

**Please cite as:**

Little J., Nicholls SG, Clifford T, Sikora L, Atwere P, Ashcroft R, Newson A, Potter B, Brehaut J, Graham I, Wilson M, Lavis J, Grimshaw J, Verweij M, Dawson A, Coyle D, Kemper A, Botkin J, Dunfield L., A critical interpretive synthesis of evidence and values in recommendations to disinvest from population Screening. Conference paper, July 2016, 11th Annual Meeting of the Society for Research Synthesis Methodology, Florence, Italy

See discussions, stats, and author profiles for this publication at: <https://www.researchgate.net/publication/305308929>

# A critical interpretive synthesis of evidence and values in recommendations to disinvest from population Screening

Conference Paper · July 2016

---

READS

91

18 authors, including:



**Tammy Clifford**

Canadian Agency For Drugs And Technologies In Health

63 PUBLICATIONS 740 CITATIONS

SEE PROFILE



**Jeremy Grimshaw**

Ottawa Hospital Research Institute

537 PUBLICATIONS 38,919 CITATIONS

SEE PROFILE

# DIMPLES:

*A critical interpretive synthesis of evidence and values in recommendations to disinvest from population Screening*

Presented by: Julian Little, July 11<sup>th</sup>, 2016

# Outline

- the interplay between values and evidence in screening policy
- methods of an ongoing systematic review
- examples of disinvestment decisions

# Population screening

- Screening is:
  - “Testing people who either do not or have not recognized the signs or symptoms of the condition being tested for. In other words, they believe themselves to be well in relation to the disease that the screening relates to”
  - “Where the stated or implied purpose is to reduce risk for that individual of future ill health in relation to the condition being tested for, or to give information about risk that is deemed valuable for that individual even though risk cannot be altered”
  - “It encompasses the whole system or programme of events necessary to achieve risk reduction. Screening is a programme not a test.” (Raffle & Gray, 2007; p37)
- Examples include: Newborn bloodspot screening, prostate cancer screening, cervical cancer screening, breast cancer screening

# Taking prostate screening as an example

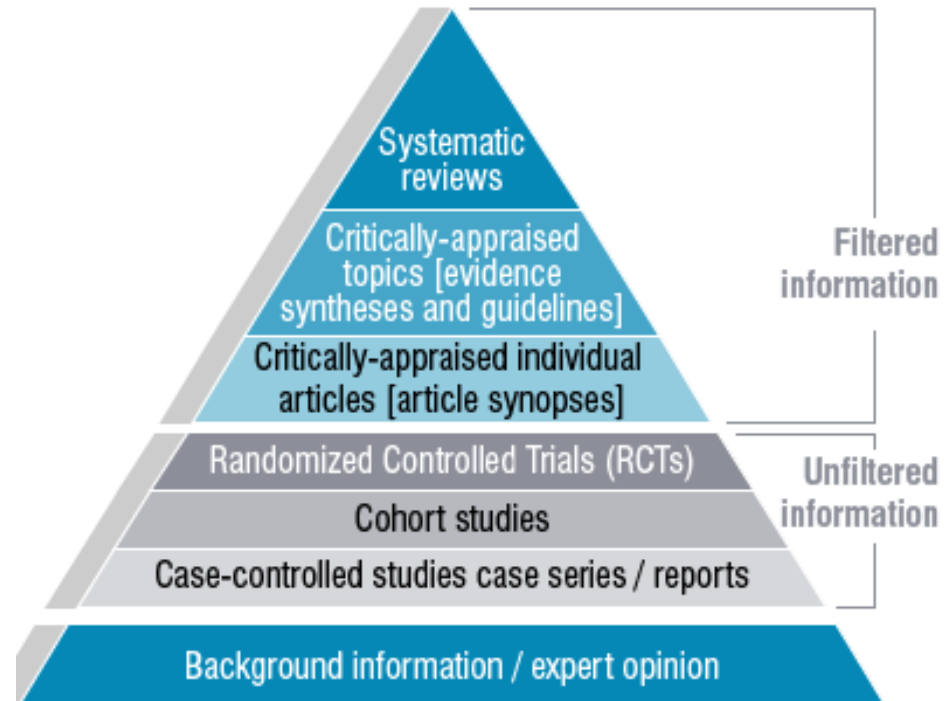
Professional Organization	Year of latest Recommendation
American Cancer Society	2012
American College of Physicians	2013
Canadian Task force on Preventive Health Care	2014
European Society for Medical Oncology	2013
Prostate Cancer Canada	2013
United States Preventive Services Task Force (USPSTF)	2012

