Recommendations to Plan a National Burden of Disease Study

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Abstract

**Background:** The *InfAct* (Information for Action) project is a European Commission Joint Action on Health Information which has promoted the potential role of burden of disease (BoD) approaches to improve the current European-Health Information System (EU-HIS). It has done so by raising awareness of the concept, the methods used to calculate estimates and their potential implications and uses in policymaking. The BoD approach is a systematic and scientific effort to quantify and compare the magnitude of health loss due to different diseases, injuries, and risk factors with estimates produced by demographic characteristics and geographies for specific points in time. Not all countries have the resources to undertake such work, and may therefore start with a more restricted objective, e.g., a limited number of diseases, or the use of simple measures of population health such as disease prevalence or life expectancy. The main objective to develop these recommendations was to facilitate those countries planning to start a national burden of disease study.

**Results:** These recommendations could be considered as minimum requirements for those countries planning to start a BoD study and includes following elements: 1. Define the objectives of a burden of disease study within the context of your country, 2. Identify, communicate and secure the benefits of performing national burden of disease studies, 3. Secure access to the minimum required data sources, 4. Ensure the minimum required capacity and capability is available to carry out burden of disease study, 5. Establish a clear governance structure for the burden of disease study and stakeholder engagement/involvement, and 6. Choose the appropriate methodological approaches. These were guided by the results from our survey performed to identify the needs of European countries for BoD studies, a narrative overview from four European countries (Belgium, Germany, The Netherlands and Scotland) and the summary of a comparative study of country health profiles with national health statistics.

**Conclusions:** These recommendations as minimum requirements would facilitate efforts by those European countries who intend to perform national BoD studies.

**Background**

*InfAct* (Information for Action) is a European Commission Joint Action on Health Information with 40 partners across the EU Member States aiming to develop a more sustainable EU health information system through improving the availability of comparable, robust and policy-relevant health status and health system performance data [1]. Through a series of three dedicated workshops, this joint action has emphasized the potential role of burden of disease (BoD) assessment and supported countries with an interest in developing a BoD study but who may lack the relevant expertise and capacity. The main aim of the *InfAct* BoD workshops was to promote the use of BoD, and to help countries integrate these methods into their routine public health activities and policy making and to improve the current European Health Information System (EU-HIS). The BoD approach also offers opportunities for improvement in current practice, such as clearer and more concise documentation, and standardization of methodologies between countries, to make BoD assessments within Europe more comparable. BoD approaches in
general provide comparative assessment frameworks, which include the key metrics such as years of life lost (YLL), years lived with disability (YLD), and disability-adjusted life years (DALYs) [2]. These estimates transform standard measures such as prevalence and mortality rates into aggregated or integrated measures, e.g. by applying severity distributions and DW and thus produces new insights and adds value to standard population health assessment. Since BoD estimates combine measures of mortality and morbidity, they allow comparisons to be made between a broad range of conditions. This means that (bearing in mind the limits of the methods) the population health impact of conditions that primarily cause premature loss of life can be compared on a like-for-like basis with conditions that cause prolonged reductions in health. Estimates may also vary considerably between areas of a country, so subnational estimates are important to describe and highlight variations and inequalities. These types of estimates are key resources to use in knowledge exchange processes with the aim of developing proportionate prevention and interventions strategies to improve overall, and inequalities, in population health at national and subnational levels. The BoD approach has many applications and produces useful outputs, which make sense to policy makers even if they do not fully understand the methods used. However, the data requirements are substantial, and the methods used to produce the summary estimates are complex. Multiple assumptions, methodological choices, and often compromises, must be made to integrate information of different types from many sources. For those countries who do not want to undertake the task of preparing their own BoD estimates, it is possible to use readily available and recent national BoD estimates published by the Institute for Health Metrics and Evaluation (IHME). These are based on the well-resourced and long-standing Global Burden of Disease (GBD) study. However, data inputs into the GBD study do not always correspond to what stakeholders at national level consider to be the best available and most up-to-date information. Countries may also disagree with some of the assumptions made to generate BoD estimates. For these and other reasons, countries who have the resources and capability to do so have performed their own BoD studies rather than rely on the GBD [3–6].

The main objective to develop these recommendations was to facilitate those countries planning to start a national burden of disease study.

