Is Bayesian Statistics an Emerging Way for Extrapolating Survival in Cost-Effectiveness Analysis: A Systematic Review

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Introduction

In health technology assessment (HTA), cost-effectiveness analysis (CEA) evaluates the clinical and economic impact of health interventions, requiring long-term survival estimates when clinical trials have limited follow-up data. As highlighted in the NICE technical support document 21 (2020) [1], Bayesian methods offer a flexible and robust framework for survival modelling by integrating external data and addressing uncertainty. This work aims to report how Bayesian is used to incorporate external information for extrapolating long-term survival in CEA.

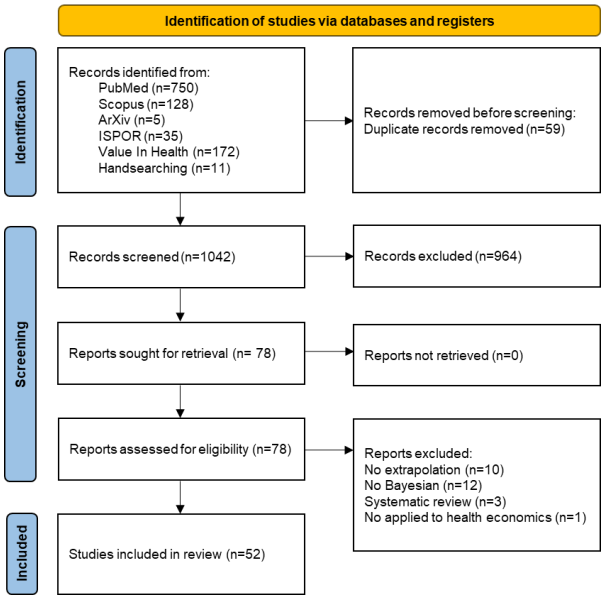
Methods

We conducted a systematic review up to October 2024 to identify both methodological and non-methodological studies using Bayesian methods for survival extrapolation. A search was performed using different electronic databases (PubMed, Scopus, ArxiV), ISPOR conference and Value In Health databases, completed by a handsearch of the references cited in the studies identified automatically.

Results

Of the 52 selected studies (77% automatically and 23% manually) of which 52% were published since 2022, 90% (n=47) focus on oncology (Figure 1).

Figure 1: PRISMA flow chart showing study extraction and selection



52% (n=27) represent articles and 48% (n=25) abstracts. 38% (n=20) are methodological works. 87% (n=45) used external data from different sources: clinical, registry, epidemiology, real world data, general population mortality and 17% (n=9) experts elicitation.

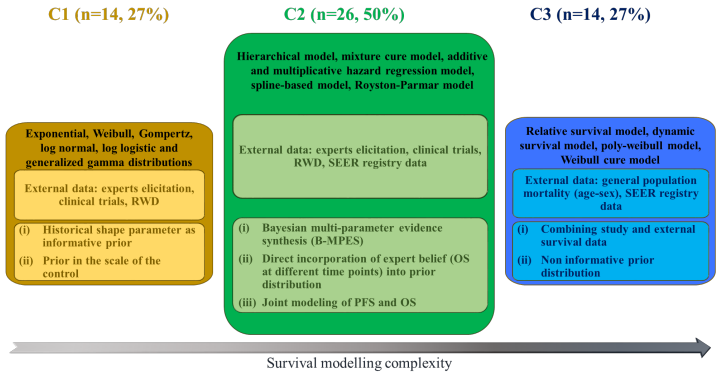
Results

We propose a classification of studies into four non-mutually exclusive categories of Bayesian modeling for survival extrapolation (C1-C4). The first three categories combine, in order of increasing complexity, survival modeling and Bayesian formulation for incorporating external information.

- **C1:** includes standard parametric models (SPM) with prior of parameters informed by historical data [2]. This approach may be considered not flexible enough.
- **C2:** includes primarily Bayesian multiple parameter evidence synthesis that allows to combine trial and external data [3], joint modelling of progression-free survival and overall survival [4], and non-SPMs.
- **C3:** groups complex hazard regression models that account for both disease-specific and expected mortality from general population mortality.

More information is provided in Figure 2.

Figure 2: Bayesian approaches for survival extrapolation with external information incorporation



RCT: randomised clinical trial, RWD: real world data, SEER: Surveillance, Epidemiology, and End Results, OS: overall survival, PFS: progression free survival

- **C4 (n=7, 13%):** Bayesian model averaging which combines survival predictions estimated from different survival models, weighted by posterior model probabilities.

R packages for survival extrapolation include **survextrap** [5] (hazard is model using M-spline function) and **survHE** [6] and for expert elicitation: **expertsurv** [7] (incorporation of expert opinions about survival probabilities at multiple time points into parametric models) and **shelf** [8] (elicitation framework).

Conclusion

This review highlights the broad spectrum of Bayesian survival models and their utility in incorporating external information to reduce uncertainty in survival extrapolation for CEA. Future research should focus on comparing these methods to identify the most suitable approach, based on intervention mechanisms and external data availability. Standardizing Bayesian survival modelling and providing clear guidance, will enhance its application in HTA and improve decision-making frameworks.

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