ISSN 1178-2293 (Online)

University of Otago
Economics Discussion Papers
No. 2003

APRIL 2020

Creating a priority list of non-communicable diseases to support health research funding decision-making

Saeideh Babashahi,[1]* Paul Hansen[1,2] & Trudy Sullivan[3]

[1] Department of Economics, University of Otago
[2] 1000minds Ltd, www.1000minds.com
3 Department of Preventive & Social Medicine, University of Otago

Address for correspondence:

Saeideh Babashahi
Department of Economics
University of Otago
PO Box 56
Dunedin
NEW ZEALAND
email: saeideh.babashahi@postgrad.otago.ac.nz
Telephone: 64 3 479 8645

Creating a priority list of non-communicable diseases to support health research funding decision-making

Saeideh Babashahi,[1]* Paul Hansen[1,2] & Trudy Sullivan[3]

[1] Department of Economics, University of Otago
[2] 1000minds Ltd, www.1000minds.com
3 Department of Preventive & Social Medicine, University of Otago

* *Corresponding author:*

Saeideh Babashahi
Department of Economics
University of Otago
Dunedin
New Zealand
email: saeideh.babashahi@postgrad.otago.ac.nz
ph: +64 3 479 8645

Abstract

To develop and pilot a framework based on multi-criteria decision analysis (MCDA) for creating a priority list of non-communicable diseases (NCDs) to support health research funding decisions, where the NCDs are prioritised with respect to their overall burden to society. The framework involves identifying NCDs to be prioritised, specifying prioritisation criteria and determining their weights from a survey of stakeholders. The mean weights from the survey are applied to the NCDs' ratings on the criteria to generate a 'total score' for each NCD, by which the NCDs are ranked (prioritised). Nineteen NCDs and five criteria were included. The criteria, in decreasing order of importance (mean weights in parentheses), are: deaths across the population (27.7%); loss of quality-of-life across the population (23.0%); cost to patients, families and the community (18.6%); cost to the health system (17.2%); and whether vulnerable groups are disproportionately affected (13.4%). The priority list of NCDs, stratified into four tiers in decreasing order of importance, is: 'Very critical' priority: coronary heart disease, back and neck pain, diabetes mellitus; 'Critical' priority: dementia and Alzheimer's disease, stroke; 'High' priority: colon and rectum cancer, depressive disorders, chronic obstructive pulmonary disease, chronic kidney disease, breast cancer, prostate cancer, arthritis, lung cancer; and 'Medium' priority: asthma, hearing loss, melanoma skin cancer, addictive disorders, non-melanoma skin cancer, headaches. The results from applying the MCDA-based framework for prioritising NCDs indicate that it is feasible and effective. The framework could also be used to support health research funding decision-making for other conditions.

Keywords: health research funding, non-communicable diseases (NCDs), priority-setting framework, multi-criteria decision analysis (MCDA), PAPRIKA method

Acknowledgements: The article arose from the first author's PhD thesis supervised by the other authors and Ronald Peeters. Thank you to the experts consulted during the course of the study, including Tony Blakely for his helpful comments on health care expenditure in New Zealand. Thank you to the survey participants. The second author (PH) co-invented and co-owns the 1000minds software mentioned in the article.

1. Introduction

Research into health problems affecting society, including health inequities borne by vulnerable population sub-groups, is very important (Khan et al., 2019; Smith et al., 2009). Although billions of dollars are invested in health research annually (Khan et al., 2019), only a small proportion of this spending targets health problems imposing the greatest burden on society (Allen, 2017; Viergever, 2013), and often with minimal attention to achieving a more equitable distribution of health outcomes. Given limited resources available for health research, prioritising areas for health research funding is necessary (Allen, 2017; Allen et al., 2017; Viergever, 2013). Health research priority-setting aims to direct funding to research areas of greatest need that will result in the biggest health gain, including reducing health inequities (Allen, 2017; Allen et al., 2017). Despite the importance of such priority-setting being generally well accepted, the development of priority lists of the most important research 'investment opportunities' remains challenging in practice (Rottingen et al., 2013; Smith et al., 2009). Notwithstanding a large number of studies into prioritising health interventions per se, very few studies have focused on prioritising health conditions, including non-communicable diseases (NCDs) (Adeyi et al., 2008; Allen, 2016; Marsh et al., 2017), to support health research funding – the subject of this paper.

NCDs are illnesses that are non-transmissible in the sense that they are not spread from person to person and are often characterised by slow deterioration and long duration (Khan et al., 2019; Strong et al., 2006; Vos et al., 2017). The four main groups of NCDs, according to the World Health Organization (WHO), are cardiovascular diseases, cancers, chronic respiratory diseases and diabetes (WHO, 2019). NCDs also include mental, neurological and musculoskeletal disorders – major causes of health burden in many countries (Carroll, 2019; Dieppe, 2013). Population aging and unhealthy lifestyles have resulted in an exponential growth of NCDs (Bigna & Noubiap, 2019; Tripathy, 2018). Globally, NCDs are the leading causes of mortality, morbidity and high health care expenditures (IHME, 2017; Muka et al., 2015; Vos et al., 2017), contributing to 73% of years of life lost (YLL) due to premature deaths and 80% of years lived with disability (YLD) (IHME, 2017; Prynn & Kuper, 2019; Roth et al., 2018).

Given the growing burden of NCDs, it is important that valid and reliable methods are used to prioritise NCDs for research funding. The WHO Global Forum for Health Research (GFHR) emphasises the importance of using structured priority-setting frameworks to direct investment into research areas, with particular attention to improving health equity and supporting vulnerable population sub-groups (Viergever et al., 2010). Multi-criteria decision analysis (MCDA) is a well-recognised tool for systematically setting priorities in the health sector (Marsh et al., 2017; Tacconelli et al., 2018). MCDA has been widely endorsed, with two reports written by the MCDA Emerging Good Practices Task Force of the International Society for Pharmacoeconomics and Outcomes Research (ISPOR) published in *Value in Health* (Hansen & Devlin, 2019; Marsh et al., 2017; Marsh et al., 2016; Thokala et al., 2016).