**Burden-eu Network**

In 2019, the burden-eu COST (European Cooperation in Science and Technology) Action (CA18218) was launched, with an aim to develop a technical platform to integrate and strengthen capacity in BoD assessment across Europe and beyond [7]. The burden-eu COST Action has four priorities for intensified collaborations: (i) increased interaction between existing BoD efforts; (ii) technical capacity building at country level; (iii) a platform to support methodological advances; and, (iv) an actionable understanding of the process underlying knowledge translation [8]. Aforementioned, the *InfAct* project has emphasized the potential role of BoD assessment; supported countries interested in developing a BoD study and improved specific expertise to develop their capacity. This technical platform would support to achieve these objectives and would continue to establish and strengthen the scientific collaborations to integrate BoD approaches in routing public health activities.
Methods

To develop the recommendations to plan a national burden of disease study, we carried out three main activities under InfAct project: 1. a survey was performed to identify the needs of European countries to perform a national burden of disease study (BoD) (additional file 1), 2. the InfAct project partners who are performing their own BoD studies (i.e., Belgium, Germany, The Netherlands and Scotland), were asked to provide an overview of key elements such as data sources used, methodological approaches applied, methodological challenges and related solutions, implication of BoD estimates in health policy and perspectives (additional file 2) as a narrative overview, and 3. using the ‘standard’ GBD metrics available in the GBD 2017 study, we have extracted a series of country health profiles of European countries from IHME website (https://vizhub.healthdata.org/gbd-compare/ and http://ghdx.healthdata.org/gbd-results-tool). We asked the InfAct partners to compare these estimates with their national health statistics (additional file 3).

Based on the results of these studies and the inputs from the experts of this domain, we developed these recommendations.

Results

Outputs of burden of disease activities performed under InfAct

We performed a survey in May 2019 among European countries to identify the current needs to perform BoD studies (Additional file 1). Among 25 participating countries, 72% mentioned that they have not carried out any BoD study in the past and have no experience from which to develop a case study on BoD. A few have already performed national BoD studies (i.e. Belgium, Germany, The Netherlands and Scotland). These countries were asked to provide a narrative overview of their experience of performing BoD studies (Additional file 2) [9]. These countries have calculated, or are calculating, BoD estimates not only at the national level but also at subnational levels. Their experience could support and guide others to initiate and integrate the burden of disease approaches into their routine public health activities. Then, we performed a study comparing the country health profiles (i.e., providing a measure of priority health conditions and risk factors, a summary breakdown of major causes, and an appreciation of health sector performance) from the GBD study with national health statistics [10] (Additional file 3). Many important differences were highlighted because countries were using different data sources compared with the GBD study, and different methods (such as differences in the standard population used in age-standardized rate calculations).

Recommendations

In collaboration with experts from InfAct project and burden-eu network, we have developed some recommendations for countries who are planning to develop national BoD studies. These recommendations could be considered as minimum requirements for those countries planning to start a
BoD study. These were guided by the results from our survey performed to identify the needs of European countries for BoD studies, a narrative overview from four European countries (Belgium, Germany, The Netherlands and Scotland) [9] and the summary of a comparative study of country health profiles with national health statistics [10]. Wide adoption of these recommendations could help harmonise and facilitate efforts by those European countries who intend to perform their national BoD studies.

1. Define the objectives of a burden of disease study within the context of your country

The BoD study is “a systematic, scientific effort to quantify the comparative magnitude of health loss due to diseases, injuries, and risk factors by age, sex, and geographies for specific points in time“, or, “a comparative assessment framework, which includes the key metrics of years of life lost (YLL), years lived with disability (YLD), and disability-adjusted life years (DALYs)” [2]. However, European countries need to define the objectives of the BoD study within the context of their country. While the standard definition of a BoD study is based on the use of DALYs for quantifying the population health impact of all relevant diseases and risk factors, we acknowledge that not all countries have the resources to achieve this comprehensive assessment, and may therefore embark with a more restricted scope, e.g. study a limited number of diseases, or the use of simple measure of population health such as disease prevalence or life expectancy (LE).

2. Identify, communicate and secure the benefits of performing national burden of disease studies

Policymakers need to be informed about the relative scale of different health problems in the population, the groups that are particularly at risk, and the trends in the state of health over time. In addition, a representative estimate of the population’s health status can be used for determining the expected health care use and is vital evidence to use when prioritizing effective interventions and evaluating their impact and cost-effectiveness [11].

