Realist review of necessity argumentations

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INTRODUCTION

Public outrage often ensues when decision makers exclude forms of care, such as orphan drugs or expensive cancer medicines, based on an incremental cost-effectiveness ratio (ICER) that is below par. This outrage not infrequently precedes a reversal of the decision (Burls et al., 2005; Clarke et al., 2001). At the same time, however, not all forms of care with a sufficiently low ICER are covered. Viagra, for example, is highly effective and not that expensive but almost never provided by the state; decision makers deem it unnecessary to do so (Bernfort, 2003; Stolk et al., 2002). Hence, (cost-) effectiveness is not the decisive factor in all funding decisions. In these situations, another factor trumps it: the perceived *necessity* of coverage. To aid operationalisation, this chapter will survey the content, use, and context of the necessity criterion, an umbrella term for need- and solidarity-related argumentations used – not just decisively, and not just in coverage decisions made by policy makers and insurance companies.

In this chapter we will review argumentations underpinning the necessity, or lack thereof, of coverage of a certain treatment or therapy, as explicated in academic literature. To cast our net wide, we have chosen to include not only 'actual' decisions, that is, coverage decisions made by policy makers and insurance companies but also what we term 'hypothetical' coverage decisions. The latter type generally comes in the form of surveys (of, e.g., decision makers or the public) or ethical or economic analyses exploring possible reasons for (denial of) coverage. By examining both actual and hypothetical decisions we hope to provide insight into all potential considerations that may be invoked when deciding whether the coverage of a therapy or treatment is thought to be necessary. This is relevant as surveys and public opinion are considered of note (and of use) within coverage decision making practice (Ham, 1997; Mitton et al., 2009), as are scholarly reflections, as exemplified by the international take-up of the Accountability for Reasonableness framework (Kapiriri et al., 2009; D. Martin et al., 2002).

Objectives and focus of review

We followed the realist review method as described in the RAMESES publication standard (G. Wong et al., 2013). This method is used to review sundry literatures on a specific policy intervention, in order to describe why and how these interventions do what they do in their context. Using this method, researchers aim to uncover what works, for whom, and in what circumstances by conceptualising meta-level theories that detail patterns of how mechanisms-in-contexts lead to certain outcomes (Greenhalgh et al., 2011; Otte-Trojel et al., 2014; Pawson et al., 2005). In such an iterative research process, we refined how argumentations bearing upon necessity of coverage (mechanism) are used in justifying both actual and hypothetical coverage decisions (outcome), as found in academic literature. These decisions are made in context: by different decision makers from different countries, and, in case of actual decisions, placed on the decision agenda by different actors. Thus, the argumentations may be seen as interventions that have a proposed or actual outworking, also depending on contexts they are situated in. This review will address the following questions:

1. Which, if any, argumentations (mechanisms) are currently used in hypothetical and actual coverage decisions to justify whether coverage of a treatment is, or is not, necessary?
2. How do these argumentations justify the hypothetical and actual coverage decisions (outcomes) for different treatments, in different countries, put on the agenda by different agents (contexts)?

METHODS

Rationale for using realist synthesis

Little attention has been given to "the problem of operationalizing for decision makers essentially qualitative and normative criteria such as whether the technology serves an "ethical" or "medically necessary" purpose" (Giacomini, 2005). Furthermore, "social and ethical parameters of value (…) are anticipated to become as critical for reimbursement decisions (…) as economic and clinical criteria" (Akhmetov & Bubnov, 2015). In light of the lack of operationalisation and its (potentially) crucial role in coverage decisions, we conducted a literature review of the argumentation types that fall under the necessity criterion.

