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School of Public Health

UNIVERSITY OF TORONTO
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# The Future of Screening Criteria for Informing New Screening Programmes

**iPAAC – Innovative Partnership For Action Against Cancer:** New Openings of Cancer Screening in Europe

05 December 2019
Helsinki, Finland

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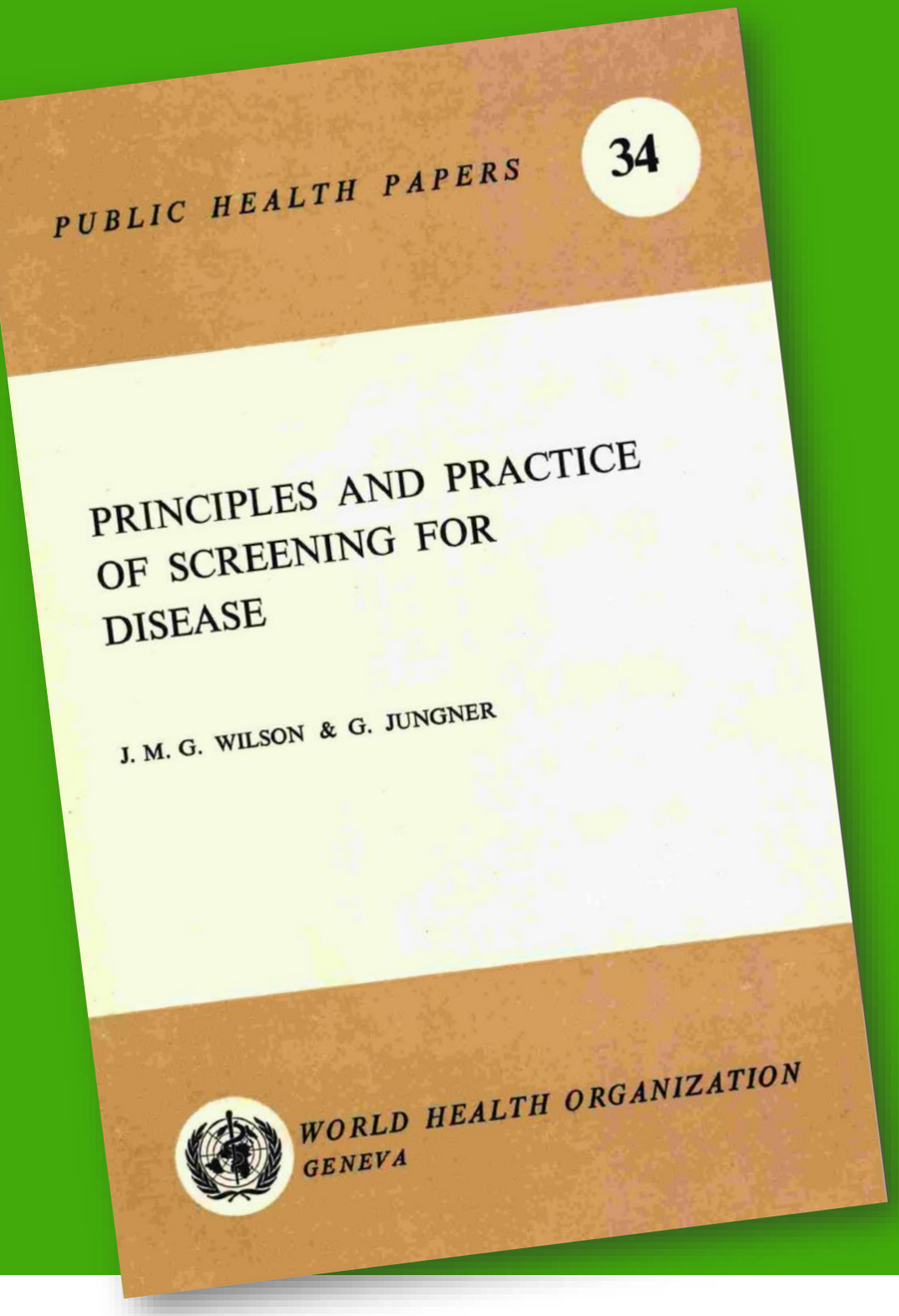


# 50 years of evolution of screening principles

RESEARCH ■ HEALTH SERVICES

## Consolidated principles for screening based on a systematic review and consensus process

Mark J. Dobrow PhD, Victoria Hagens MA, Roger Chafe PhD, Terrence Sullivan PhD, Linda Rabeneck MD MPH

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### ABSTRACT

**BACKGROUND:** In 1968, Wilson and Jungner published 10 principles of screening that often represent the de facto starting point for screening decisions today; 50 years on, are these principles still the right ones? Our objectives were to review published work that presents principles for population-based screening decisions since Wilson and Jungner's seminal publication, and to conduct a Delphi consensus process to assess the review results.

**METHODS:** We conducted a systematic review and modified Delphi consensus process. We searched multiple databases for articles published in English in 1968 or later that were intended to guide population-based screening decisions, described development and modification of principles, and presented principles as a set or list. Identified sets were compared for basic characteristics (e.g., number, categorization), a citation analysis was conducted, and principles were iteratively synthesized and consolidated into categories to assess evolution. Participants in the consensus process assessed the level of agreement with the importance and interpretability of the consolidated screening principles.