The following is a list of some potential uses of BoD estimates:

- Health and policy improvement
  - Drawing attention to the effects of non-health outcomes on overall population health and rank the impact of diseases on population health expressed in terms of lost life years due to illness (YLD) and death (YLL) in a single summary measure (DALY)
  - Comparing the health of one national or subnational population with that of another (including international benchmarking).
  - Monitoring changes over time in the health of a given population.
  - Identifying and quantifying health inequalities within countries.
Priority setting by health condition and risk factor

- Informing debates on priorities for health service delivery and planning.
- Informing debates on priorities for research and development.

Resource allocation

- Rational and proportionate allocation of resources: trends in specific conditions and differences in outcomes across ages and between sexes can yield insights about where new investments in health resources are needed.
- Analyzing the potential benefits of health interventions for use in cost-effectiveness analyses [12]

Data improvement and quality of the Health Information System

- Performing a national BoD study helps to appraise and improve the completeness and quality of available data to be used, consequently this helps to improve the country’s health information system.

Helping to build capacity

- Performing a BOD study helps to build capacity in BOD assessment, and population health and epidemiology in general
- Relevant training programmes and workshops can increase awareness and build local capacity and expertise to use BoD methods.

Strengthening collaborations

- BoD work can be used to strengthen collaborations with other countries and international organisations (such as WHO, IHME and through the burden-eu COST Action) to integrate and strengthen capacity in BoD assessments across Europe and beyond [8]

3. Secure access to the minimum required data sources

As a minimum data requirement to perform a BoD study, high quality cause specific mortality data (best differentiated at the level of three- or four-digit ICD-10 codes) and other disease-specific statistics are needed. These may include one or more of the following sources: national health administrative data sources (general practitioner registration, hospital discharge data and/or health insurance data), available disease-specific registries, health surveys, vital and causes of death statistics (i.e. census, birth, and death registries). Additional information can be integrated from the scientific literature or from the GBD study. Data used could be either linked or unlinked, at the individual level or at an aggregated level. It is essential to use the best available high-quality data. Which data sources are needed also depends on the objectives of the analyses. As a first step, causes of death and vital statistics can be used to calculate YLL, and later national health administrative data sources, registries, or health surveys can be used to determine disease prevalence and YLD.
4. Ensure the minimum required capacity and capability is available to carry out burden of disease study

BoD assessments are a collaborative effort, particularly when they are performed across a widespread range of health conditions and risk factors. It provides an opportunity to mobilise the scientific knowledge and competencies with a multidisciplinary approach and networking from various domains. To perform a BoD study, the minimum required workforce would include epidemiologists, data managers, biostatisticians/statisticians, public health experts and demographers.

5. Establish a clear governance structure for the burden of disease study and stakeholder engagement/involvement

National public health institutes are ideally placed to be responsible for or for co-ordinating, national BoD studies in collaborations with various partners to share their expertise. Stakeholder engagement is important at every stage [13] to share information, coordinate and obtain experts opinion on BoD indicators.

6. Choose the appropriate methodological approaches

Countries undertaking their own analyses need to select appropriate approaches that fit within their country contexts. When making methodological choices about BoD methods, one should be aware of certain standardized methods proposed by the IHME if comparability with GBD is an aim. If country and subnational contexts are different from the GBD study, then these estimates will retain strong within-country value but their utility in comparison with GBD estimates are limited. This may not matter depending on the purpose of the estimates. Some reference guides are available that can help countries to follow various methodological approaches. For example, WHO 2001 a practical guide on national BoD studies [14].

A recent study highlighting the key methodological decisions in national BoD assessment [15], the narrative overview of national BoD studies [9] and the comparison of country health profiles with national health statistics [10] have emphasized the importance of the following aspects of the methods used:

- Methodological choices comprise (some of these are optional ones, others demand a choice):
  - Use of national or GBD (standard/ideal) life tables to estimate YLL [16]
  - Methods used to redistribute garbage codes/ill-defined codes and invalid ICD-10 codes when making counts of death by cause [4, 17]
  - Use of bespoke national or GBD disability weights and severity distributions when calculating YLDs [18]
  - Multimorbidity adjustment method [19]
Methods for estimating uncertainty levels is optional whether to quantify uncertainty, and if so, whether to use a quantitative or qualitative approach.

Distribution of potential risk factors in population by age, sex and geographical level to calculate the relative risk (RR), attributable fraction (AR) and population attributable fraction (PAF) and to calculate risk attribution to disease burden.

- Geographic level (i.e., municipalities, metropolitan, subnational and national level).
- Choice of a reference population used in standardized rate calculations: For European countries, it is likely to be most appropriate to use the 2013 European standard population (ESP2013) [20]. For within country comparisons a local standard population may be more appropriate especially if the country has a markedly different age/sex structure from the rest of Europe.

