Proposed guidelines for open source health economic models and a framework for applying them in youth mental health

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Abstract

Summary: Despite growing support for open source health economic models (OSHEMs), uptake remains limited and better guidance about how to implement them is required. One potential benefit of high quality OSHEMS is the development of more valid, useful and extensive models to inform the design of population mental health strategies. In this article, we describe a framework that we have developed for an OSHEM in youth mental health. The framework includes a set of 20 good practice standards and toolkits for developing models that meet those standards. Framework standards are intended to support the development of OSHEMS that are TIMELY - Transparent (6 standards), Iterative (4 standards), Modular (2 standards), Epitomising (3 standards), Licensed (3 standards) and Yielding (2 standards). Framework toolkits include repositories for model data and code as well as six novel R packages. The R packages provide a novel syntax, template modules and modelling tools for authoring model modules, function libraries, datasets and reports. We discuss the rationale for each standard included in our framework and provide a worked example of the application of the framework to implement a utility mapping study.

 $\begin{tabular}{lll} \textbf{Code:} & Development versions of all code repositories referenced in this article are available in $$https://github.com/ready4-dev/$. Archived code releases are available in $$https://zenodo.org/$ communities/ready4 . \end{tabular}$

 $\textbf{Data:} \quad \text{All data repositories referenced in this article are available in $$https://dataverse.harvard.edu/dataverse/ready4 .}$

1 Introduction

Computational models have become essential tools for health policy development [1,2]. Although influential and widely used, these models routinely contain errors [3], are rarely adequately validated [4] and can be difficult to reproduce [5–7]. To help address these issues, there is support for greater use of open source

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health economic models (OSHEMs) that grant liberal permissions to access and re-use model source code and data [8].

Complex modelling projects may be more tractable when multiple modelling teams re-use, test and extend one another's work [9]. OSHEMSs might therefore mitigate some barriers to improving the longevity and reach of some health economic models. For example, health economic models should be updated and refined as new evidence emerges and decision contexts change [10], but this occurs infrequently [11]. Reference models that can be re-used in multiple health economic modelling projects have been advocated for [12] but require greater investments in model transparency and validation [13,14].

Despite the potential benefits of OSHEMs, actual implementations are rare [14–16]. Barriers to adoption of OSHEMs include concerns about intellectual property, confidentiality, model misuse and the resources required to support open source implementations [8,17]. As many health economic models are owned by pharmaceutical companies and consultancies, commercial considerations may also limit the uptake of OSHEMs [14]. Adherence to good practice guidance is an essential requirement for healthcare modelling [2], but guidelines for implementing OSHEMs remain scarce, piecemeal and need improving [18].

In this paper, we describe:

- a proposed set of guidelines for quality implementation of OSHEMS;
- a framework for applying these guidelines to the development of a reference OSHEM in youth mental health;
- a worked example of a modelling project developed with the framework.

2 Guidelines

Guidelines on health economic model transparency were published ten years ago [13] and made recommendations on documenting models; however, notably missing were recommendations on the sharing of model code and data. More recent and more general modelling guidance [2] recommends the sharing of code and data through digital repository services and the use of version control systems. Based on these and other existing sources of guidance and our own experience, we propose 20 guidelines for implementing OSHEMs that are **TIMELY**:

- Transparent: people can easily see how a model has been implemented and tested;
- Iterative: a model is routinely updated to maintain and improve validity;
- Modular: models and their components can be combined to extend their scope;
- Epitomising: model code is sufficiently general to be be re-used in multiple decision contexts;
- Licensed: a model, its components and derivatives are persistently re-usable by other modellers; and
- Yielding: a model can be simply, flexibly and reliably used as a decision aid.

2.1 Transparent Models

A range of tools and practices are available to help make model code and data accessible, comprehensible and citable. The most efficient way to widely disseminate code and data may be to use existing open science infrastructure [2] to make permanently accessible and uniquely identified digital artefacts. Model code and data also need to be clearly documented, potentially with different versions for technical and non-technical users [13]. Consistent use of meaningful naming conventions when authoring code is recommended [19,20]. Code can be made easier to follow by using the practices of abstraction [21], where only simple, high level commands are routinely exposed to reviewers, and polymorphism [22], where the same command (e.g. "simulate") can be reused to implement different algorithms of the same type. Programs to implement model analyses can be made comprehensible to even non-technical users through the use of literate programming techniques to render documents that integrate computer code with plain English descriptions.