**RESULTS:** We identified 41 sets and 367 unique principles. Each unique principle was coded to 12 consolidated decision principles that were further categorized as disease/condition, test/intervention or program/system principles. Program or system issues were the focus of 3 of Wilson and Jungner's 10 principles, but comprised almost half of all unique principles identified in the review. The 12 consolidated principles were assessed through 2 rounds of the consensus process, leading to specific refinements to improve their relevance and interpretability. No gaps or missing principles were identified.

**INTERPRETATION:** Wilson and Jungner's principles are remarkably enduring, but increasingly reflect a truncated version of contemporary thinking on screening that does not fully capture subsequent focus on program or system principles. Ultimately, this review and consensus process provides a comprehensive and iterative modernization of guidance to inform population-based screening decisions.

In 1968, Wilson and Jungner published *Principles and Practice of Screening for Disease*,[1] a seminal work that highlighted 10 principles that should be considered when making a screening decision (Box 1). These screening principles were set out as normative statements regarding what should be known about the relative importance of a health problem, the natural progression of the disease or condition, the characteristics of available screening tests and follow-up treatments, and the cost-effectiveness of screening, before proceeding with a screening decision. Health care professionals, screening experts and policy-makers from all parts of the world use these principles to guide screening decisions. But despite the popularity of these principles, screening decisions remain challenging.[2,3] Recent controversies regarding screening for cancer[4–6] and screening in newborns[7] highlight the persistent complexity of screening decisions and the intense scrutiny under which they are made. The Wilson and Jungner principles of screening often represent the de facto starting point for these inherently contentious and costly sets of decisions. But after almost 50 years, are these principles still the right ones? Since their original publication, there has not been a systematic attempt to examine how screening principles have evolved or an assessment of what constitutes a comprehensive set of screening principles to guide contemporary screening decisions.

The objectives of this study were to review published work that presents principles for guiding population-based screening decisions since the publication of Wilson and Jungner's principles in 1968, and to conduct a Delphi consensus process to assess a synthesis of the review results.

### Methods

We employed a systematic review to identify, synthesize and consolidate existing principles of screening, followed by a modified Delphi consensus process with international screening





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# Systematic Review and Consensus Process

# Systematic Review

- **41** sets of screening principles

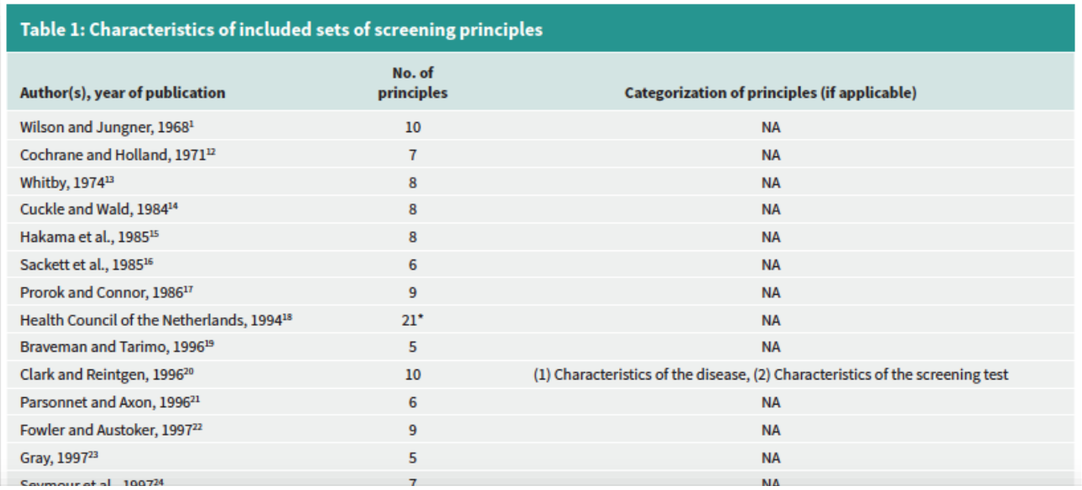

**Table 1: Characteristics of included sets of screening principles**

| Author(s), year of publication | No. of principles | Categorization of principles (if applicable) |
|---|---|---|
| Wilson and Jungner, 1968[1] | 10 | NA |
| Cochrane and Holland, 1971[12] | 7 | NA |
| Whitby, 1974[13] | 8 | NA |
| Cuckle and Wald, 1984[14] | 8 | NA |
| Hakama et al., 1985[15] | 8 | NA |
| Sackett et al., 1985[16] | 6 | NA |
| Prorok and Connor, 1986[17] | 9 | NA |
| Health Council of the Netherlands, 1994[18] | 21* | NA |
| Braveman and Tarimo, 1996[19] | 5 | NA |
| Clark and Reintgen, 1996[20] | 10 | (1) Characteristics of the disease, (2) Characteristics of the screening test |
| Parsonnet and Axon, 1996[21] | 6 | NA |
| Fowler and Austoker, 1997[22] | 9 | NA |
| Gray, 1997[23] | 5 | NA |
| Seymour et al., 1997[24] | 7 | NA |