In this paper, an MCDA-based framework for prioritising NCDs with respect to their overall burden to society – developed and piloted in New Zealand (NZ) – is presented. The resulting

priority list of NCDs is intended to be used to *support* research funding decision-making. "*Support*" is emphasised because additional considerations such as the cost of the research and its likelihood of success are also important factors (and not included in this framework) when research projects are being assessed and, ultimately, funds are allocated in pursuit of 'value for money'. The framework is similar (though, on a logistically smaller scale) to the one used recently by the WHO to create a priority list of antibiotic-resistant bacteria to support research and development into new antibiotics (Tacconelli et al., 2018). The present study is the first to prioritise NCDs for health research funding.

2. Methods

Consistent with the recommendations of ISPOR's MCDA Task Force (Marsh et al., 2016; Thokala et al., 2016) and the process discussed in the article by Hansen and Devlin (Hansen & Devlin, 2019), the MCDA-based framework for prioritising NCDs with respect to their overall burden to society (and hence their importance for health research funding) involves three key components: (1) identifying NCDs to be prioritised for health research funding; (2) specifying prioritisation criteria and levels of performance within each criterion; and (3) determining weights for the criteria (and their levels), representing their relative importance to stakeholders.

Applying the information from these three components, each NCD can be rated according to its 'performance' on each of the criteria. Each NCD's ratings are aggregated using a linear (i.e. additive) equation – also referred to as a 'weighted-sum model' or 'points system' – to produce a 'total score' for each NCD (Hansen & Ombler, 2008; Tacconelli et al., 2018). Finally, based on their total scores (in the range 0-100%), the NCDs are ranked (prioritised).

Each of the three key components mentioned above is now explained in turn, followed by a brief discussion of the check of test-retest reliability of the survey used for determining the criteria weights. The NCDs' ratings on the criteria and their ranking are then explained. Finally, a brief explanation of further analyses to explore possible associations between participants' preferences – i.e. their criteria weights – and their socio-demographic and background characteristics is provided.

Identifying NCDs to be prioritised

The English-language literature was searched to identify NCDs responsible for large health burdens and spending in NZ and those that disproportionately affect vulnerable groups such as children, poor people, Māori (NZ's indigenous people) and other ethnic minorities (Blakely et al., 2019; MoH, 2009, 2016; OECD, 2017; Roth et al., 2018). This literature – academic and grey – included published articles, papers, reports, health system reviews and the websites of major international and national organisations such as the WHO, World Bank, Institute for Health Metrics and Evaluation, Organisation for Economic Co-operation and Development, NZ Ministry of Health and Health Research Council. Experts in NCDs from a wide range of clinical, research and policy-making backgrounds in NZ were also consulted to corroborate interpretations and improve understanding of the information gleaned from the literature search.

Specifying prioritisation criteria

The literature was searched again, but more systematically this time, to discover criteria used for priority-setting in health systems around the world, potentially in a variety of applications (e.g. health technology prioritisation, etc). Articles, reports and grey literature for the period 1990-2019 were searched using PubMed's and Google Scholar's search engines and combinations of these keywords: priority-setting, prioritisation criteria, social and ethical issues, decision-making, health research funding and/or disease, non-communicable disease(s) and NCD(s).

Because the weights on the criteria and the levels within each criterion are to be determined using a survey (explained in the next sub-section), the criteria need to be specified concisely using language that most survey participants would be expected to understand. Likewise, as well as being concise and simple, the levels should be generic in nature – e.g. ranging from 'low' to 'high' – and sufficiently granular to be able to distinguish between the NCDs included in the study. The final specification of the criteria and levels was validated and refined following in-depth interviews with the experts referred to in the previous sub-section, and pilot-testing of the framework with several participants from diverse backgrounds.

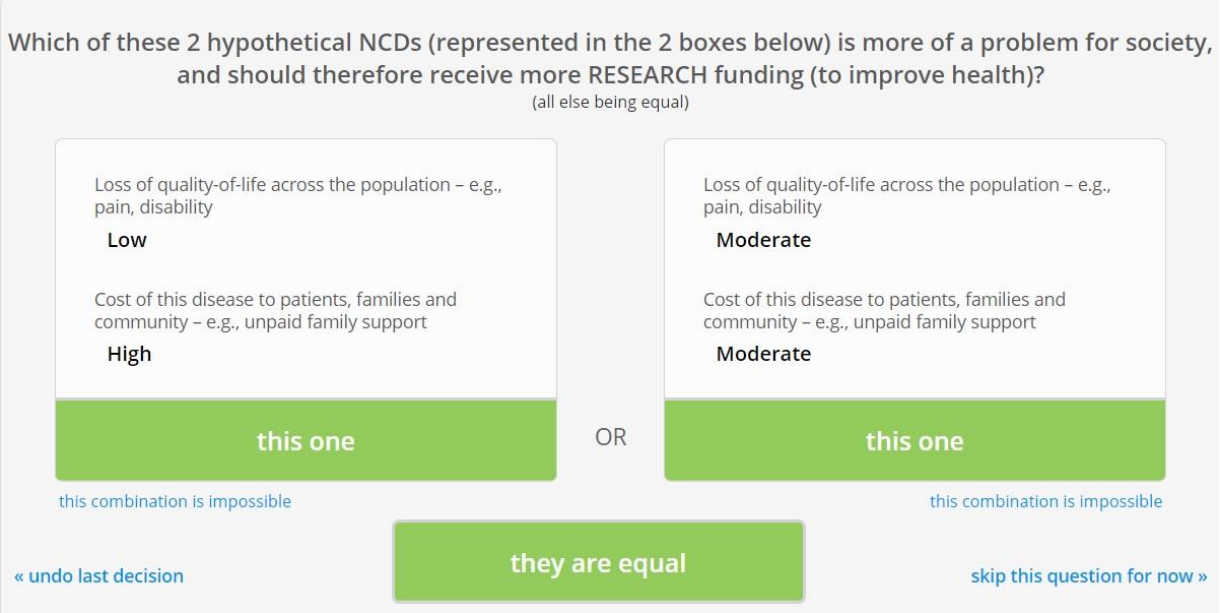
Determining criteria weights

The weights were determined by surveying people from three health sector groups: (1) patients or members of the general public; (2) health providers (e.g. nurses or doctors); and (3) health policy-makers or researchers. Consistent with the literature, patients and the general public were included because they are the ultimate beneficiaries of health research (Kapiriri, 2018). The other two groups were included because of their expert knowledge and interest in NCDs. Convenience and purposive sampling with 'snowballing' (Etikan et al., 2016; Goodman, 1961; O'Haire et al., 2011; Street et al., 2014) – whereby participants who were initially contacted were asked to forward the survey link to other eligible and potentially interested people – was used to identify the participants, who were initially given two weeks to complete the survey, with a reminder email sent out after 10 days.