- Data standards should align with existing data and metadata standards where appropriate: i.e. WHO approved terminologies/ontologies including ICD-10.
- BoD indicators should be reported according to the GATHER (Transparency, as per Guidelines for Accurate and Transparent Health Estimates Reporting) guidelines [21]

Discussion

Current focus and remaining challenges

The current focus of these recommendations is to provide an opportunity to mobilise existing scientific knowledge and competencies with a multidisciplinary approach and to encourage networking from various domains at European and international levels. The proposed recommendations could be considered as minimum requirements for those countries planning to start a BoD study. To implement and to integrate BoD approaches in steering public health activities, the technical platform of burden-eu COST Action would provide operational support to achieve these objectives and would continue to strengthen scientific collaborations.

The previous study on comparisons of country health profiles with national health statistics has highlighted the importance of key aspects such as differences in data sources, choice of a standard population in standardized rate calculations, and the differences between methods used to calculate BoD estimates when developing BoD studies [10]. These are the key areas where countries could invest to improve the quality of available data sources for reliable estimates and to build their capacity and skills for calculating BoD estimates at a small scale (i.e., including minimum number of diseases).

The strength of this study is that these recommendations were reviewed by BoD experts and may facilitate efforts to start, or to integrate, the BoD approach in routine public health activities. These recommendations were developed based mainly on the current experience of using BoD approaches in the national public health institutes of European Union countries and did not take into account the full experience of those research institutes in each country who may be involved in additional BoD activities.
Conclusions

The InfAct project is a European Commission Joint Action on Health Information, which has promoted the potential role of the burden of disease (BoD) approaches to improve the current European Health Information System (EU-HIS). The InfAct BoD workshops have raised the awareness of the concept, the methods used to calculate estimates and their implications and uses in policymaking. The BoD is a collaborative effort built on shared experiences and techniques. The BoD approach is a systematic and scientific effort to quantify and compare the magnitude of health loss due to different diseases, injuries, and risk factors with estimates produced by demographic characteristics and geographies for either a single geographical location or several locations of interest for specific points in time. We acknowledge that not all countries have the resources to undertake such work, and may therefore start with a more restricted scope, e.g., a limited number of diseases, or the use of simple measures of population health such as disease prevalence or life expectancy. For those countries planning to start a BoD study, these recommendations and adoption of these proposed minimum requirements, would promote to harmonise and facilitate efforts by European countries.

Abbreviations

InfAct: Information for Action (i.e., joint action of European countries to establish a more sustainable health information system)

BoD: Burden of Disease

EU-HIS: European Health Information System

IHME: Institute for Health Metrics and Evaluation

GBD: Global Burden of Disease

DALYs: Disability Adjusted Life Years

YLL: Years of Life Lost due to premature mortality

YLD: Years Lived with Disability

ESP2013: European Standard Population 2013

ICD: International Statistical Classification of Diseases and Related Health Problems

WHO: World Health Organization

GATHER: Guidelines for Accurate and Transparent Health Estimates Reporting

Declarations
Ethics approval and consent to participate

Not applicable

Consent for publication

All authors gave the consent for publication.

Availability of data and materials

Not applicable

Competing interests

H. Van Oyen is one of the co-authors of this paper and the editor in chief of “Archives of Public Health”. B. Devleesschauwer and H. Hilderink are the co-authors of this paper and are the editors of article collection on “burden of disease” of “Archives of Public Health. R. Haneef is the first author of this paper and the section editor of “health information system” of “Archives of Public Health”. All other authors declare that they have no competing interests related to the work.

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Authors’ contributions

Conceived and designed the study: RH JS AG BD HH TZ JN. Performed the study: RH JS. Analysis of the data, interpretation of the results and writing of the manuscript: All authors contributed. All authors read and approved the final manuscript.