**Table 1: Characteristics of included sets of screening principles**

| Author(s), year of publication | No. of principles | Categorization of principles (if applicable) |
|---|---|---|
| Wilson and Jungner, 1968[1] | 10 | NA |
| Cochrane and Holland, 1971[12] | 7 | NA |
| Whitby, 1974[13] | 8 | NA |
| Cuckle and Wald, 1984[14] | 8 | NA |
| Hakama et al., 1985[15] | 8 | NA |
| Sackett et al., 1985[16] | 6 | NA |
| Prorok and Connor, 1986[17] | 9 | NA |
| Health Council of the Netherlands, 1994[18] | 21* | NA |
| Braveman and Tarimo, 1996[19] | 5 | NA |
| Clark and Reintgen, 1996[20] | 10 | (1) Characteristics of the disease, (2) Characteristics of the screening test |

# Appendix 3. Citation Analysis of Reviewed Sets of Screening Principles

| Citing Source \ Cited Source | Wilson and Jungner | Cochrane and Holland | Whitby | Cuckle and Wald | Hakama et al | Sackett et al | Prorok and Connor | Health Council of the Netherlands | Parsonnet and Axon | Braveman and Tarimo | Clark and Reintgen | Gray | Fowler and Austoker | Seymour et al | Schoemaker | UK National Screening Committee | Nielsen and Lang | Prorok et al | AAP Newborn Screening Taskforce | UK National Screening Committee | Katz | Hanselaar | Canadian National Committee | N Zealand National Health Committee | UK National Screening Committee | Harris and Kinsinger | Strong et al | Miller | Andermann et al | Bryant and Hamdy | Mandel and Smith | Lewis et al | Grootendorst et al | Andermann et al | Harris et al | Martin-Moreno et al | Petros | Adriaensen et al | Forman et al | Lewis et al | UK National Screening Committee |
|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|
| Wilson and Jungner (1968) | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | |
| Cochrane and Holland (1971) | C | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | |
| Whitby (1974) | C | C | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | |
| Cuckle and Wald (1984) | C | C | - | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | |
| Hakama et al (1985) | - | - | - | - | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | |
| Sackett et al (1985) | - | - | - | - | - | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | |
| Prorok and Connor (1986) | C | - | | - | C | - | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | |
| Health Council of the Netherlands (1994) | C | - | | - | - | - | - | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | |
| Parsonnet and Axon (1996) | - | - | | - | - | - | - | - | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | |
| Braveman and Tarimo (1996) | C | - | | - | - | - | - | - | - | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | |
| Clark and Reintgen (1996) | - | - | | - | - | - | - | - | - | - | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | |
| Gray (1997) | C | - | | - | - | - | - | - | - | - | - | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | |
| Fowler and Austoker (1997) | C | C | | C | - | - | - | - | - | - | - | - | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | |
| Seymour et al (1997) | C | - | | - | - | - | - | - | - | - | - | - | - | | | | | | | | | | | | | | | | | | | | | | | | | | | | | |
| Schoemaker (1998) | C | C | | - | - | - | - | - | - | - | - | - | - | - | | | | | | | | | | | | | | | | | | | | | | | | | | | | |
| UK National Screening Committee (1998) | C | C | | - | - | - | - | - | - | - | - | A | - | - | - | | | | | | | | | | | | | | | | | | | | | | | | | | | |
| Nielsen and Lang (1999) | - | - | | - | - | - | - | - | - | - | - | - | - | - | - | - | | | | | | | | | | | | | | | | | | | | | | | | | | |
| Prorok et al (1999) | C | - | | - | A | - | A | - | - | - | - | - | - | - | - | - | - | | | | | | | | | | | | | | | | | | | | | | | | | |
| AAP Newborn Screening Taskforce (2000) | - | - | | - | - | - | - | - | - | - | - | - | - | C | - | - | - | - | | | | | | | | | | | | | | | | | | | | | | | |
| UK National Screening Committee (2000) | C | C | | - | - | - | - | - | - | - | - | A | - | - | - | C | - | - | - | | | | | | | | | | | | | | | | | | | | | | | |
| Katz (2001) | - | - | | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | | | | | | | | | | | | | | | | | | | | | | |
| Hanselaar (2002) | C | - | | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | | | | | | | | | | | | | | | | | | | | | |
| Canadian National Committee (2002) | C | - | | - | - | - | - | - | C | - | - | C | - | - | - | - | - | - | - | - | - | - | | | | | | | | | | | | | | | | | | | |
| New Zealand National Health Committee (2003) | C | C | | C | - | - | - | - | - | - | - | C | C | - | - | - | - | - | - | C | - | - | - | | | | | | | | | | | | | | | | | | |
| UK National Screening Committee (2003) | C | C | | - | - | - | - | - | - | - | - | A | - | - | - | A | - | - | - | A | - | - | - | - | | | | | | | | | | | | | | | | | |
| Harris and Kinsinger (2005) | - | - | | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | | | | | | | | | | | | | | | | |
| Strong et al (2005) | C | - | | - | C | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | | | | | | | | | | | | | | | |
| Miller (2005) | C | - | | C | C | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | A | | | | | | | | | | | | | | |
| Andermann et al (2008) | C | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | C | - | - | - | - | - | C | - | - | | | | | | | | | | | | | |
| Bryant and Hamdy (2008) | C | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | | - | | | | | | | | | | | |
| Mandel and Smith (2008) | C | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | | - | | | | | | | | | | | |
| Lewis et al (2008) | C | - | - | - | - | - | - | - | - | - | - | - | - | - | - | C | - | - | - | C | - | - | - | - | C | - | - | | - | - | | | | | | | | | | |
| Grootendorst et al (2009) | C | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | | C | - | - | | | | | | | | | |
| Andermann et al (2009) | C | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | C | - | - | - | - | - | C | - | - | | C | - | - | - | | | | | | | | |
| Harris et al (2011) | C | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | A | - | | C | - | - | - | - | | | | | | | |
| Martin-Moreno et al (2012) | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | | - | - | - | - | - | - | | | | | | |
| Petros (2012) | C | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | | C | - | - | - | - | - | - | | | | | |
| Adriaensen et al (2013) | C | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | | - | - | - | - | - | C | - | - | | | | |
| Forman et al (2013) | C | C | - | - | - | - | - | - | - | - | - | - | - | C | - | - | - | - | - | - | - | - | - | - | - | - | - | | C | - | - | - | - | - | - | - | - | | | |
| Lewis et al (2014) | C | - | - | - | - | - | - | - | - | - | - | - | - | - | - | C | - | - | - | C | - | - | - | - | C | - | - | | - | - | - | A | - | - | - | - | - | - | - | |
| UK National Screening Committee (2015) | C | C | - | C | - | - | - | - | - | - | - | C | - | - | - | A | - | - | - | A | - | - | - | - | A | - | - | | - | - | - | - | - | - | - | - | - | - | - | - |