The survey applied the PAPRIKA method for determining weights – an acronym for Potentially All Pairwise RanKings of all possible Alternatives (Hansen & Ombler, 2008) – as implemented by 1000minds software (www.1000minds.com). As mentioned earlier, this method and software was used by the WHO to create its priority list of antibiotic-resistant bacteria for research funding (Tacconelli et al., 2018). PAPRIKA and 1000minds have also been widely used for patient (Hansen et al., 2012) and health technology prioritisation (Golan & Hansen, 2012; Sullivan & Hansen, 2017), disease classification (Aringer et al., 2019) and health preferences research (Sullivan et al., 2020).

The PAPRIKA method, in the present context, involves participants being asked to pairwise rank two hypothetical NCDs defined on two criteria at a time and involving a trade-off – in terms of which is more of a problem for society and therefore more eligible for research funding. An example of a pairwise-ranking question from 1000minds software appears in Figure 1.

Figure 1: **Example of a pairwise-ranking question (screenshot from 1000minds software)**



Such pairwise-ranking questions are repeated with different pairs of hypothetical NCDs – always defined on two criteria at a time and involving a trade-off. Each time a participant answers a question, the PAPRIKA method applies the logical property of transitivity to identify and eliminate all other pairs of hypothetical NCDs defined on two criteria at a time that are pairwise ranked, thereby minimising the number of questions asked. For example, if a participant ranks NCD *A* ahead of NCD *B* and *B* ahead of NCD *C*, then *A* is ranked ahead of *C* (and so would not be asked about). Each time a person answers a question, PAPRIKA adapts with respect to choosing the next question (always one whose answer is not implied by earlier answers) based on all preceding answers (Hansen & Ombler, 2008). This adaptivity and the transitivity-based elimination procedure ensures the number of questions a participant is asked is minimised while ensuring they end up having pairwise ranked all hypothetical NCDs defined on two criteria at a time, either explicitly or implicitly (by transitivity).

The number of questions to be answered by participants was also reduced, thereby reducing the elicitation burden, by utilising the software's interpolation feature. For example, if a criterion has five levels – e.g. 'low', 'low to moderate', 'moderate', 'moderate to high' and 'high' – then only the first ('low'), third ('moderate') and fifth ('high') levels are included in the pairwise-ranking questions. The weights for the second ('low to moderate') and fourth ('moderate to high') levels are interpolated using Bézier interpolation (Farin et al., 2002) – i.e. in essence, by fitting a smoothed curve through the weights for the first, third and fifth levels. Thus, the granularity arising from having the full set of levels available for rating the NCDs on the criteria is maintained while the number of questions that survey participants are asked to answer is limited.

Two checks related to the quality of each participant's data were performed by the software. First, three pairwise-ranking questions were repeated at the end of each participant's survey to check the consistency of their answers. Second, any participants who answered all their

questions by clicking "they are equal" (see Figure 1 again) – i.e. *universal* indifference – were identified, to gauge participants' engagement with the survey.

From each participant's answers to the pairwise-ranking questions, PAPRIKA uses linear programming methods to determine weights for the criteria (and for the levels within each criterion), representing their relative importance to the participant. Participants were also asked questions about their socio-demographic and background characteristics, and how easy or difficult they found answering the pairwise-ranking questions.

Checking test-retest reliability

As a check of the survey's test-retest reliability, a sub-sample of 40 participants completed the survey twice, almost two weeks apart. The mean criteria weights obtained from the two implementations of the survey were tested for statistically significant differences.

Rating NCDs on the criteria

Each NCD was rated on the criteria using information from the literature, including YLL, YLD and economic burden. This rating exercise was performed by the first author (SB) in consultation with the other authors and the experts involved at the other stages of the study, as mentioned earlier. The latest YLL and YLD data for NZ were obtained from the website of the Institute for Health Metrics and Evaluation (IHME, 2019). NCDs' economic burdens were derived from reports and cost of illness (CoI) studies for NZ. International and national reports and papers related to the NZ context were reviewed to identify NCDs disproportionately affecting vulnerable groups (Blakely et al., 2019; MoH, 2009, 2016).

As there is very little information relating to indirect health care costs available, only direct health care expenditure for each NCD was considered in the analysis. Cost estimates from Blakely et al (2019), the best data source available at the time, were used to estimate the cost burdens on the NZ Government – i.e. publicly-funded health care – and patients (and families). According to that study, approximately 82% of direct health care costs are publicly-funded, with the remaining 18% paid by individuals – e.g. co-payments, out-of-pocket payments and private health insurance (Blakely et al., 2019). (Although this is the best data source available, the estimated costs should be treated with caution.) Costs were adjusted for inflation using the NZ general consumer price index (CPI) for 2017 via the Reserve Bank of New Zealand inflation calculator (The Reserve Bank of New Zealand, 2017).

Ranking NCDs

Applying the mean criteria weights from the survey to the ratings of the NCDs generates a 'total score' for each NCD in the range 0-100%. Based on their total scores, the NCDs are ranked (prioritised).

Predicting participants' preferences

Two-step cluster analysis was performed to identify clusters of participants with similar preferences (i.e. criteria weights). A chi-squared test (along with Cramér's V) was used to explore possible associations (and their effect size) between participants' preferences and

their socio-demographic and background characteristics. As participants were sampled mainly from the higher educated population, a t-test was used to determine whether there are statistically significant differences between the preferences of participants with higher and lower levels of education.

3. Results

NCDs to be prioritised

Twenty-one NCDs were initially identified. However, two of them – anxiety and dental disorders – were excluded due to a paucity of information. These remaining 19 NCDs, in alphabetical order, are: addictive (drug and alcohol use) disorders, arthritis, asthma, back and neck pain, breast cancer, chronic kidney disease, chronic obstructive pulmonary disease, colon and rectum cancer, coronary heart disease, dementia and Alzheimer's disease, depressive disorders, diabetes mellitus (mainly type 2), headaches, hearing loss, lung cancer, melanoma skin cancer, non-melanoma skin cancer, prostate cancer and stroke.

