

A systematic review of the methods for evaluating the benefits and harms of antenatal and newborn screening programmes adopted by health economic assessments

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Abstract

Extensive resources and complex organisational arrangements are required to deliver antenatal and newborn screening programmes in industrialised nations. National screening committees and health technology assessment agencies consider cost-effectiveness assessments alongside viability, accuracy of screening and confirmatory tests, effectiveness of treatments and appropriateness criteria. It is not clear what approaches health economic assessments have adopted to measure and value benefits and harms of antenatal and newborn screening programmes. Therefore, this study aims to systematically review and critique published and grey literature on methods for identifying, measuring and valuing the benefits and harms of antenatal and newborn screening adopted by economic assessments in OECD countries.

The PICOS (Population, Intervention, Comparator, Outcome and Study design) framework was applied to the literature searches. Bibliographic database searches of the published literature were supplemented by manual reference searching of bibliographies, contacts with experts in the field, citation searching and author searching. The grey literature was identified from a pool of relevant websites informed by a recent systematic review of national policy recommendations on newborn screening that identified websites of national and regional screening organisations with documentation about antenatal and/or newborn screening recommendations. The search strategies combined terms for health economic assessments with those for antenatal and newborn screening programmes. Studies were selected for review if they reported health economic assessments of antenatal or newborn screening programmes from any OECD country and published from 2000 onwards. The study selection process followed PRISMA guidelines. Data were extracted into a bespoke form created to understand the handling of benefits and harms by authors, as well as adherence to the Consolidated Health Economic Evaluation Reporting Standards (CHEERS) statement. A protocol for this study was registered in the International Prospective Register of Systematic Reviews (PROSPERO) (registration number CRD42020165236).

Preliminary results suggested that the reporting of information to understand what and how benefits and harms are incorporated in these studies is currently limited. Authors rarely provide sufficient information on their methods to assess how benefits and harms are incorporated in terms of screening outcomes (true

positives, false positive, true negatives and false negatives). The reporting of the structure of underpinning decision models and accompanying modelling assumptions is also poor. Often, authors focus on limited biomedical outcomes within their incremental cost effectiveness ratios that limit comparability across studies. These expressions of cost-effectiveness often do not capture the breadth of benefits and harms modelled by authors.

Findings from this review should inform recommendations about whether the techniques used to measure and value benefits and harms adopted by health economic assessments of antenatal and newborn screening programmes are appropriate or whether a new framework for these economic evaluations is needed.