Legend: The article in the 'Cited Source' column was cited by all of the articles denoted by 'C' in the 'Citing Source' rows.
The article in the 'Cited Source' column, denoted by the 'A', was not cited by the article in the 'Citing Source' row, however, one or more authors contributed to both citing/cited sources.

# Appendix 3. Citation Analysis of Reviewed Sets of Screening Principles – Updated for Dobrow et al 2018

| Citing Source \ Cited Source | Wilson and Jungner | Cochrane and Holland | Whitby | Cuckle and Wald | Hakama et al | Sackett et al | Prorok and Connor | Health Council of the Netherlands | Parsonnet and Axon | Braveman and Tarimo | Clark and Reintgen | Gray | Fowler and Austoker | Seymour et al | Schoemaker | UK National Screening Committee | Nielsen and Lang | Prorok et al | AAP Newborn Screening Taskforce | UK National Screening Committee | Katz | Hanselaar | Canadian National Committee | N Zealand National Health Committee | UK National Screening Committee | Harris and Kinsinger | Strong et al | Miller | Andermann et al | Bryant and Hamdy | Mandel and Smith | Lewis et al | Grootendorst et al | Andermann et al | Harris et al | Martin-Moreno et al | Petros | Adriaensen et al | Forman et al | Lewis et al | UK National Screening Committee | Dobrow et al |
|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|
| Wilson and Jungner (1968) | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | |
| Cochrane and Holland (1971) | C | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | |
| Whitby (1974) | C | C | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | |
| Cuckle and Wald (1984) | C | C | - | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | |
| Hakama et al (1985) | - | - | - | - | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | |
| Sackett et al (1985) | - | - | - | - | - | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | |
| Prorok and Connor (1986) | C | - | - | - | C | - | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | |
| Health Council of the Netherlands (1994) | C | - | - | - | - | - | - | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | |
| Parsonnet and Axon (1996) | - | - | - | - | - | - | - | - | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | |
| Braveman and Tarimo (1996) | C | - | - | - | - | - | - | - | - | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | |
| Clark and Reintgen (1996) | - | - | - | - | - | - | - | - | - | - | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | |
| Gray (1997) | C | - | - | - | - | - | - | - | - | - | - | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | |
| Fowler and Austoker (1997) | C | C | - | C | - | - | - | - | - | - | - | - | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | |
| Seymour et al (1997) | C | - | - | - | - | - | - | - | - | - | - | - | - | | | | | | | | | | | | | | | | | | | | | | | | | | | | | |
| Schoemaker (1998) | C | C | - | - | - | - | - | - | - | - | - | - | - | - | | | | | | | | | | | | | | | | | | | | | | | | | | | | |
| UK National Screening Committee (1998) | C | C | - | - | - | - | - | - | - | - | - | A | - | - | - | | | | | | | | | | | | | | | | | | | | | | | | | | | |
| Nielsen and Lang (1999) | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | | | | | | | | | | | | | | | | | | | | | | | | | | |
| Prorok et al (1999) | C | - | - | - | A | - | A | - | - | - | - | - | - | - | - | - | - | | | | | | | | | | | | | | | | | | | | | | | | | |
| AAP Newborn Screening Taskforce (2000) | - | - | - | - | - | - | - | - | - | - | - | - | - | C | - | - | - | - | | | | | | | | | | | | | | | | | | | | | | | |
| UK National Screening Committee (2000) | C | C | - | - | - | - | - | - | - | - | - | A | - | - | - | C | - | - | - | | | | | | | | | | | | | | | | | | | | | | | |
| Katz (2001) | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | | | | | | | | | | | | | | | | | | | | | | |
| Hanselaar (2002) | C | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | | | | | | | | | | | | | | | | | | | | | |
| Canadian National Committee (2002) | C | - | - | - | - | - | - | C | - | - | C | - | - | - | - | - | - | - | - | - | - | - | | | | | | | | | | | | | | | | | | | | |
| New Zealand National Health Committee (2003) | C | C | - | C | - | - | - | - | - | - | C | C | - | - | - | - | - | - | - | C | - | - | - | | | | | | | | | | | | | | | | | | | |
| UK National Screening Committee (2003) | C | C | - | - | - | - | - | - | - | - | A | - | - | - | A | - | - | - | - | A | - | - | - | - | | | | | | | | | | | | | | | | | | |
| Harris and Kinsinger (2005) | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | | | | | | | | | | | | | | | | | |
| Strong et al (2005) | C | - | - | - | C | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | | | | | | | | | | | | | | | | |
| Miller (2005) | C | - | - | C | C | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | A | | | | | | | | | | | | | | | |
| Andermann et al (2008) | C | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | C | - | - | - | - | - | C | - | - | - | | | | | | | | | | | | | | |
| Bryant and Hamdy (2008) | C | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | | | | | | | | | | | | | |
| Mandel and Smith (2008) | C | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | | | | | | | | | | | | |
| Lewis et al (2008) | C | - | - | - | - | - | - | - | - | - | - | - | - | - | - | C | - | - | - | C | - | - | - | - | C | - | - | - | - | - | - | | | | | | | | | | |
| Grootendorst et al (2009) | C | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | C | - | - | - | | | | | | | | | |
| Andermann et al (2009) | C | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | C | - | - | - | - | - | C | - | - | - | C | - | - | - | - | | | | | | | | |
| Harris et al (2011) | C | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | A | - | - | C | - | - | - | - | - | | | | | | | |
| Martin-Moreno et al (2012) | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | | | | | | |
| Petros (2012) | C | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | C | - | - | - | - | - | - | - | | | | | |
| Adriaensen et al (2013) | C | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | - | C | - | - | | | | |
| Forman et al (2013) | C | C | - | - | - | - | - | - | - | - | - | - | - | C | - | - | - | - | - | - | - | - | - | - | - | - | - | - | C | - | - | - | - | - | - | - | - | - | | | |
| Lewis et al (2014) | C | | | | | | | | | | | | | | | C | | | | C | | | | | C | | | | | | A | - | - | - | - | - | - | - | | | |
| UK National Screening Committee (2015) | C | C | - | C | - | - | - | - | - | - | - | C | - | - | - | A | - | - | - | A | - | - | - | - | A | - | - | - | - | - | - | - | - | - | - | - | - | - | - | |
| Dobrow et al (2018) | C | C | C | C | C | C | C | C | C | C | C | C | C | C | C | C | C | C | C | C | C | C | C | C | C | C | C | C | C | C | C | C | C | C | C | C | C | C | C | C | C |