Prioritisation criteria

The literature search revealed three main groups of criteria used for prioritising NCDs for research funding: (1) burden of disease (BoD) – i.e. morbidity and mortality, including health-related quality of life; (2) cost burden and efficiency; and (3) additional considerations such as ethical and social issues (Drummond et al., 2015; Golan et al., 2011; Shmueli et al., 2017; Thokala et al., 2016; Tromp & Baltussen, 2012).

The first of these three groups of prioritisation criteria, BoD – often described in terms of quality- or disability-adjusted life years (QALYs or DALYs) or the latter's two components, YLL and YLD (IHME, 2017; Prynn & Kuper, 2019; Roth et al., 2018; Tromp & Baltussen, 2012; Vos et al., 2017) – is the most widely-used criterion for evaluating health losses and prioritising health conditions, including NCDs. The second group recognises that NCDs impose substantial health care costs (Allen, 2017; Muka et al., 2015; Smith et al., 2009) in terms of publicly-funded health care costs, and expenses incurred by patients and families' including out-of-pocket expenses, as well as the value of unpaid and informal family support (Drummond et al., 2015). The third main group is associated with reducing health inequities affecting vulnerable population sub-groups such as children, poor people and ethnic minorities, including indigenous people, as these groups experience a disproportionately larger share of the disease burden (Best, 2012). As reported in Table 1, five criteria were specified for prioritising NCDs, with 2-7 levels within each criterion. Table 1 also summarises the health system goals addressed by each criterion and the data source used to assess the NCDs on each criterion (Tromp & Baltussen, 2012).

Table 1: Criteria for prioritising NCDs

Criterion	Level	Health system goal addressed	Data source
Deaths across the population – i.e. reduced life expectancy	None (or low) Low to moderate Moderate Moderate to high High High to very high Very high	Mortality	Latest NZ YLL data (2017) extracted from the IHME website.
Loss of quality-of-life across the population – e.g. pain, disability	Low Low to moderate Moderate Moderate to high High	Morbidity	Latest NZ YLD data (2017) extracted from the IHME website.
Cost of the disease to the health system – i.e. publicly-funded health care	Low Low to moderate Moderate Moderate to high High	Health system costs	CoI studies, reports and papers – e.g. from MoH, BODE[3] and PHARMAC.
Cost of the disease to patients, families and community – e.g. unpaid family support	Low Low to moderate Moderate Moderate to high High	Societal costs	
Disproportionately affects vulnerable groups – e.g. Māori, children, poor people	Yes No	Equity	Reports and papers –e.g. from WHO, IHME, OECD, MoH, HRC, DHBs.

BODE[3]: Burden of Disease Epidemiology, Equity and Cost-Effectiveness Programme. BODE[3] is a funded program by the Health Research Council (HRC) of New Zealand (NZ) to provide health economics data. CoI: Cost of Illness. DHBs: 20 District Health Boards in NZ are responsible for providing health care services to the population within their districts. IHME: Institute for Health Metrics and Evaluation. MoH: Ministry of Health. OECD: Organisation for Economic Co-operation and Development. PHARMAC: the Pharmaceutical Management Agency is an NZ crown entity that decides which pharmaceutical products and devices are subsidised for use in the public sector. YLD: Years Lived with Disability. YLL: Years of Life Lost due to premature deaths. WHO: World Health Organization.

Criteria weights

The survey for determining the criteria weights required participants to answer 20 pairwise-ranking questions on average, taking 15-20 minutes in total. The survey was completed by 517 participants; however, 27 (5%) answered all three repeated questions contradictorily (inconsistently), and another 14 (3%) answered 'they are equal' for all questions (universal indifference). These participants were excluded from the data set because both behaviours are suggestive of the participants not having engaged seriously with the pairwise-ranking exercise

or not having understood the questions. The remaining 476 participants answered at least one of the three repeated questions identically (consistently), 356 (75%) answered two questions consistently and 168 (35%) answered all three consistently.

The socio-demographic and background characteristics of these 476 participants are summarised in Table 2, where, inter alia, it can be seen that almost 31% were patients or members of the general public, 35% were health providers (e.g. nurses or doctors) and 34% were health policy-makers or researchers. Almost 53% of participants said they found completing the survey and answering the pairwise-ranking questions relatively easy.

The mean weights of the criteria and their levels, representing their relative importance to participants, are reported in Table 3 with their standard deviations (SD). The most important criterion for prioritising NCDs with respect to their overall burden to society (and hence their importance for health research funding, all else being equal) is 'deaths across the population' (mean weight = 27.7%), followed by 'loss of quality-of-life across the population' (23.0%), 'cost to patients, families and community' (18.6%), 'cost to the health system' (17.2%) and – the least-important criterion – 'disproportionately affects vulnerable groups' (13.4%). Criteria weights indicate the relative strength of participants' preferences with respect to reducing the multi-dimensional burden of NCDs on society.

Test-retest reliability

For the 40 people who completed the MCDA survey twice, the results of a paired sample t-test revealed no statistically significant differences between the mean criteria weights from the first and second surveys.

Ratings of NCDs on the criteria

The ratings of the 19 NCDs on the five criteria are reported in Table 4.

Table 2: Summary of characteristics of survey participants (n=476)

Characteristic	n (%)
Gender	
Male	201 (42.2)
Female	274 (57.6)
Gender diverse	1 (0.2)
Age (years)	
18-24	15 (3.2)
25-34	62 (13.0)
35-44	108 (22.7)
45-54	129 (27.1)
55-64	114 (23.9)
65 and over	48 (10.1)
Ethnicity	
NZ European	302 (63.4)
Māori	45 (9.5)
Chinese	42 (8.8)
Pacific	18 (3.8)
Indian	29 (6.1)
Others	40 (8.4)
Qualification	
No qualification	30 (6.3)
Secondary school	47 (9.9)
Post-secondary school qualification	72 (15.1)
University degree equivalent	327 (68.7)
Region	
North Island	325 (68.3)
South Island	151 (31.7)
Work status	
Working	312 (65.5)
Not working	73 (15.3)
Retired	91 (19.1)
Participant (or immediate family member) with NCD(s)	
Yes	373 (78.4)
No	103 (21.6)
Use of health care services	
Never	4 (0.8)
Occasionally	373 (78.4)
Frequently	99 (20.8)
Background	
Patient or member of the general public	149 (31.3)
Health provider – e.g. nurse or doctor	166 (34.9)
Health policy-maker or researcher	161 (33.8)
Ease of completing the survey and answering pairwise-ranking questions	
Relatively easy	250 (52.5)
Relatively difficult	226 (47.5)

Percentages for ethnicity do not sum to 100 as some people identify with multiple groups.