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References

1. INFACT: Joint Action on Health Information: https://www.inf-act.eu/. 2018.
2. Murray CJL, Ezzati M, Flaxman AD, Lim S, Lozano R, Michaud C, Naghavi M, Salomon JA, Shibuya K, Vos T et al: GBD 2010: design, definitions, and metrics. The Lancet 2012, 380(9859):2063-2066.
3. Melse JM, Essink-Bot ML, Kramers PG, Hoeymans N: A national burden of disease calculation: Dutch disability-adjusted life-years. Dutch Burden of Disease Group. Am J Public Health 2000, 90(8):1241-1247.
4. Grant I, Mesalles-Naranjo O, Wyper G, Kavanagh J, Tod E, Fischbacher C, McCartney G, Stockton D: Burden of Disease study in Scotland: https://www.nrscotland.gov.uk/files//statistics/rgar-invited-chapter/rgar17-invited-chapter.pdf. 2017.
5. Devleesschauwer B, Renard F: Belgian National Burden of Disease study: https://www.sciensano.be/en/projects/belgian-national-burden-disease-study. 2016.
6. Rommel A, von der Lippe E, Plaß D, Wengler A, Anton A, Schmidt C, Schüssel K, Brückner G, Schröder H, Porst M et al: BURDEN 2020—Burden of disease in Germany at the national and regional level. Bundesgesundheitsblatt - Gesundheitsforschung - Gesundheitsschutz 2018, 61(9):1159-1166.
7. COST A: European Burden of Disease Network: https://www.burden-eu.net/. 2019.
8. Devleesschauwer B: European burden of disease network: strengthening the collaboration. European Journal of Public Health 2020, 30(1):2-3.
9. INFACT: Narrative overview of National Burden of Disease studies: A case study report: https://www.inf-act.eu/sites/inf-act.eu/files/2020-12/D9.4%20A_Narrative%20overview%20of%20national%20BoD%20studies.pdf 2020.
10. INFACT: Comparison of Country Health Profiles (i.e., GBD metric) with National Health Statistics in European Countries: https://www.inf-act.eu/sites/inf-act.eu/files/2020-12/D9.4%20B_Comparison%20of%20Country%20Health%20Profiles.pdf 2020.
11. Devleesschauwer B, Maertens de Noordhout C, Smit GSA, Duchateau L, Dorny P, Stein C, Van Oyen H, Speybroeck N: Quantifying burden of disease to support public health policy in Belgium: opportunities and constraints. *BMC Public Health* 2014, 14(1):1196.

12. Murray CJL, Salomon JA, Mathers CD: A critical examination of summary measures of population health / Christopher J. L. Murray, Joshua A. Salomon and Colin Mathers. *Examen critique des mesures synthétiques de l’ état de santé d’ une population : résumé* 2000.

13. Delnord M, Tille F, Abboud LA, Ivankovic D, Van Oyen H: How can we monitor the impact of national health information systems? Results from a scoping review. *European Journal of Public Health* 2019, 30(4):648-659.

14. WHO: National Burden of Disease Studies_A Practical Guide: https://www.who.int/healthinfo/nationalburdenofdiseasemanual.pdf. 2001.

15. von der Lippe E, Devleesschauwer B, Gourley M, Haagsma J, Hilderink H, Porst M, Wengler A, Wyper G, Grant I: Reflections on key methodological decisions in national burden of disease assessments. *Archives of Public Health* 2020, 78(1):137.

16. Devleesschauwer B, McDonald SA, Speybroeck N, Wyper GMA: Valuing the years of life lost due to COVID-19: the differences and pitfalls. *International Journal of Public Health* 2020, 65(6):719-720.

17. Wengler A, Gruhl H, Plaß D, Leddin J, Rommel A, von der Lippe E: Redistributing Ill-defined Causes of Death – A Case Study from the Burden 2020-Project in Germany: https://www.researchsquare.com/article/rs-117779/v1. 2021.

18. Wyper GMA, Grant I, Fletcher E, Chalmers N, McCartney G, Stockton DL: Prioritising the development of severity distributions in burden of disease studies for countries in the European region. *Arch Public Health* 2020, 78:3.

19. Hilderink HB, Plasmans MH, Snijders BE, Boshuizen HC, Poos MJ, van Gool CH: Accounting for multimorbidity can affect the estimation of the Burden of Disease: a comparison of approaches. *Arch Public Health* 2016, 74:37.

20. Wyper GMA, Grant I, Fletcher E, McCartney G, Fischbacher C, Stockton DL: How do world and European standard populations impact burden of disease studies? A case study of disability-adjusted life years (DALYs) in Scotland. *Arch Public Health* 2020, 78:1.

21. Stevens GA, Alkema L, Black RE, Boerma JT, Collins GS, Ezzati M, Grove JT, Hogan DR, Hogan MC, Horton R et al: Guidelines for Accurate and Transparent Health Estimates Reporting: the GATHER statement. *Lancet* 2016, 388(10062):e19-e23.