An essential component of quality assuring health economic models is verification - ensuring that calculations are correct and consistent with model specifications [23]. One useful concept for informing model users about the extensiveness of verification checks is code coverage [24] - the proportion of model code that has been explicitly tested. Finally, transcription errors - mistakes introduced when transferring data between sources, models and reports - are very common in health economic models [3]. The risk of these errors might be lower if there was full transparency across all steps in a study workflow. Scientific computing tools now make it relatively straightforward to author programs that reproducibly execute all steps in data ingest, processing and reporting [19].

Guidelines:

- T1: Uniquely identified copies of model code and data are permanently archived in open online repositories
- T2: Model code and data are documented
- T3: Model code uses a simple and consistent syntax
- T4: Model analyses and reporting are implemented using literate programming
- T5: Code coverage is reported
- T6: All parts of a study analysis and reporting workflow can be reproduced and/or replicated

2.2 Iterative Models

To avoid OSHEMs going stale - losing validity and usefulness with time - they should be routinely updated. Each update of code and data should be uniquely identifiable and retrievable, a goal that can be facilitated by use of version control tools [2]. Labeling each change using semantic versioning [25] conventions can signal the potential importance of an update to users of model code and data. Continuous integration [26] tools can help verify that each code update passes multiple quality tests. Finally, using deprecation conventions that take an informative and staged approach to retiring old code and data reduces the risk that model revisions have unintended consequences on third party users. ->

Guidelines:

- I1: Model code and data are version controlled
- I2: Model code and data use semantic versioning
- I3: Continuous integration is used to verify model code updates
- I4: Deprecation conventions are used to retire model code and data

2.3 Modular Models

Modular health economic models link multiple self-contained components that can be independently reused and extended by other projects [27,28]. Many types of mental health data are highly sensitive with strict confidentiality requirements. For this reason, not all data included in some mental health models can be made widely available for others to re-use. A modular approach that ensures that model code and data are decoupled (stored in different files) can help model developers to restrict access to confidential model data, while providing open access to all other model components. An important consideration when combining model components (or modules) is to ensure that interactions between two modules do not compromise the validity of either. Using the coding practice of encapsulation [21] can help ensure that model modules can be safely combined [29].

Guidelines:

- M1: Model code and data are stored and managed separately
- M2: Model code defines encapsulating data structures

2.4 Epitomising Models

A key challenge to generalising health economic models is that they are typically developed to inform a decision problem with a highly specific jurisdictional context. However, a number of choices about how these models are implemented can significantly increase the re-usability of model code in other contexts.

Writing code as collections of functions (short, self-contained and reusable algorithms that each perform a discrete task) is recommended as good practice for scientific computing [19]. When bundled for distribution as libraries, functions have the potential to be widely re-used, often in contexts very different than those they were originally developed for. A special type of function, called a method, can only be applied to a pre-defined class of data structure. Due to the coding concept of inheritance [21], the more restricted nature of methods can be used to enhance the re-usability of model code in different decision contexts [29]. For example, when generalising a model developed for the Australian context to a UK context, one could create a class that initially inherits all of the methods defined for the Australian model and then write new or replacement methods as needed for the UK model. Whatever type of functions are written for a modelling project, it is good practice to make available test or toy data to demonstrate their use [19].

Guidelines:

- E1: Model code is distributed as libraries of classes and functions
- E2: Model code defines inheriting data-structures
- E3: Test data is available to demonstrate generalised applications of model code

2.5 Licensed Models

To make model code and data widely re-usable by others, it is important to provide users with appropriate and explicit permissions. In the context of open source models, there are two broad categories of licensing options. Some guidance strongly recommends the use of permissive licensing [19] that provides users with great flexibility as to the purposes (including commercial) for which the content could be re-used. An alternative approach is to use copyleft licenses [30] that can require content users to distribute any derivative works they create under similar open source arrangements. The most suitable approach may differ for code and data.

For code, it may be appropriate to adopt the prevailing open source licensing practice within the programming language being used. For data, it may not be sufficient to simply choose between a permissive license like the Public Domain Dedication (CC0) [31] or a copyleft option such as the Attribution-Share Alike (CC-BY-SA) [32]. Responsible custodianship of some de-identified mental health data may involve using or adapting template terms of use [33] which have a number of ethical clauses (for example, prohibiting efforts to re-identify research participants). Licenses may or may not specify that model re-users must give appropriate acknowledgement to model authors. Citation tools can be distributed with each individual code or data item to inform re-users of the desired attribution.