Table 3: Mean criteria weights (in decreasing order of importance)

Criterion and levels	Mean weight (SD)
Deaths across the population – i.e. reduced life expectancy	
None (or low)	0.0 (0.0)
Low to Moderate	6.9
Moderate	12.7 (5.0)
Moderate to high	17.0
High	20.4 (6.5)
High to Very high	24.1
Very high	**27.7** (8.1)
Loss of QoL across the population – e.g. pain, disability	
Low	0.0 (0.0)
Low to Moderate	1.0
Moderate	11.9 (4.8)
Moderate to high	17.5
High	**23.0** (6.3)
Cost of the disease to the health system – i.e. publicly-funded health care	
Low	0.0 (0.0)
Low to Moderate	4.3
Moderate	8.6 (4.7)
Moderate to high	12.9
High	**17.2** (6.7)
Cost of the disease to patients, families and community – e.g. unpaid family support	
Low	0.0 (0.0)
Low to Moderate	4.7
Moderate	9.3 (4.2)
Moderate to high	14.0
High	**18.6** (6.3)
Disproportionately affects vulnerable groups – e.g. Māori, children, poor people	
No	0.0 (0.0)
Yes	**13.4** (6.3)

SD: Standard Deviation is only available for the levels presented in the survey. Note that other levels are interpolated using Bézier interpolation, as explained in the context. Values reported are percentages. Bolded values sum to one (100%) and represent the relative weights of the criteria overall.

Table 4: Ratings of the NCDs on the criteria

NCD	Deaths across the population – i.e. reduced life expectancy	Loss of QoL across the population – e.g. pain, disability	Cost of the disease to the health system – i.e. publicly-funded health care	Cost of the disease to patients, families and community – e.g. unpaid family support	Disproportionately affects vulnerable groups – e.g. Māori, children, poor people
Addictive disorders	None (or low)	Low to moderate	Low	Low	Yes
Arthritis	None (or low)	Low to moderate	Moderate	Moderate	Yes
Asthma	None (or low)	Low to moderate	Low to moderate	Low to moderate	Yes
Back and neck pain	None (or low)	High	High	High	Yes
Breast cancer	None (or low) to Moderate	Low	Moderate	Moderate	Yes
CHD	Very high	Low	High	High	Yes
CKD	None (or low) to Moderate	Low	Moderate	Moderate	Yes
Colon and rectum cancer	Moderate	Low	Moderate	Moderate	Yes
COPD	Moderate	Low to moderate	Low to moderate	Low to moderate	Yes
Dementia and Alzheimer's disease	Moderate to High	Low to moderate	High	High	No
Depressive disorders	None (or low)	Moderate	Moderate	Moderate	Yes
Diabetes mellitus	None (or low) to Moderate	Moderate	High	High	Yes
Headaches	None (or low)	Moderate	Low	Low	No
Hearing loss	None (or low)	Moderate	Low	Low	Yes
Lung cancer	Moderate	Low	Low to moderate	Low to moderate	Yes
Melanoma skin cancer	None (or low) to Moderate	Low	Low	Low	Yes
Non-melanoma skin cancer	None (or low)	Low	Low	Low	Yes
Prostate cancer	None (or low) to Moderate	Low	Moderate	Moderate	Yes
Stroke	Moderate to High	Low to moderate	Moderate	Moderate	Yes

QoL: Quality-of-life; CHD: Coronary heart disease; CKD: Chronic kidney disease; COPD: Chronic obstructive pulmonary disease

Ranking of NCDs

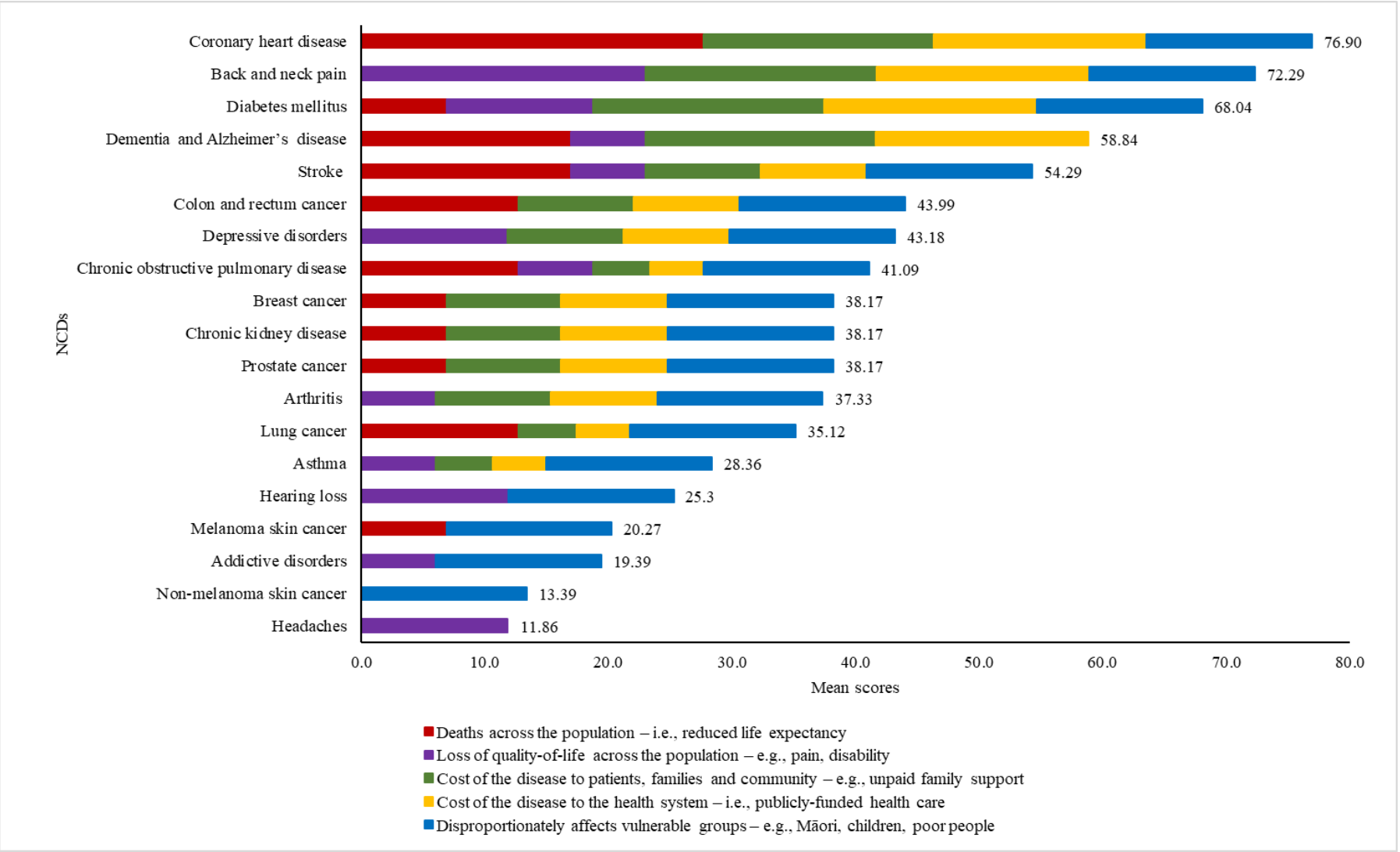
Applying the mean criteria weights (Table 3) to the NCDs' ratings (Table 4) resulted in a total score for each of the 19 NCDs – in the range 0-100% – as shown in Figure 2. With a total score of 77%, coronary heart disease (CHD) is the top-ranked NCD, followed by back and neck pain (72%) and diabetes mellitus (68%), and so on for the other 16 NCDs.