# When the evidence recommendations disagree

- Canadian Agency for Drugs Technology and Health (CADTH) review (Prostate Cancer Screening, 2013):

“Almost all the included guidelines reported that *their recommendations were based on a balance between the benefits and harms* of screening; however, *the specific outcomes reviewed in each guideline and the weights given for each outcome varied from one guideline to another.*”

- Clinical Practice Guidelines (CPG) issued from the Royal Australasian College of General Practitioners (7<sup>th</sup> edn. 2009) stated that men aged 55–69 years **should not** be offered PSA testing routinely whereas CPG from the Urological Society of Australia and New Zealand stated *they should* (2009)(Scott & Guyatt, 2013).



# Evidence review process: Variation

**Table 3: Comparison of criteria across general and genetic screening**

No	Criteria	W&J	Aus <sup>52</sup>	Can <sup>44a</sup>	Den <sup>45</sup>	Fin <sup>38</sup>	Fra <sup>35</sup>	Ger <sup>41</sup>	Ita <sup>36</sup>	Neth <sup>33</sup>	NZ <sup>34</sup>	Spa <sup>42</sup>	Swe <sup>55</sup>	UK <sup>4</sup>	USA <sup>35a</sup>	WHO <sup>37</sup>	EU <sup>18</sup>	And 08 <sup>6</sup>	And 09/10 <sup>8,9</sup>	Blanc <sup>7</sup>	Fowler <sup>34</sup>	Gray <sup>34</sup>	Harris <sup>54</sup>	Holland <sup>47</sup>	HCN <sup>b</sup>	Goel <sup>b</sup>	ESHG <sup>b</sup>	EC ELSI <sup>b</sup>	PHGF <sup>b</sup>	Bonham <sup>b</sup>	
1	Suitable or well-defined candidate for screening			X	X						X				X	X		X				X									
2	<b>The condition should be an important health problem</b> "The overall burden of disease due to genetic conditions should merit a public health response that examines the possibility of prevention. EURORDIS advocates that this should be a global approach- rather than a piecemeal policy for each disorder separately." (PHGF 2010)	X	X		X				X	X		X	X	X	X	X	X	X	X					X		X	X	X	X	X	
	a. Burden of condition – Incidence & prevalence			X		X	X								X	X															
	b. Mortality and morbidity						X										X														
	c. Socioeconomic impact						X										X														

Seedat F, Cooper J, Cameron L, et al. *International comparisons of screening policy-making: A systematic review*, 2014. URL:

[https://www.gov.uk/government/uploads/system/uploads/attachment\\_data/file/444227/FINAL\\_REPORT\\_International\\_Screening.pdf](https://www.gov.uk/government/uploads/system/uploads/attachment_data/file/444227/FINAL_REPORT_International_Screening.pdf)

## Stopping screening: controversy

### NICE condemn breast cancer women to death while fatties jump the queue for gastric bands

18:41, 9 August 2014

OPINION

BY CAROLEMALONE



★ Recommended In News

**Mirror**

‘Ludicrous’ advice against prostate cancer screening could cause needless deaths: study

**NATIONAL POST**



# Screening decisions, evidence-based medicine, and values

- CTFPHC recommendations on prostate screening (Oct 2014).
  - [the CTFPHC] “rated the quality of evidence and strength of recommendations using the Grading of Recommendations Assessment, Development and Evaluation (GRADE) system. However, **it paid insufficient attention to patient values, patient preferences and costs.**” (Krahn, 2014)
- Are agreements/disagreements about evidence, or values? (Atkins et al., 2005)
  - “Disease prevention can only be understood in a political as well as a medical context. **Medical knowledge provides a technological base for the analysis of policy options. However, it is the political process, reflecting the power and values of the several constituencies in society, that will determine which choices are made.**”(Eisenberg, 1984)

# Research objective(s)

- The **goal** of this review is to conduct a **critical interpretive synthesis** (CIS) to better understand the stated rationales for disinvestment from established population screening programs. In doing so we will:
  - Identify **examples of population screening programs** recommended for disinvestment and describe their characteristics
  - Generate a **matrix of factors associated with recommendations to disinvest** from existing screening programs and **compare these with established criteria for the implementation** of population screening
  - Elicit the **values which underpin disinvestment recommendations.**
  - Generate a **categorization framework of disinvestment decision-types**