Guidelines:

- L1: Model code is made available for re-use under copyleft or permissive licenses
- L2: Non-confidential model data is licensed for liberal re-use (subject to additional terms for de-identified human data)
- L3: Model code and data are distributed with tools to support appropriate citation

2.6 Yielding Models

OSHEMs can be time and skills intensive for modellers to develop - but they should be easy for others to use. Statistical models are a common output of health economic evaluations, but they are often not reported in a format that enables others to confidently and reliably re-use them [34]. Open source approaches can help address this by disseminating code artefacts that enable easy and appropriate use of a statistical model to make predictions with new data. However, great care must be exercised when doing so if models are derived from data on human subjects as some software artefacts by default contain a copy of the source dataset. Such dataset copies must therefore be replaced (for example, with synthetic data) and the amended artefact's predictive performance then retested before any public release. Another way to make OSHEMs easier to use is to develop simple user-interfaces for non-technical users.

Guidelines:

- Y1: Statistical models are distributed with validated tools to support their safe and appropriate re-use
- Y2: Simple user-interfaces allow non-technical users to configure and run models

3 Framework

3.1 readyforwhatsnext

Our approach to model development is to undertake a number of discrete modelling projects of the people, places, platforms and programs that shape the mental health and wellbeing of young people and to progressively link them together by means of a common framework. To model people we are developing synthetic representations of populations of interest [35] that describe relevant individual characteristics and their relationships, algorithms that map psychological measures to health utility [36] and choice models for predicting the helpseeking behaviour of young people [37]. Our in-development model of places [38] has the aim of synthesising geometry and spatial attribute data to characterise the geographic distribution of relevant demographic, environmental, epidemiological and service infrastructure features. We are in the early stages of a multi-annual project to develop a service platform model that will represent the processes and operations of a complex primary youth mental health service. We also plan to extend and update our prior work reviewing economic evidence relating to youth mental health programs [39] so that it can be integrated with this model.

Our initial work on **readyforwhatsnext** is focused on Victoria, Australia but the framework we are using to develop it is designed to facilitate extension by ourselves and others to different jurisdictional decision contexts. Progress is reported on a project website [40].

3.2 Framework toolkits

We have developed a toolkit to help streamline the process of developing OSHEMs that meet the TIMELY standards. The toolkit is comprised of online repositories and software.

3.2.1 Framework repositories

We created a GitHub organisation where all framework software source code is stored, documented, version controlled and continuously integrated [41]. To store citable archived copies of release copies of our software, we created a Zenodo community [42]. Finally, to manage datasets for use in models developed with the framework, we created a dedicated collection within the Harvard Dataverse [43].

3.2.2 Framework software

A coding framework for OSHEMs developed in the language R includes standardised approaches to directory structure and naming conventions [20].

As a foundation for implementing the framework, we authored a development version R package that defines a novel syntax and a template class for model modules [44]. To enable the syntax and module template be applied to modelling projects, we then created five additional development version R packages that provide tools for authoring:

- documented model modules [45];
- documented functions (including methods), written in a consistent house style [46];
- citable, quality assured R packages [47];
- model datasets [48];
- model analyses and reports [49].

The six R packages, their primary focus, the TIMELY standards they support and the third-party packages they depend on are summarised in Table 1. When used in conjunction with framework repositories, the six packages extend existing R packages provide strong support for implementing 16 of the TIMELY standards. However, the software only weakly supports implementing the standards relating to disseminating statistical models (Y1) and user-interface development (Y2) and does not yet provide any workflow tools to help implement the standards for code coverage (T5) and deprecation conventions (I4). Standards not supported or weakly supported by our software can be met with existing developer tools in R and we plan to progressively integrate these third-party tools with our own in future releases of our software. Another future priority is to submit production versions of each R package for review by and archiving on the Comprehensive R Archive Network (CRAN) [50].