Consistent with the presentational style and terminology used by the WHO for its priority list of antibiotic-resistant bacteria (Tacconelli et al., 2018), the ranking of the 19 NCDs were stratified into four tiers of priority: 'very critical', 'critical', 'high' and 'medium'. The four tiers are (total score ranges in parentheses): 'Very critical' priority (68-77%): coronary heart disease, back and neck pain, diabetes mellitus; 'Critical' priority (54-59%): dementia and Alzheimer's disease, stroke; 'High' priority (35-44%): colon and rectum cancer, depressive disorders, chronic obstructive pulmonary disease, chronic kidney disease, breast cancer, prostate cancer, arthritis, lung cancer; and 'Medium' priority (12-29%): asthma, hearing loss, melanoma skin cancer, addictive disorders, non-melanoma skin cancer, headaches.

Predicting participants' preferences

The results of the cluster analysis (and chi-squared test and Cramér's V) indicate that the variation in participants' preferences is generally unrelated to their socio-demographic and background characteristics, suggesting that people's preferences are largely idiosyncratic. The results of the t-test indicate that there are no statistically significant differences between the preferences of participants with higher and lower levels of education.

Figure 2: NCDs total scores



Values reported are percentages.

4. Discussion

In this study, 19 NCDs were prioritised based on five criteria, where their weights were determined from a survey of NZ health sector stakeholders, and information about the NCDs' performance on the criteria. Like the WHO's priority list of antibiotic-resistant bacteria (Tacconelli et al., 2018), for ease of communication (e.g. with researchers and policy-makers), the priority list of NCDs was stratified into four tiers of priority. NCDs in the 'very critical' tier – coronary heart disease, back and neck pain and diabetes mellitus – have high rates of YLL or YLD and high health system costs. In contrast, NCDs in the (lowest) 'medium' tier – asthma, hearing loss, melanoma skin cancer, addictive disorders, non-melanoma skin cancer and headaches – have the lowest burden. The intended use of such a (tiered) priority list is to support research funding decision-making. Additional considerations such as the cost of the research and its likelihood of success would also need to be included when research projects are being assessed and, ultimately, funds are allocated in pursuit of 'value for money' (Tuffaha et al., 2019; Tuffaha et al., 2018).

The inclusion of back and neck pain in the 'very critical' tier is consistent with the findings from global burden of disease (GBD) studies (Roth et al., 2018; Vos et al., 2017). For example, Blakely et al (2019) point out that "in GBD 2016, New Zealand had 1.31 times higher morbidity burden for back pain than expected based on its level of sociodemographic development" (p. 17). As well as having high rates of YLD, back and neck pain is associated with high health care costs – reflecting the correlation between YLD and health care costs, a common finding in other studies (Blakely et al., 2019; Fun et al., 2019; Kinge et al., 2017; Wieser et al., 2018).

In NZ, the Ministry of Health, the Ministry of Business, Innovation and Employment and the Health Research Council (HRC) are closely involved with setting health research priorities at the national level (HRC, 2019). The HRC, in conjunction with the Healthier Lives National Science Challenge (a national research collaboration), recently embarked on setting health research priorities for 2017-27 for cancer, cardiovascular diseases, diabetes and obesity. The priority list created in the present study suggests that, in addition to these NCDs, other NCDs – e.g. back and neck pain and dementia and Alzheimer's (i.e. in the 'very critical' and 'critical' priority tiers) – should probably also be considered as priorities for research funding in NZ.

Study limitations

This study has several limitations, mainly related to data deficiencies. First, anxiety disorders and dental disorders – the former associated with relatively major health burden (IHME, 2019; Lee et al., 2017) and the latter with high health care expenditure (mostly not publicly-funded) (MoH, 2017) – were excluded from the priority-setting framework due to a paucity of available data. Second, the 19 NCDs included in the framework are largely heterogeneous – e.g. stroke can range from being mild and transient to a major disabling event requiring nursing home care for the rest of a person's life – however, recognising such heterogeneity and representing the NCDs with more granularity was impossible because the required

information was unavailable. Third, due to a lack of detailed data on NCDs' health care costs, the proportion proposed by Blakely et al. (2019) – indicating that 82% of NZ health care expenditure is publicly-funded and the remaining 18% is privately-funded – was used in this study to estimate the cost burden by the government and patients (and their families) (Blakely et al., 2019). Fourth, there was very little evidence on NCDs-related health care expenditures by the private sector and non-governmental organisations (e.g. Cancer Society); however, given 82% of NZ's total health care spending is publicly-funded, this may not be a serious limitation (Blakely et al., 2014, 2015; Blakely et al., 2019). Overall, there is a need to improve the quality (and availability) of data on NCDs. Further efforts are needed to conduct CoI studies from the perspective of both health care providers and patients to generate data for economic evaluation studies and to assist policy-makers in allocating resources.