A realist review describes an intervention from different types of literatures, in our case actual coverage decisions (qualitative analyses of coverage decisions or policies), as well as hypothetical ones (economic analyses, ethical analyses, surveys, interviews, and opinion pieces). It searches these articles not just for information on the intervention (that is, the argumentation) but also for how the context (country, agenda setter) may have influenced the use of the intervention and its outcome (the decision including decision type: hypothetical or actual decision). This is subsequently summarised in context-mechanism-outcome patterns. From these patterns, meta-level theories are formulated that explain the working of these interventions-in-context. The primary reason for choosing the realist review method is practical; this method provided a focused lens to zoom in on particular aspects of actual and hypothetical coverage decisions, which in turn aided comparison of a broad variety of articles. Using this method for a non-classical intervention proved, moreover, an interesting methodological issue to grapple with. The second reason for utilising this method lies in its philosophical underpinnings. A realist philosophy holds that actors can and do effectuate change in context but are themselves shaped by the contexts they are part of. In this sense, it is likely to be acceptable to (social) scientists and policy makers alike.

Scoping the literature and searching processes

As an exploratory foray into grey literature and policy documents yielded too few explicated argumentations, we focused on peer-reviewed literature. For our primary background search thereof (Pawson et al., 2005), we used the conceptualisation of the necessity criterion in the Netherlands (Couwenbergh et al., 2013), as a request for operationalisation of this criterion from the Dutch National Health Care Institute catalysed this study. We subsequently discovered similar and/or underlying conceptualisations and related terminology in other countries, like 'need' and 'solidarity', which helped inform our search

strategy. The primary search was conducted in Embase (see Table 1 for search terms) and translated to Medline and Web of Science (Bramer et al., 2014), which is recognised to be an effective combination for reviews (Wichor M. Bramer). We used three general elements separated by the Boolean operator 'AND' as this kept the total number of articles workable (under 6,000). These general elements are a) the type of provision, b) the process of decision making, and c) the content in terms of criteria. Utilising a), we aimed for a representative sample, therefore a wide variety of provision were included (benefit package, health insurance, and/or health catalogue or service). For elements b) and c) specificity was the goal; we zoomed in specifically on coverage decisions (also often termed 'rationing' or 'priority setting' decisions) and precisely on those decisions that employ the necessity criterion. In selecting the exact search terms, we aimed for results that included the articles retrieved and selected from the primary background search, for example (Bernfort, 2003; Hoedemaekers & Oortwijn, 2003; Stolk et al., 2002). For each of the three elements we included relevant thesaurus terms (Emtree terms for Embase and MeSH terms for Medline). We excluded conference papers, letters, notes, and editorials, as well as articles written in any language other than English but did not employ any date restrictions (WM Bramer).

('insurance'/de OR 'health insurance'/de OR 'child health insurance'/de OR 'national health insurance'/de OR 'private health insurance'/de OR 'public health insurance'/de OR 'national health service'/de OR 'reimbursement'/de OR (insurance* OR reimburse* OR (national NEAR/3 (service OR coverage)) OR (cover* NEAR/6 deci*) OR ((partial* OR polic* OR universal OR unlimited OR limited OR temporar* OR permanent* OR recommend* OR plan OR plans) NEAR/3 coverage) OR (basic NEAR/3 package*) OR (health NEAR/3 catalogue*)):ab,ti)
AND
('decision making'/de OR 'ethical decision making'/de OR 'medical decision making'/de OR (decision* OR decide OR rationing OR priorit* OR (analys* NEAR/3 (inclusion OR exclusion)) OR (coverage NEAR/3 (negativ* OR positiv* OR determin* OR deny OR denial*))):ab,ti) AND ('resource allocation'/de OR (coverage OR inclusion* OR funding OR (resource* NEAR/3 allocat*) OR 'should be provided' OR 'what to provide'):ab,ti)
AND
('health care cost'/de OR 'cost of illness'/de OR 'economic evaluation'/exp OR ethics/de OR bioethics/de OR 'medical ethics'/de OR 'ethical decision making'/de OR 'health care policy'/de OR 'needs assessment'/de OR (necess* OR cost* OR (disease* NEAR/3 burden*) OR expenditure* OR solidarit* OR (therapeutic NEAR/3 (value* OR need*)) OR (budget* NEAR/3 impact*) OR ethic* OR 'health benefit*' OR (benefit NEAR/3 (risk OR analysis)) OR 'health technology assessment*' OR 'health care poli*' OR (need* NEAR/3 (assess* OR healthcare OR health-care))):ab,ti)
NOT
([Conference Abstract]/lim OR [Letter]/lim OR [Note]/lim OR [Editorial]/lim)
AND
[english]/lim