Table 1: Framework software to enable the ready forwhatsnext model to meet TIMELY guidelines

Package	Focus	Guideline	Depends on these R packages
ready4	Syntax	T3 M3	assertthat bib2df dataverse dplyr fs generics Hmisc kableExtra knitr lifecycle magrittr methods piggyback purrr rlang rvest stats stringi stringr testit testthat tibble tools utils
ready4fun	Functions	T2-3 E1	desc devtools dplyr Hmisc knitr lifecycle lubridate magrittr methods piggyback pkgdown purrr readxl ready4 ready4show ready4use rlang sinew stats stringi stringr testit testthat tibble tidyr tools usethis utils xfun
ready4class	Modules	T2-3 M2 E1-2	devtools dplyr fs gtools Hmisc knitr lifecycle magrittr methods purrr ready4 ready4fun ready4show rlang stats stringi stringr testit testthat tibble tidyr usethis utils
ready4pack	Libraries	T1 I1-3 M1-2 E1 L1,3	dataverse dplyr knitr lifecycle magrittr methods purrr ready4 ready4class ready4fun rlang stringr testthat tibble tidyr utils
ready4use	Data	T1-2 M1 E3 L2-3	data.table dataverse dplyr fs Hmisc knitr lifecycle magrittr methods piggyback purrr readxl ready4 ready4show rlang stats stringi stringr testit testthat tibble tidyr utils
ready4show	Reporting	T2,4,6 Y1-2	dataverse DescTools dplyr flextable grDevices here Hmisc kableExtra knitr knitrBootstrap lifecycle magrittr methods officer purrr ready4 rlang rmarkdown stringi stringr testthat tibble tidyr utils xtable

Table 2: TIMELY Checklist applied to utility mapping study

	Standard	Met?	Description
T1	${ m Publicly} \ { m archived}$	Yes	5 libraries (youthvars, scorz, specific, TTU and youthu) [51–55], 3 programs [56–58], 2 sub-routines [59,60] and 2 datasets [35,61] are normanoutly archived with unique identifiers
T2	Documented	Yes	All code libraries have documenting websites with URLs that concatenate 'https://ready4-dev.github.io/' and the
			package name (e.g. https://ready4-dev.github.io/youthvars). All three Markdown programs are self-documenting, with one [56] including additional instructions in a README file. Only one sub-routine [60] is documented with a meaningful README file. All datasets have meaningful metadata descriptors.
T3	Consistent	Yes	All libraries, programs and sub-routines use the same house style, which allows most library documentation to be written by algorithms from the ready4fin package [46]. All libraries except [55] use framework syntax, as does one program [56].
T4	Literately	Yes	All programs use literate programming.
T5	programmed Code	$_{ m o}^{ m N}$	No current reporting of code coverage.
9L	coverage Reproducible	Yes	All parts of the study workflow from raw data ingest through to data processing, analysis, reporting and dissemination of
ř). P	
	Version $controlled$	Yes	All code is version controlled using Git and GitHub. All source code is available in a GitHub organisation [41].
12	Semanticly	Yes	Semantic version is used in all code. As no code library has yet been submitted to CRAN, only the development version
	versioned		extensions of each version number have been incremented to date.
I3	Continuously integrated	Yes	All six libraries use continuous integration (CI). CI results for each library can be viewed at a URL that concatenates 'https://oithub.com/readv4_dev/' the nackage name and '/actions'
	mogranoa		(e.g. https://github.com/ready4-dev/youthvars/actions)
1 4	Deprecation	Yes	Retired code is deprecated using lifecycle package tools (e.g. everything after "## DEPRECATED FNS" in https://github.com/ready4-dev/youthvars/blob/main/data-raw/fns/add.R.). Package vignettes and datasets are also deprecated e.g. https://ready4-dev.github.io/youthvars/articles/Replication_DS.html)
M1	Separate code and	Yes	All development code is stored on repos in a GitHub organisation [41] and all archived releases are available in a Zenodo community [42]. All non-confidential data is stored in repositories within a Harvard Dataverse collection [43].
M2	data Encapsulated	Yes	Four [51–54] out of five libraries include encapsulated modules. Examples are the items beginning with Scorz, Specific
			and Youthvars that are listed in this table: https://ready4-dev.github.io/ready4/articles/V_01.html#current-ready4-framework-modules as well as the S4 classess from the TTH nackage listed here: https://readv4.dev.github.io/TTHT/reference/index.html
E1	Libraries	Yes	All code libraries include functions. The most complete list of functions for each library is available by clicking the 'Manual - Developer (PDF)' link on each package's documentation homepage (see item T2 above).