# Research objective(s)

- The **goal** of this review is to conduct a **critical interpretive synthesis** (CIS) to better understand the stated rationales for disinvestment from established population screening programs. In doing so we will:
  - Identify **examples of population screening programs** recommended for disinvestment and describe their characteristics
  - Generate a **matrix of factors associated with recommendations to disinvest** from existing screening programs and **compare these with established criteria for the implementation** of population screening
  - Elicit the **values which underpin disinvestment recommendations**. (i.e. theory/concept development)
  - Generate a **categorization framework of disinvestment decision-types**

# Critical interpretative synthesis

- **Issues**

- Diffuse literature
- Heterogenous literature (large proportion grey literature)
- Qualitative and quantitative studies
- Not aggregative; interpretive

- **Strategy**

- A critical interpretive synthesis (CIS) approach

# Critical interpretive Synthesis (CIS)

**Box 1** Key characteristics of critical interpretive synthesis (CIS)<sup>6,7</sup>

Purpose	To further understanding of a topic/question by drawing on broadly relevant literature to develop concepts and theories that integrate those concepts. The topic might not be precisely bounded, and the initial question might be refined as the review progresses.
Process	The process of CIS is iterative, interactive, dynamic and recursive, with recognition of a need for flexibility and reflexivity. Searching, sampling, critique and analysis may happen concurrently.
Search strategy	Formal bibliographic searches may feature, but use will also be made of the research team's awareness of relevant literature from various fields and sources. The strategy may evolve organically.
Sampling	Sampling of studies may be selective and purposive (not necessarily aiming for comprehensive identification and inclusion of all relevant literature). Inclusion criteria can be flexible and to some extent emergent. Reflexivity informs sampling. Ongoing selection of potentially relevant literature is informed by emerging theoretical framework.
Quality appraisal	Some formal appraisal of methodological quality may be appropriate, but judgements about the credibility and contribution of studies may be deferred until synthesis, as methodologically weak papers may still prove theoretically or conceptually insightful.
Data analysis	Inductive – aims towards the development of a synthesizing argument. CIS involves an interrogation rather than aggregation of concepts and themes. Formal data extraction may be useful but is not essential to the approach.
Findings/results	CIS results in the generation of a 'synthesizing argument' linking existing constructs from the findings to 'synthetic constructs' (new constructs generated through synthesis). This network of relationships and categories is submitted to rigorous scrutiny as the review progresses.
Discussion, contribution	CIS aims to offer a theoretically sound and useful account that has explanatory power and is demonstrably grounded in the evidence. It explicitly acknowledges the 'authorial voice' and that some aspects of its production will not be auditable or reproducible.

*Entwistle V, Firnigl D, Ryan M, Francis J, Kinghorn P. Which experiences of health care delivery matter to service users and why? A critical interpretive synthesis and conceptual map. Journal of Health Services Research and Policy 2012, 17:70*

# Comparison of Systematic review & CIS

Systematic Review/Meta analyses	Critical Interpretive Synthesis (CIS)
Test theories	Generate theories
Aggregative synthesis	Interpretive synthesis
Summarizing data	Theory grounded in included studies
Explicit searching strategies- clear account of search for reproducibility reasons	'organic' search process- electronic databases, websites, reference chaining, use of experts within review team
Identification of all relevant material	Potentially relevant papers to provide a sampling frame
Specific boundaries	Boundaries diffused and ill-defined
Summary of ALL available data	Development of concepts and theory, not an exhaustive summary of all data- purposive sampling and theoretical sampling
Study design hierarchy	No hierarchy of study designs
Data extraction forms to identify characteristics of studies in a systematic way	Informal methods and data forms
Analyses	Identify recurring themes and developing a critique
Answerable question	Meaningful question/ exploratory

# Eligibility Criteria

- Qualitative or quantitative studies that report on revisions and or amendments to an established population screening program- an iterative process of modifying and refining of review question, a reflection of the complexity, scope and nature of the review
  - Studies on population-based screening programs- newborns/children/adults
  - Recommendation(s) to disinvest- deintensify and or deimplement
  - Recommendation can be international, national or provincial
  - No restriction on time recommendation was made

# Iterative process

Year	Age	Modality	Recommendation
2002	Any age	PSA test	Insufficient evidence to make a recommendation
2008	Less than 75 years	PSA test	Insufficient evidence to make a recommendation  Not Recommended
	75 Years or older	PSA test	
2012	Any Age	PSA Test	Not Recommended

- USPSTF-United States Preventive Services Task force on Prostate Screening
- PSA- prostate -specific antigen, DRE- Digital Rectal Exams