Another limitation of the study is that the sample of participants used for the survey to determine the criteria weights is not representative of the NZ population. However, the variation in participants' preferences (weights) was found to be related more to their personal preferences than to their background characteristics; also, no significant differences between the preferences of participants with high levels and low levels of education respectively were found. Because participants were recruited using convenience and purposive sampling with 'snowballing', calculating a response rate is impossible; nonetheless, the number of participants (n=517, with 476 usable responses) was larger than for other studies that conducted online surveys to set research priorities across health interventions (O'Haire et al., 2011; Street et al., 2014). The survey included in the present framework could be repeated using a more representative sample.

5. Conclusion

To the best of the authors' knowledge, the MCDA-based framework developed and piloted in this study is the first attempt to create a priority list of NCDs to support health research funding decision-making. The framework enables multiple criteria to be incorporated for evaluating a wide range of NCDs, and for multiple stakeholders to be involved. The priority list of NCDs created confirms that it is important to recognise their multi-dimensional nature – e.g. mortality, morbidity and health care costs – when evaluating their relative priority. The successful application of the framework in this study confirms that it is feasible and effective. The framework could also be used to support priority-setting for health research funding for other health conditions.

References

Adeyi, O., Smith, O., & Robles, S. (2008). Public policy and the challenge of chronic noncommunicable diseases. *International Journal of Epidemiology, 37*(3), 686-687.

Allen, L. (2016). Non-communicable disease research. *International Journal of Non-Communicable Diseases, 1*(3), 131-133.

Allen, L. (2017). Non-communicable disease funding. *The Lancet, 5*, 92.

Allen, L., Cobiac, L., & Townsend, N. (2017). Quantifying the global distribution of premature mortality from non-communicable diseases. *Journal of Public Health, 1*, 1-6.

Aringer, M., Costenbader, C., Daikh, D., & et al. (2019). 2019 European League against Rheumatism/American College of Rheumatology classification criteria for systemic lupus erythematosus. *Arthritis & Rheumatology, 71*, 1400-1412.

Best, R. (2012). Disease politics and medical research funding: Three ways advocacy shapes policy. *American Sociological Review, 77*(5), 780-803.

Bigna, J., & Noubiap, J. (2019). The rising burden of non-communicable diseases in Sub-Saharan Africa. *The Lancet Global Health, 7*(10), 1295-1296.

Blakely, T., Atkinson, J., Kvizhinadze, G., & et al. (2014). Health system costs by sex, age and proximity to death, and implications for estimation of future expenditure. *The New Zealand Medical Journal, 127*(1393), 12-25.

Blakely, T., Atkinson, J., Kvizhinadze, G., & et al. (2015). Updated New Zealand health system cost estimates from health events by sex, age and proximity to death: Further improvements in the age of 'big data'. *The New Zealand Medical Journal, 128*(1422), 13-23.

Blakely, T., Kvizhinadze, G., Atkinson, J., Dieleman, J., & Clarke, P. (2019). Health system costs for individual and comorbid noncommunicable diseases: An analysis of publicly-funded health events from New Zealand. *PLOS Medicine, 16*(1), e1002716.

Carroll, W. (2019). The global burden of neurological disorders. *The Lancet, 18*(5), 418-419.

Dieppe, P. (2013). Chronic musculoskeletal pain. *British Medical Journal, 16*(346), f3146.

Drummond, M., Sculpher, M., Claxton, G., & Torrance, G. (2015). Methods for the Economic Evaluation of Health Care Programmes (4th ed): New York: Oxford University Press.

Etikan, I., Musa, S., & Alkassim, R. (2016). Comparison of convenience sampling and purposive sampling. *American Journal of Theoretical and Applied Statistic, 1-4*.

Farin, G., Hoschek, J., & Kim, M. (2002). Handbook of Computer Aided Geometric Design: Elsevier Science Ltd, North Holland.

Fun, W., Sararaks, S., Tan, E., & et al. (2019). Research funding impact and priority setting – advancing universal access and quality healthcare research in Malaysia. *BMC Health Services Research, 19*(248), 1-8.

Golan, O., & Hansen, P. (2012). Which health technologies should be funded? A prioritization framework based explicitly on value for money. *Israel Journal of Health Policy Research, 1*, 44.

Golan, O., Hansen, P., Kaplan, G., & et al. (2011). Health technology prioritization: Which criteria for prioritizing new technologies and what are their relative weights. *Health Policy, 102*(2-3), 126-135.

Goodman, L. (1961). Snowball sampling. *Annals of Mathematical Statistics, 32*(1), 148-170.

Hansen, P., & Devlin, N. (2019). Multi-criteria decision analysis (MCDA) in healthcare decision-making. *The Oxford Encyclopedia of Health Economics. Oxford University Press*.

Hansen, P., Hendry, A., Naden, R., Ombler, F., & Stewart, R. (2012). A new process for creating points systems for prioritizing patients for elective health services. *Clinical Governance: An International Journal, 17*, 200-209.

Hansen, P., & Ombler, F. (2008). A new method for scoring additive multi-attribute value models using pairwise rankings of alternatives. *Journal of Multi-Criteria Decision Analysis, 15*(3-4), 87-107.

HRC. (2019). The New Zealand health research prioritisation framework. www.hrc.govt.nz/sites/default/files/2020-01/NZ%20Prioritisation-Framework-FA-web_0.pdf.

IHME. (2017). Findings from the Global Burden of Disease Study 2017. www.healthierlives.co.nz/about/about-healthier-lives.

IHME. (2019). New Zeland burden of disease. https://vizhub.healthdata.org/gbd-compare/.

Kapiriri, L. (2018). Stakeholder involvement in health research priority setting in low income countries: The case of Zambia. *Research Involvement and Engagement, 4*, 41.

Khan, M., Rahman-Shepherd, A., Painter, H., & Fletcher, H. (2019). How can we improve priority-setting for investments in health research? A case study of tuberculosis? *Health Research Policy and Systems, 17*(1), 68.

Kinge, J., Saelensminde, K., Dieleman, J., Vollset, S., & Norheim, O. (2017). Economic losses and burden of disease by medical conditions in Norway. *Health Policy, 121*(6), 691-698.

Lee, C., Duck, I., & Sibley, C. (2017). Ethnic inequality in diagnosis with depression and anxiety disorders. *The New Zealand Medical Journal, 130*(1454), 10-20.

Marsh, K., Goetghebeur, M., Thokala, P., & Baltussen, R. (2017). Multi-Criteria Decision Analysis to Support Healthcare Decisions. Springer, New York.