Met? Description	All modules (see item M2) inherit from the Ready4Module class and can be inherited from. Two synthetic datasets and their data dictionaries are publicly available in a data repository [35]. One (ymh_clinical_tb.RDS) closely resembles the study dataset and was released so that the main study algorithm [56] can be rerun by those without access to the confidential study dataset. The other (eq5d_ds_dict.RDS) is deliberately different to the source dataset in both variable naming convention and the concepts used for predictors and outcome measures. It was created to demonstrate generalised applications of study algorithms.	All code libraries, programs and sub-routines use GPL-3 licenses.	Datasets use amended version of template provided by Harvard Dataverse [33].	All libraries have a CITATION file in the inst directory. All code repositories have a CITATION.cff file. All datasets have citation generating metadata.	Model catalogues (PDF files beginning with 'AAA_TTU_MDL_CTG') are available in the study results dataset [61] and describe the predictive performance of all models under a variety of usage regimes (including when the source dataset in the R model object is replaced with fake data). The youthu library [55] includes tools for searching for and applying models compatible with different types of input data. An example program to demonstrate this functionality is available in both RMarkdown [57] and rendered PDF formats (the 'Application.pdf' file in the study results dataset [61]).	
Met?	Yes	Yes	Yes	Yes	Yes	$N_{\rm o}$
Standard	Inheriting Test data	Copyleft code	Liberal, safe data terms	Citation tools	Prediction tools	User interface
	E3	L1	L2	L3	Y1	Y2

4 Application

We used framework toolkits to develop model code and datasets for implementing a utility mapping study that has previously been described [36]. Table 2 assesses that study against each TIMELY standard, 18 of which the study was able to meet. The two current standards where the study falls short are in reporting code coverage and including a user-interface. Both these items are current development priorities for our team.

5 Discussion

Mental disorders impose high health, social and economic burdens worldwide [62,63]. Much of this burden is potentially avertable [64], but poorly financed and organised mental health systems are ill-equipped for this challenge [65,66]. The large and widespread additional mental health burdens recently observed during the COVID-19 pandemic [67] and predicted as a potential future consequence of climate change [68], highlight the need to improve the resilience and adaptability of these systems. To help stem growing demand for mental health services, policymakers have also been encouraged to place greater emphasis on tackling the social determinants of mental disorders [69].

Major mental health reform programs, can involve the identification, prioritisation, sequencing, targeting and monitoring of multiple interdependent initiatives.

The significant deficits in our understanding of the systems in which mental disorders emerge and are treated [70] suggest that there is ample scope for mental health systems models to progressively improve their validity over time. Prospective work could address the weak theoretical underpinnings for understanding complex mental health systems [71]. For example, it remains unclear why increased investments in mental health care have yet to discernibly reduce the prevalence and burden of mental disorders[72]. The literature, and evidence base, regarding how the requirements, characteristics and performance of mental health services are shaped by spatiotemporal context needs to be further developed [73]. There is also a need for better evidence to identify the social determinants of mental disorders most amenable to preventative interventions, and for which population sub-groups such interventions would be most effective [74].

The development, validation and updating of more complex mental health economic models implemented over longer time frames may be too onerous a burden for a single modelling team.

Developing networks of modellers working on common health conditions has been recommended as a strategy for improving model validity [18] and some of us are part of a nascent initiate of this type in mental health [75]. Similarly, developing partnerships between modellers and decision-makers across the life-cycle of a modelling project can help ensure models are appropriately conceptualised, implemented and have practical utility as decision aids [76,77].

6 Conclusion

We have identified a number of standards that we believe are appropriate to implementing quality OSHEMs in youth mental health. Most of these standards are probably relevant to OSHEMs in other health conditions, though some such as the copyleft licensing may be less relevant to modellers using different tools. Our framework toolkits can help support standardised approaches to dynamic systems model development that are important for collaborative and interdependent projects.

Availability of data and materials

Development versions of all code repositories referenced in this article are available in https://github.com/ready4-dev/. Archived code releases are available in https://zenodo.org/communities/ready4.

 $All \ data \ repositories \ referenced \ in \ this \ article \ are \ available \ in \ https://dataverse.harvard.edu/dataverse/ready4$

Ethics approval

Framework development did not involve human subject research and was not ethically reviewed. The worked example of framework application is a previously reported study that was reviewed and granted approval by the University of Melbourne's Human Research Ethics Committee, and the local Human Ethics and Advisory Group (1645367.1).

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Conflict of Interest

None declared.

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