# Consolidated Principles

## Appendix 4 (as supplied by the authors). Mapping of Individual Screening Principles to Consolidated Screening Principles

| CONSOLIDATED SCREENING PRINCIPLES (Post-Review and Consensus Process) | INDIVIDUAL SCREENING PRINCIPLES (from 41 reviewed sets of screening principles)<br>(*individual principle applies to more than 1 consolidated screening principle; original Wilson and Jungner principles in bold font) |
|---|---|
| | **I. DISEASE/CONDITION PRINCIPLES (26 unique principles)** |
| **1. Epidemiology of the disease/ condition**<br><br>The epidemiology of the disease/ condition should be adequately understood, and the disease/ condition should be an important health problem (e.g., high or increasing incidence/prevalence and/or causes substantial morbidity/mortality).<br><br>This consolidated decision principle includes 31 unique principles representing 32 distinct sources. | - **The condition sought should be an important health problem**[1-3]<br>- A genetic screening programme must relate to a health problem or to a condition which can lead to such a problem in those being tested or in their descendants[4]<br>- *Define clearly the adverse health outcome the program is intended to reduce. Define clearly the population that the program intends to screen[5]<br>- Disease is serious[6]<br>- Disease: high morbidity, mortality, and cost[7]<br>- Disease: high prevalence and incidence[7]<br>- *Disease: the disease should cause a sufficient burden of suffering to warrant attention and should have a detectable preclinical phase of sufficient length to allow early detection[8]<br>- Disorder associated with significant morbidity or mortality[9]<br>- Does the burden of the disability from the target disease warrant action?[10]<br>- Important health problem[11]<br>- Important health problem (i.e. common and serious)[12,13]<br>- Important public health concern[14]<br>- Is the condition to be detected of public importance?[15]<br>- Known incidence in populations relevant to UK[9]<br>- Prevalence: known[16]<br>- Screening protocols should be directed toward diseases with a relatively high incidence[17]<br>- The condition is an important health problem[18]<br>- The condition should be an important health problem[19-23]<br>- *The condition should be an important health problem as judged by its frequency and/or severity. The epidemiology, incidence, prevalence and natural history of the condition should be understood, including development from latent to declared disease and/or there should be robust evidence about the association between the risk or disease marker and serious or treatable disease[24]<br>- The condition sought should be a common and/or serious health problem[25]<br>- The condition to be screened for should have a high death or disability rate[17]<br>- The criteria for inclusion of a screening test are: a) the condition is an important health problem that occurs frequently enough to justify screening an entire population[26]<br>- The disease must be neither too rare, nor too common[6]<br>- The disease or condition should be an important problem (morbidity and mortality)[27]<br>- The disease or condition should be common (prevalence and incidence)[27]<br>- The disease should be a serious health problem and the cause of substantial mortality and morbidity[28]<br>- The disease should be a serious health problem, being common in occurrence and the cause of substantial mortality and morbidity[29]<br>- The disease should be an important health problem[30]<br>- The disease should be an important public health problem in terms of its frequency and/or severity. Historically, the development of this principle was in the general context of screening for infectious and chronic diseases and not related specifically to cancer. Today some of the cancer sites |