Table 1 - Search terms used in Embase

Selection and appraisal of documents

The first author (TKV) scanned titles, abstracts, and keywords in Endnote to include decisions that were made on the macro (government) or meso (local health authorities, sickness funds, and insurance companies) level (Polikowski & Santos-Eggimann, 2002). BB, the last author, scanned a random subset of 537 studies. Together, an agreement rate of 96% was reached and further disagreement was resolved through discussion. This first round of inclusions amounted to a total of 666 studies. Next, TKV read all candidate papers in full and excluded 594 of the 666, ending up with 72 studies. Through snowballing, a further 26 such studies were added, bringing the total number to 98 (see Figure 1).

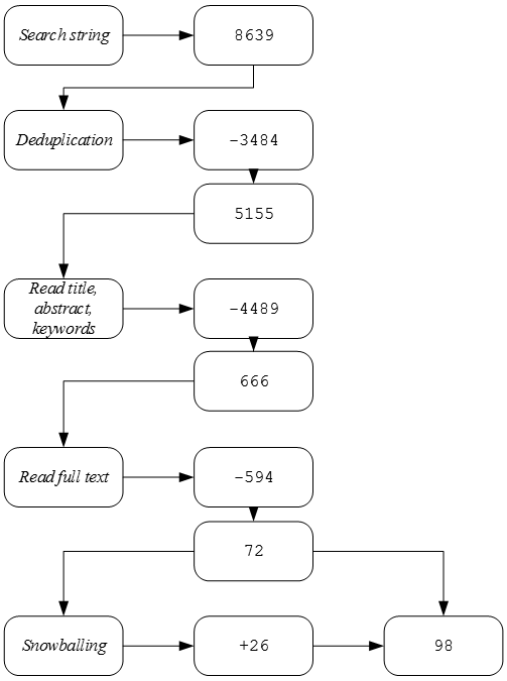

Figure 1 - Document flow diagram

The inclusion criteria were:

a) the article describes a hypothetical or actual coverage decision made concerning a specific form of care, treatment, therapy or medical technology,
b) on the meso or macro level, and
c) employing necessity-related considerations.

We correspondingly excluded articles that:

a) describe generalised criteria not applied to a specific form of care,
b) concern individual decisions (that is, for one patient) as they may concern the exception rather than the rule, and
c) employ only effectiveness and/or cost-effectiveness or other quantitative argumentations.

In doing so, we aimed to find studies containing concrete, qualitative, in-use argumentations that influence different types of coverage decisions as formulated by different actors on different levels regarding different forms of care.

Data extraction

TKV extracted data from these 98 articles using Microsoft Excel and ATLAS.ti. During this process, 'mode of agenda setting' emerged as a potential context of influence. Conversely, 'type of disease'

was removed as it proved unfruitful. To score the context, we noted country, year, macro- or meso level, and decision maker per identified argumentation, which was in turn scored as mechanism. The coverage decision was scored as the outcome, with eight outcome categories, of which three were used in case of an actual coverage decision:

- Yes – the treatment is unconditionally covered/conditions unspecified,
- Conditional – the treatment is covered for specific patient groups/indications,
- No – the treatment is never covered.

Three categories were scored as hypothetical decisions (generally based on theory, interviews, or surveys):

- Should – the treatment should be unconditionally covered/conditions unspecified,
- Should conditional – the treatment should be covered for specific patient groups or indications,
- Should not – the treatment should never be covered.

Furthermore, two categories were added later on to signify when an author or actor noted whether an argumentation should, or should not, have a bearing upon the decision.

- Valid – the consideration should be taken into account,
- Not valid – the consideration should not be taken into account.

BB extracted data from a random subset of 11 studies, reaching an agreement rate of 73%, and full agreement was reached in deliberation.

Analysis and synthesis process

Data analysis was undertaken by TKV, and candidate context-mechanism-outcome combinations were formulated according to the following pattern:

Based on these argumentation types, (mechanism)
used in country C, put on the agenda by A, (contexts)
lead to actual or hypothetical in- or exclusion of treatment T. (outcome)

Discussion with BB and AdB enabled the formulation of four meta-level theories.