Identification

Records identified through electronic search strategy (n=9570)

Documents from reference lists and grey literature (n=In progress)

Screening

Total records screened after duplicates removed (n=8088)

**Records excluded (n=6853)**  
 \*not a population screening program  
 \*Opportunistic screening  
 \*no recommendation  
 \*Recommendation for treatment/surveillance

Eligibility

Full-text articles assessed for eligibility (n=1235)

**Records excluded (n=1224)**

Included

Studies included (n=11)

# Results: A range of screening programs

- Toxoplasmosis (Denmark): “no evidence of lasting benefit” (Prosser et al, 2012)
- Neuroblastoma: ended because of “no benefit from population screening, in terms of mortality rates.” (Botkin, 2005)
- Cervical Cancer
- Prostate cancer
- Mammography

# Results: An emphasis on evidence (as opposed to recommendations)

- The language of (for) disinvestment in the literature is vague
- Emphasis on evidence, abstract from a programmatic decision about what to do with the data
  - ‘...there is a large body of evidence supporting 2-yearly screening of women aged 50-69 years’ (Evans and Whelehan, 2011)
  - ‘... no evidence to suggest that a single screen between the ages of 47 and 50 within a programme screening at 3-year intervals will reduce mortality’ (Evans and Whelehan, 2011)
  - .... ‘the emphasis should be on utilizing evidence-based medicine to reduce overdiagnosis and overtreatment through less frequent screening’ (Makovey 2013)

Makovey I, Stephenson AJ, Haywood S. Response to the U.S. Preventative Services Task Force decision on prostate cancer screening. *Curr Urol Rep.* 2013 Jun; 14(3):168-73

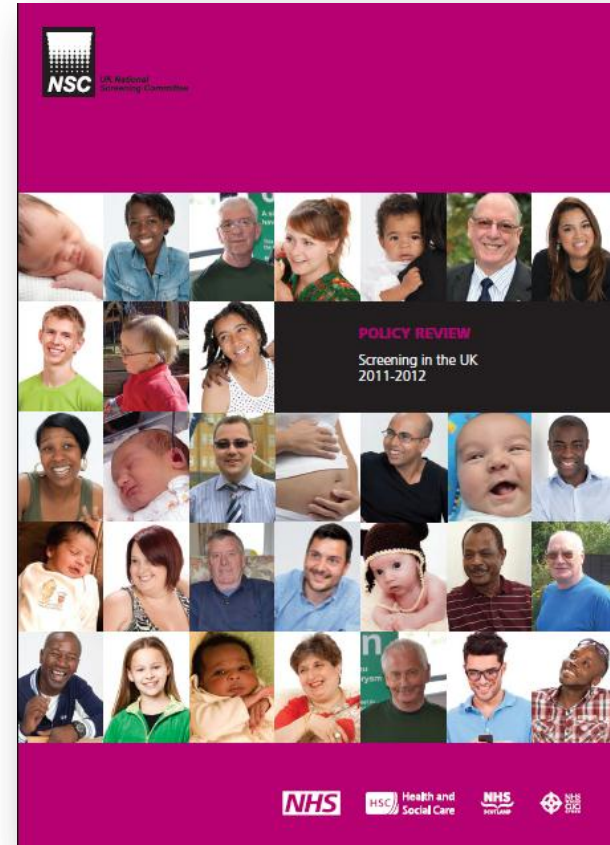
Evans A, Whelehan P. Breast screening policy: are we heading in the right direction? *Clin Radiol.* 2011 Oct;66(10):915-9.

# Results: Actively seeking the absent

- Recommendations tend to focus on micro, not macro issues:
  - Morbidity (lack of reduction in)
  - Mortality (as above)
  - Test quality
- Little if any mention of cost or cost-effectiveness
  - Contrast with many jurisdictional processes where cost per QALY or other metric is used for IMPLEMENTATION