# 12 Consolidated Principles

| Domains | Consolidated Principles |
|---|---|
| **Disease / Condition** | 1. Epidemiology of the disease or condition |
| | 2. Natural history of disease or condition |
| | 3. Target population for screening |
| **Test / Intervention** | 4. Screening test performance characteristics |
| | 5. Interpretation of screening test results |
| | 6. Post screening test options |
| **Program / System** | 7. Screening program infrastructure |
| | 8. Screening program coordination and integration |
| | 9. Screening program acceptability and ethics |
| | 10. Screening program benefits and harms |
| | 11. Economic evaluation of screening program |
| | 12. Screening program quality and performance management |

# 12 Consolidated Principles

| Domains | Consolidated Principles |
| --- | --- |
| Disease / Condition | 1. Epidemiology of the disease or condition |
| Test / Intervention | |
| Program / System | 10. Screening program benefits and harms<br>11. Economic evaluation of screening program<br>12. Screening program quality and performance management |

**Program/System** principles account for:

- 6 of 12 (50%) consolidated principles
- 171 of 367 (47%) unique principles

# 12 Consolidated Principles

| Domains | Consolidated Principles – Overlap with Wilson/Jungner Principles |
|---|---|
| **Disease / Condition** | 1. Epidemiology of the disease or condition |
| | 2. Natural history of disease or condition |
| | 3. Target population for screening |
| **Test / Intervention** | 4. Screening test performance characteristics |
| | 5. Interpretation of screening test results |
| | 6. Post screening test options |
| **Program / System** | 7. Screening program infrastructure |
| | 8. Screening program coordination and integration |
| | 9. Screening program acceptability and ethics |
| | 10. Screening program benefits and harms |
| | 11. Economic evaluation of screening program |
| | 12. Screening program quality and performance management |

# 12 Consolidated Principles

| Domains | Consolidated Principles – Overlap with Wilson/Jungner Principles |
|---|---|
| Disease / Condition | 1. Epidemiology of the disease or condition |
| | 2. Natural history of disease or condition (2) |
| | 3. Target population for screening |
| Test / Intervention | 4. Screening test performance characteristics (2) |
| | 5. Interpretation of screening test results |
| | 6. Post screening test options |
| Program / System | 7. Screening program infrastructure |
| | 8. Screening program coordination and integration |
| | 9. Screening program acceptability and ethics |
| | 10. Screening program benefits and harms |
| | 11. Economic evaluation of screening program |
| | 12. Screening program quality and performance management |

*Table A2.1 Comparison of Round 1 and Round 2 Versions of the Consolidated Principles*

| Round 1 Version of Consolidated Principles (Post-Systematic Review) | Round 2 Version of Consolidated Principles (Includes Post-Round 1 Refinements) |
|---|---|
| **Principle 1.** Epidemiology of the disease/condition: The epidemiology of the disease/condition should be adequately understood, and the disease/ condition should be an important health problem (e.g., high or increasing incidence/prevalence and causes substantial morbidity/mortality). | **Principle 1.** Epidemiology of the disease/condition: The epidemiology of the disease/condition should be adequately understood, and the disease/condition should be an important health problem (e.g., high or increasing incidence/prevalence and/or causes substantial morbidity/mortality). |
| **Principle 2.** Natural history of disease/condition: The natural history of the disease/condition should be adequately understood, the disease/condition is well-defined, and there should be a detectable preclinical phase. | **Principle 2.** Natural history of disease/condition: The natural history of the disease/condition should be adequately understood, the disease/condition is well-defined and, where relevant, there should be a detectable preclinical phase. |
| **Principle 3.** Target population for screening: The target population for screening should be clearly defined (e.g., with an appropriate target age-range), identifiable, accessible, and likely to participate. | **Principle 3.** Target population for screening: The target population for screening should be clearly defined (e.g., with an appropriate target age-range), identifiable, and contactable. |
| **Principle 4.** Screening test performance characteristics Screening test performance should be appropriate for the purpose, with all key components of the test being accurate (e.g., sensitive, specific, positive predictive value), reliable/reproducible, safe/ethical/acceptable, simple and cost-effective to perform/ administer to the target population. | **Principle 4.** Screening test performance characteristics: Screening test performance should be appropriate for the purpose, with all key components specific to the test (rather than the screening program) being accurate (e.g., in terms of sensitivity, specificity, positive predictive value) and reliable/reproducible. The test should be acceptable to the target population and it should be possible to perform/administer it safely, affordably and efficiently. |
| **Principle 5.** Target population for post-screening care: Screening test results should be clearly interpretable and determinate (e.g., with known distribution of test values and well-defined and agreed cut-off points) to allow identification of the screening participants who should (and should not) be offered diagnostic testing and other post-screening care. | **Principle 5.** Interpretation of screening test results: Screening test results should be clearly interpretable and, where appropiate, determinate (e.g., with known distribution of test values and well-defined and agreed cut-off points) to allow identification of the screening participants who should (and should not) be offered diagnostic testing and other post-screening care. |
| **Principle 6.** Post-screening care: There should be an agreed upon course of action for screening participants with positive screening results that involves diagnostic testing, treatment/intervention and follow-up care that will modify/alter the natural history and clinical pathway for the disease/condition, is available/accessible/acceptable to those affected and results in improved outcomes (e.g., survival, function, quality of life). The burden of post-screening care on all participants should be understood and the impact of false-positive tests should be minimized. | **Principle 6.** Post-screening test options: There should be an agreed upon course of action for screening participants with positive screening test results that involves diagnostic testing, treatment/intervention and follow-up care that will modify/alter the natural history and clinical pathway for the disease/condition, is accessible and acceptable to those affected and results in improved outcomes (e.g., increased functioning/quality of life, decreased cause-specific mortality). The burden of post-screening care on all participants should be understood/acceptable and the impact of false-positive and false-negative tests should be minimal. |
| **Principle 7:** Screening program infrastructure: There should be adequate infrastructure (e.g., financial resources, health human resources, information technology, facilities, equipment, test | **Principle 7.** Screening program infrastructure: There should be adequate existing infrastructure (e.g., financial resources, health human resources, information technology, facilities, equipment, |

