exercise may also have had important effects on the judgments of the members of the groups. The presentations provided overviews of the state of the science from recognised experts in the field. Inevitably these experts will have highlighted specific issues based on their knowledge and experience. And the ensuing discussions were inevitably led by individuals with specific views, although chairing ensured an open debate. Whether this consensus procedure introduces bias, as opposed to simply being informed debate, is of course an issue for all consensus methods. This can be partially guarded against through transparency of the consensus decision-making process and clarity of evidence presented. In the present case transparency has been ensured by following standard procedures and reporting them openly, and by making the presentations publically available on the internet.[30]

None of the research questions came close to receiving unanimous maximum ratings (i.e. a score of 250) and the small difference between the total scores of the top five items indicates that no questions were considered of outstanding priority. Instead, several issues of broadly similar importance emerged. Undoubtedly, prognostication is a complex subject with implications within several diverse domains (e.g. personal, clinical, policy, research).

In summary, a wide range of research questions relating to prognostication were identified, but the strongest priority to emerge from the consensus data concerned the validity of prognostic tools. The importance of this issue lies in the ability of a measure to accurately predict outcomes beyond the original sample, in order that it can have wider clinical value. The ongoing PIPS study is a good example of an attempt to develop and validate a prognostic tool for use in palliative care.

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Table 1. Speaker presentations (available at http://www.ceco.org.uk/resourcedocs/prognostication.pdf)[30]

Speaker	**Affiliation**	**Title of presentation**
Professor Chris Todd	School of Nursing, Midwifery, and Social Work, University of Manchester, Manchester, United Kingdom	Why might we want to prognosticate?
Dr Paul Glare	Memorial Sloan Kettering Cancer Centre, New York, United States	Prognosis in advanced care: a core clinical skill for the 21st century
Professor Karel GM Moons	Julius Center for Health Sciences and Primary Care, University Medical Centre Utrecht, Netherlands	Prognostic research: state of the science and future challenges
Dr Patrick Stone	St George's Hospital Medical School, London, United Kingdom	Practical difficulties with undertaking prognostic research in palliative care patients: lessons from the PiPS study
Dr Michael Downing	Victoria Hospice, Victoria, British Columbia, Canada	Implementing a prognostic indicator in clinical practice

Table 2. Research questions generated through four discussion groups

Question number	Research question
Q1	How variable is validity between centres?
Q2	How do we judge the impact of a prognostic score in clinical practice?
Q3	Is choice of death important in prognostic indices?
Q4	Is socioeconomic status important to prognosis predictions?
Q5	How valid are prognostication tools?
Q6	What is the most user-friendly validated tool?
Q7	Are there psychosocial or spiritual factors that influence prognosis?
Q8	Does 'tone' of care influence prognosis for older people with dementia?
Q9	Could prognostic tools be adapted for children?
Q10	Are the Gold Standard Framework prognostic indicators validated?
Q11	Can prognostication tools be used in non-cancer populations?
Q12	Could prognostication tools be used as a trigger to initiate palliative care referral?
Q13	Can downstream tools be used upstream?
Q14	What sentinel physical or emotional events alter prognosis?
Q15	How do prognosis estimates differ between clinicians, carers, and patients?
Q16	What do clinicians use to estimate prognosis? [combined with Q28]
Q17	How do we increase awareness in non-palliative care physicians?
Q18	Does being given prognosis information increase/decrease survival?
Q19	What is the optimum time period to measure change for prediction?
Q20	Can we use prognostic scores as entry criteria for research? [combined with Q35]
Q21	What are the main disease trajectories?
Q22	How do the predictions of different grades of professionals and multi-disciplinary teams compare in relation to survival?
Q23	What are the preferred methods of presenting information to patients and others? [combined with Q40]
Q24	How do prognosis predictions influence patient views?
Q25	How does the understanding and priorities of patients change over time?
Q26	What is the impact of prognosticating on patients, outcomes, costs, and service delivery?
Q27	What are the experiences of patients in the disclosure of prognosis?
Q28	What are clinicians considering when they make prognostic judgements? [combined with Q16]
Q29	Would the routine use of a prognostic tool in hospice settings affect informed communication?
Q30	Does improving symptoms lead to changes in prognosis?
Q31	What are the information needs of patients and families regarding survival predictions?
Q32	What other health outcomes beyond survival do palliative care patients rate as important?
Q33	How well do tools compare with clinical judgements?
Q34	Would it be possible to predict disease stage?
Q35	Can prognostic models be used to determine research study eligibility criteria? [combined with Q20]
Q36	What are the risks/benefits for patients and health-care professionals of disclosing prognosis?
Q37	What do patients really mean when they ask 'how long do I have?'
Q38	Who wants to know about prognosis and why?
Q39	Do predictive models change clinical decisions?
Q40	What is the best way to present survival data to patients? [combined with Q23]

Table 3. Five research priorities emerging from consensus exercise

Rank	Research question	Score
1	How valid are prognostic tools? (Q5)	83
=2	Can we use prognostic criteria as entry criteria for research? (Q2)	79
=2	How do we judge the impact of a prognostic score in clinical practice? (Q16)	79
4	What is the best way of presenting survival data to patients? (Q23)	73.5
5	What is the most user-friendly validated tool? (Q6)	73

Figure 1. Fifteen highest ranked questions