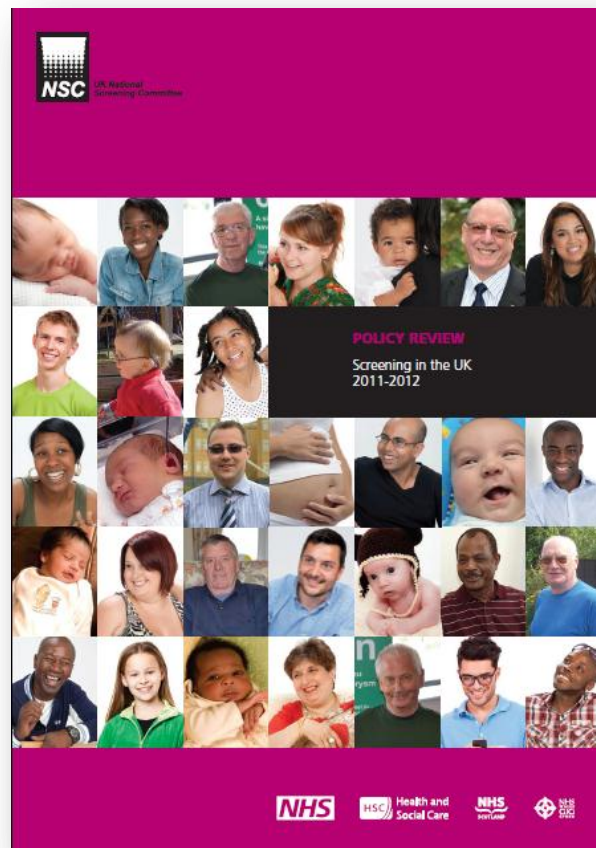
# Implicit values revealed: rationales inconsistent with established criteria

- There are concerns about the reliability of the current test
- While there is evidence that long term steroid treatment is beneficial, the optimum time at which it should be started remains uncertain
- There is insufficient evidence that identifying newborns with DMD by screening improves long term health compared to current practice
- It is not clear from the research what the impact of early diagnosis is on parents' subsequent reproductive decision making
- (UK National Screening Committee. *Policy Review. Screening in the UK 2011-2012*, UK National Screening Committee: London, UK 2012., P..13)



# Implicit values revealed: rationales inconsistent with established criteria

- There are concerns about the reliability of the current test
- While there is evidence that long term steroid treatment is beneficial, the optimum time at which it should be started remains uncertain
- There is insufficient evidence that identifying newborns with DMD by screening improves long term health compared to current practice
- **It is not clear from the research what the impact of early diagnosis is on parents' subsequent reproductive decision making**
- (UK National Screening Committee. *Policy Review. Screening in the UK 2011-2012*, UK National Screening Committee: London, UK 2012., P..13)



# Summary

- CIS provides an approach that is flexible given a diffuse and heterogenous literature
- Can incorporate traditional systematic review processes (e.g. systematic search), but has a different outlook
- Interpretative, as opposed to aggregative, analysis may throw into light missing elements or may emphasis contrasts that instigate further consideration/assessment
- To date little explicit and published consideration of disinvestment from screening
- A focus on health, not health systems
- Value systems are embedded and need further exploration and explication

Thank you for your attention



# Acknowledgements

- CIHR Knowledge Synthesis grant (KRS-140993)

- TEAM DETAILS

- Stuart G. Nicholls, University of Ottawa, Ottawa, Canada; Children’s Hospital of Eastern Ontario, Ottawa, Canada
- Tammy Clifford, Canadian Agency for Drugs and Technologies in Health, Ottawa, Canada; University of Ottawa, Ottawa, Canada
- Lindsey Sikora, University of Ottawa, Ottawa, Canada
- Jamie Brehaut, Ottawa Hospital Research Institute, Ottawa, Canada; University of Ottawa, Ottawa, Canada
- Richard Ashcroft, Queen Mary University of London, London, UK
- Ainsley Newson, The University of Sydney, Sydney, Australia
- Beth Potter, University of Ottawa, Ottawa, Canada
- Michael Wilson, McMaster University, Hamilton, Canada
- Ian Graham, Ottawa Hospital Research Institute, Ottawa, Canada; University of Ottawa, Ottawa, Canada
- Jeremy Grimshaw, Ottawa Hospital Research Institute, Ottawa, Canada; University of Ottawa, Ottawa, Canada
- Marcel Verweij, Wageningen University, Wageningen, Netherlands
- Angus Dawson, The University of Sydney, Sydney, Australia
- Doug Coyle, University of Ottawa, Ottawa, Canada
- Lesley Dunfield, Canadian Agency for Drugs and Technologies in Health, Ottawa, Canada
- Alex Kemper, Duke Clinical Research Institute, Durham, USA
- Jeff Botkin, University of Utah, Salt Lake City, USA
- John Lavis, McMaster University, Hamilton, Canada
- Spencer Anderson, University of Ottawa, Ottawa, Canada
- Fareeha Jafer, University of Ottawa
- Pearl Atwere, University of Ottawa, Ottawa, Canada



# DIMPLES

**D**eintensification and  
de**IMPLE**mentation from  
population **S**creening