**Table 2: Final refined set of consolidated screening principles**

| Domain | Consolidated screening principles (after systematic review and modified Delphi consensus process) |
|---|---|
| Disease/condition principles | **1. Epidemiology of the disease or condition**<br>The epidemiology of the disease or condition should be adequately understood, and the disease or condition should be an important health problem (e.g., high or increasing incidence or prevalence, or causes substantial morbidity or mortality). |
| | **2. Natural history of disease or condition**<br>The natural history of the disease or condition should be adequately understood, the disease or condition is well-defined, and there should be a detectable preclinical phase. |
| | **3. Target population for screening**<br>The target population for screening should be clearly defined (e.g., with an appropriate target age range), identifiable and able to be reached. |
| Test/intervention principles | **4. Screening test performance characteristics**<br>Screening test performance should be appropriate for the purpose, with all key components specific to the test (rather than the screening program) being accurate (e.g., in terms of sensitivity, specificity and positive predictive value) and reliable or reproducible. The test should be acceptable to the target population and it should be possible to perform or administer it safely, affordably and efficiently. |
| | **5. Interpretation of screening test results**<br>Screening test results should be clearly interpretable and determinate (e.g., with known distribution of test values and well-defined and agreed cut-off points) to allow identification of the screening participants who should (and should not) be offered diagnostic testing and other postscreening care. |
| | **6. Postscreening test options**<br>There should be an agreed on course of action for screening participants with positive screening test results that involves diagnostic testing, treatment or intervention, and follow-up care that will modify the natural history and clinical pathway for the disease or condition; that is available, accessible and acceptable to those affected; and that results in improved outcomes (e.g., increased functioning or quality of life, decreased cause-specific mortality). The burden of testing on all participants should be understood and acceptable, and the effect of false-positive and false-negative tests should be minimal. |
| Program/system principles | **7. Screening program infrastructure**<br>There should be adequate existing infrastructure (e.g., financial resources, health human resources, information technology, facilities, equipment and test technology), or a clear plan to develop adequate infrastructure, that is appropriate to the setting to allow for timely access to all components of the screening program.* |
| | **8. Screening program coordination and integration**<br>All components of the screening program* should be coordinated and, where possible, integrated with the broader health care system (including a formal system to inform, counsel, refer and manage the treatment of screening participants) to optimize care continuity and ensure no screening participant is neglected. |
| | **9. Screening program acceptability and ethics**<br>All components of the screening program* should be clinically, socially and ethically acceptable to screening participants, health professionals and society, and there should be effective methods for providing screening participants with informed choice, promoting their autonomy and protecting their rights. |
| | **10. Screening program benefits and harms**<br>The expected range and magnitude of benefits (e.g., increased functioning or quality of life, decreased cause-specific mortality) and harms (e.g., overdiagnosis and overtreatment) for screening participants and society should be clearly defined and acceptable, and supported by existing high-quality scientific evidence (or addressed by ongoing studies) that indicates that the overall benefit of the screening program outweighs its potential harms. |
| | **11. Economic evaluation of screening program**<br>An economic evaluation (e.g., cost-effectiveness analysis, cost–benefit analysis and cost–utility analysis) of the screening program, using a health system or societal perspective, should be conducted (or a clear plan to conduct an economic evaluation) to assess the full costs and effects of implementing, operating and sustaining the screening program while clearly considering the opportunity costs and effect of allocating resources to other potential nonscreening alternatives (e.g., primary prevention, improved treatments and other clinical services) for managing the disease or condition. |
| | **12. Screening program quality and performance management**<br>The screening program should have clear goals or objectives that are explicitly linked to program planning, monitoring, evaluating and reporting activities, with dedicated information systems and funding, to ensure ongoing quality control and achievement of performance targets. |

*Components of a screening program include recruitment, testing, information access, diagnosis, referral, treatment, follow-up, patient education and support, staff training and program management and evaluation.