Marsh, K., M, I., Thokala, P., R, B., Boysen, M., Kaló, Z., & et al. (2016). Multiple criteria decision analysis for health care decision making - Emerging Good Practices: Report 2 of the ISPOR MCDA Emerging Good Practices Task Force. *Value in Health, 19*(1), 125-137.

MoH. (2009). Report on New Zealand cost-of-illness studies on long-term conditions. www.health.govt.nz/system/files/documents/publications/nz-cost-of-illness-jul09.pdf.

MoH. (2016). Health Loss in New Zealand 1990–2013: A report from the New Zealand burden of diseases, injuries and risk factors study. www.health.govt.nz/system/files/documents/publications/health-loss-in-new-zealand-1990-2013-aug16.pdf.

MoH. (2017). Publicly funded dental care. www.health.govt.nz/your-health/services-and-support/health-care-services/visiting-dentist/publicly-funded-dental-care.

Muka, T., Imo, D., Jaspers, L., & et al. (2015). The global impact of non-communicable diseases on healthcare spending and national income: a systematic review. *European Journal of Epidemiology, 30*(4), 251-277.

O'Haire, C., McPheeters, M., Nakamoto, E., & et al. (2011). Engaging stakeholders to identify and prioritize future research needs. Methods for engaging stakeholders to identify and prioritize future research needs. Methods Future Research Needs Report No. 4. (Prepared by the Oregon Evidence-based Practice Center and the Vanderbilt Evidence-based Practice Center under Contract No. 290-2007-10057-I.) AHRQ Publication No. 11-EHC044-EF. Rockville, MD: Agency for Healthcare Research and Quality, 2011.

OECD. (2017). Health policy in New Zealand. www.oecd.org/els/health-systems/Health-Policy-in-New-Zealand-March-2017.pdf.

Prynn, J., & Kuper, H. (2019). Perspectives on disability and non-communicable diseases in low- and middle-income countries, with a focus on stroke and dementia. *International Journal of Environmental Research and Public Health, 16*(18), 3488.

Roth, G., Abate, D., Abate, K., & et al. . (2018). Global, regional, and national age-sex-specific mortality for 282 causes of death in 195 countries and territories, 1980-2017: A systematic analysis for the global burden of disease study 2017. *The Lancet, 392*, 1736-1788.

Rottingen, J., Regmi, S., Eide, M., & et al. (2013). Mapping of available health research and development data: What's there, what's missing, and what role is there for a global observatory? *The Lancet, 382*, 1286-1307.

Shmueli, A., Golan, O., Paolucci, F., & Mentzakis, E. (2017). Efficiency and equity considerations in the preferences of health policy-makers in Israel. *Israel Journal of Health Policy Research, 6*(18), 1-11.

Smith, N., Mitton, C., Peacock, S., Cornelissen, E., & MacLeod, S. (2009). Identifying research priorities for health care priority-setting: A collaborative effort between managers and researchers. *BMC Health Services Research, 9*, 165.

Street, J., Duszynski, K., Krawczyk, S., & et al. (2014). The use of citizens' juries in health policy decision-making: A systematic review. *Social Science & Medicine, 109*, 1-9.

Strong, K., Mathers, C., Epping-Jordan, J., & Beaglehole, R. (2006). Preventing chronic disease: A priority for global health. *International Journal of Epidemiology, 35*(2), 492-494.

Sullivan, T., & Hansen, P. (2017). Determining criteria and weights for prioritizing health technologies based on the preferences of the general population: A New Zealand pilot study. *Value in Health, 20*(4), 679-686.

Sullivan, T., Hansen, P., Ombler, F., Derrett, S., & Devlin, N. (2020). A new tool for creating personal and social EQ-5D-5L value sets, including valuing 'dead'. *Social Science & Medicine, 246*, 112707.

Tacconelli, E., Carrara, E., Savoldi, A., & et al. . (2018). Discovery, research, and development of new antibiotics: The WHO priority list of antibiotic-resistant bacteria and tuberculosis. *The Lancet Infectious Diseases, 18*(3), 318-327.

The Reserve Bank of New Zealand. (2017). The Reserve Bank of New Zealand inflation calculator. www.rbnz.govt.nz/monetary-policy/inflation-calculator.

Thokala, P., Devlin, N., Marsh, K., & et al. (2016). Multiple criteria decision analysis for health care decision making – An introduction: Report 1 of the ISPOR MCDA Emerging Good Practices Task Force. *Value in Health, 19*(1), 1-13.

Tripathy, J. (2018). Research priorities in non-communicable diseases in developing countries: Time to go beyond prevalence studies. *Public Health Action, 8*(2), 98-99.

Tromp, N., & Baltussen, R. (2012). Mapping of multiple criteria for priority setting of health interventions: An aid for decision makers. *BMC Health Services Research, 12*, 454.

Tuffaha, H., Aitken, J., Chambers, S., & Scuffham, P. (2019). A framework to prioritize health research proposals for funding: Integrating value for money. *Applied Health Economics and Health Policy, 17*(6), 761-770.

Tuffaha, H., El Saifi, N., Chambers, S., & Scuffham, P. (2018). Directing research funding to the right research projects: A review of criteria used by research organizations in Australia in prioritising health research projects for funding. *BMJ Open, 8*(12), e026207.

Viergever, R. (2013). The mismatch between the health research and development (R&D) that is needed and the R&D that is undertaken: An overview of the problem, the causes, and solutions. *Global Health Action, 6*, 22450.

Viergever, R., Terry, R., & MP, M. (2010). Health research prioritization at WHO: An overview of methodology and high level analysis of WHO led health research priority setting exercises. Geneva: World Health Organization, www.who.int/rpc/publications/en/.

Vos, T., Abajobir, A., Abate, K., & et al. (2017). Global, regional, and national incidence, prevalence, and years lived with disability for 328 diseases and injuries for 195 countries, 1990–2016: A systematic analysis for the global burden of disease study 2016. *The Lancet*, *390*(10100), 1211-1259.

WHO. (2019). Noncommunicable diseases. www.who.int/gho/ncd/en/.

Wieser, S., Riguzzi, M., Pletscher, M., Huber, C., Telser, H., & Schwenkglenks, M. (2018). How much does the treatment of each major disease cost? A decomposition of Swiss National Health Accounts. *European Journal of Health Economics*, *19*(8), 1149-1161.