| | |
|---|---|
| Disease/condition principles | **1. Epidemiology of the disease or condition**<br>The epidemiology of the disease or condition should be adequately understood, and the disease or condition should be an important health problem (e.g., high or increasing incidence or prevalence, or causes substantial morbidity or mortality). |
| | **2. Natural history of disease or condition**<br>The natural history of the disease or condition should be adequately understood, the disease or condition is well-defined, and there should be a detectable preclinical phase. |
| Not included in Wilson/Jungner principles of screening | **3. Target population for screening**<br>The target population for screening should be clearly defined (e.g., with an appropriate target age range), identifiable and able to be reached. |
| Test/intervention principles | **4. Screening test performance characteristics**<br>Screening test performance should be appropriate for the purpose, with all key components specific to the test (rather than the screening program) being accurate (e.g., in terms of sensitivity, specificity and positive predictive value) and reliable or reproducible. The test should be acceptable to the target population and it should be possible to perform or administer it safely, affordably and efficiently. |
| | **5. Interpretation of screening test results**<br>Screening test results should be clearly interpretable and determinate (e.g., with known distribution of test values and well-defined and agreed cut-off points) to allow identification of the screening participants who should (and should not) be offered diagnostic testing and other postscreening care. |
| | **6. Postscreening test options**<br>There should be an agreed on course of action for screening participants with positive screening test results that involves diagnostic testing, treatment or intervention, and follow-up care that will modify the natural history and clinical pathway for the disease or condition; that is available, accessible and acceptable to those affected; and that results in improved outcomes (e.g., increased functioning or quality of life, decreased cause-specific mortality). The burden of testing on all participants should be understood and acceptable, and the effect of false-positive and false-negative tests should be minimal. |

| | |
|---|---|
| Program/system principles | **7. Screening program infrastructure**<br>There should be adequate existing infrastructure (e.g., financial resources, health human resources, information technology, facilities, equipment and test technology), or a clear plan to develop adequate infrastructure, that is appropriate to the setting to allow for timely access to all components of the screening program.* |
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| | **11. Economic evaluation of screening program**<br>An economic evaluation (e.g., cost-effectiveness analysis, cost–benefit analysis and cost–utility analysis) of the screening program, using a health system or societal perspective, should be conducted (or a clear plan to conduct an economic evaluation) to assess the full costs and effects of implementing, operating and sustaining the screening program while clearly considering the opportunity costs and effect of allocating resources to other potential nonscreening alternatives (e.g., primary prevention, improved treatments and other clinical services) for managing the disease or condition. |
| Not included in Wilson/Jungner principles of screening | **12. Screening program quality and performance management**<br>The screening program should have clear goals or objectives that are explicitly linked to program planning, monitoring, evaluating and reporting activities, with dedicated information systems and funding, to ensure ongoing quality control and achievement of performance targets. |

*Components of a screening program include recruitment, testing, information access, diagnosis, referral, treatment, follow-up, patient education and support, staff training and program management and evaluation.

## Box 1: Wilson and Jungner's principles of screening[1]

- The condition sought should be an important health problem.
- The natural history of the condition, including development from latent to declared disease, should be adequately understood.
- There should be a recognizable latent or early symptomatic stage.
- There should be a suitable test or examination.
- The test should be acceptable to the population.
- There should be an agreed policy on whom to treat as patients.
- There should be an accepted treatment for patients with recognized disease.
- Facilities for diagnosis and treatment should be available.
- The cost of case-finding (including diagnosis and treatment of patients diagnosed) should be economically balanced in relation to possible expenditure on medical care as a whole.
- Case-finding should be a continuing process and not a "once and for all" project.

# Implications/Considerations

# Shifting *Principles*

- Are Wilson and Jungner's 10 principles showing their age?
  - 50 years of evolution of screening principles has led to shift toward more operational and implementation issues
  - The Wilson/Jungner principles do not fully capture the extended focus of subsequent work toward program/system considerations
  - While the Wilson/Jungner principles were ahead of their time, they tend to reflect a truncated version of contemporary thinking on screening
  - Our 12 consolidated screening principles build on 50 years of evolution, but principles are not static

# Shifting *Evidence*

- With shifting principles, evidence needs also shift
  - Differing characteristics of the evidence base by domain
    - For disease/condition and test/intervention principles: evidence base is typically high-quality experimental or observational studies
    - For program/system principles: evidence base is much less developed and more context-dependent
  - Broader/more sophisticated conception of evidence needed
    - Research, contextual, experiential evidence – the '*necessary but not sufficient*' caveat
    - Global and local evidence – rigour needed for both

# Shifting *Decision Context*

- With shifting principles and shifting evidence, the decision context also shifts
  - Nature of programmatic screening decisions
    - Highly complex and scrutinized
    - Not single yes-no decisions, but rather multiple linked decisions
    - Process often runs over multiple years
  - Expertise required to make screening decisions
    - Involve multiple experts/stakeholders to generate, identify, interpret and apply a broader and more diverse evidence base
    - Evidence for disease/condition and test/intervention principles typically assessed by clinical and epidemiologic experts
    - Evidence for program/system principles requires a more diverse set of experts and stakeholders (e.g., health service program managers, policy analysts, information system specialists, health economists, ethicists and members of both average and high-risk population groups)

# The future of screening criteria for informing new screening programmes

- Acknowledge shifting principles, shifting evidence, shifting decision contexts

- Strive for ***good questions before good answers***
  - Clear, rational logic (i.e., principles) should drive decision-making, not emergent evidence
  - Address new challenges (e.g., screening of high-risk populations) under lens of screening principles

- Clarify **screening governance**
  - Clarify who has overarching responsibility for screening decisions
  - Clarify appropriate set of screening principles that will guide decision-making
  - Clarify which experts/stakeholders should contribute to specific components of screening decisions
  - Clarify evidence sources/development sought (e.g., research/contextual/experiential and global/local)
  - Clarify responsibility for combining multiple evidence-informed inputs together
  - Clarify responsibility for monitoring screening decisions on an ongoing basis

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# Questions? Comments?

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