RESULTS

Document characteristics

The 98 reviewed studies contain studies that are (in part) based on qualitative analyses of coverage decisions or policies (34 studies), interviews/surveys (26 studies), and case studies (twenty studies), but also ethical analyses (fourteen studies), opinion pieces (fourteen studies), economic analyses (eight studies), and reviews (seven studies), from over twenty (primarily Western) countries.

Main findings

General overview

In this chapter twenty argumentation types are described that are in use in hypothetical and actual coverage decisions. These twenty argumentation types are derived from over 400 argumentations found in 98 articles, for an overview of the argumentation types in alphabetical order, see Table 2.

Argumentation type	Description
1) Definition of Illness	Whether the ailment is considered an illness for which treatment is necessary
2) Dignity	Whether (lack of) coverage is considered to affect the dignity of the patient to such an extent that it needs to be amended
3) Equity/Fairness/Justice	Whether coverage would be necessary to counter injustice/inequity/lack of fairness in (access to) treatment
4) Human Right	Whether (lack of) coverage is considered to affect the human rights of the patient to such an extent that it needs to be amended
5) Individual Cost	Whether lack of coverage would stop patients from buying necessary care themselves due to prohibitive cost
6) Individual Responsibility	Whether the individual is considered responsible for paying for this treatment
7) Medical Necessity	Whether or not a treatment is considered to be "medically necessary" or a "medical necessity"
8) Morbidity/Severity	Whether the physical and/or psychosocial morbidity associated with a certain ailment constitutes such a need that coverage is considered necessary
9) Moral Hazard	Whether there is considered to be a possibility of over-usage (i.e., unnecessary increase in demand, when people use more than they need as a result of coverage)
10) Need	The extent to which the patient is considered to be in need for which treatment is necessary
11) (No) Alternative	Whether or not viable alternatives are considered to be present which would make coverage more or less necessary
12) Patient-Diagnosis	Whether an illness is self-reported rather than diagnosed by a doctor
13) Range of Normality	Whether the experience of the patient is considered normal or abnormal to such an extent that coverage is deemed necessary
14) Rule of Rescue	Whether the identifiability of individuals close to death is considered to heighten the necessity of coverage
15) Similar Treatments	Whether similar treatments are covered or not (meaning that this *type* of treatment is considered necessary)
16) Small Number of Patients	Whether the small size of the patient population is considered to heighten the necessity of coverage (due to, amongst others, the inequality in terms of research expenditure or difficulties in obtaining high-quality data)
17) Societal Impact	Whether coverage is considered necessary to allay the impact this disease has on people beyond the patient
18) Societal Functioning	Whether coverage would aid a person's necessary functioning in society
19) Societal Responsibility	Whether society is considered responsible for this necessary treatment
20) Vulnerability/Compassion	Whether a compassionate response to vulnerable groups, e.g. children, in the form of coverage is considered to be a necessity

Table 2 - Overview of the twenty argumentation types that fall under the necessity criterion and their respective descriptions

The 98 reviewed articles are divided up into seven treatment sets (cancer therapies, orphan drugs, infertility treatments, Viagra, cosmetic surgery, obesity treatment, and smoking cessation therapy). What follows is a detailed description of the use of these twenty argumentation types per treatment set in justifying positive, negative, and/or conditional coverage decisions, both actual and hypothetical. A handful of included articles is not described because there was too little information on that type of treatment: Alzheimer's disease (Bernardi & Pegoraro, 2003), genetic tests (Fischer & Rogowski, 2014; Trosman et al., 2015), and medical devices (Kisser et al., 2016). Notably, no new argumentation types are described in these articles.

1. Cancer therapies

For cancer therapies, we observed solely positive coverage decisions. For these, higher incremental cost-effectiveness ratios than usual are acceptable (Chabot & Rocchi, 2010), as evidenced by positive decisions made despite the fact that coverage would be inadvisable based on high cost (Drummond & Mason, 2007), or "lack of clear clinical consensus about [a therapy's] benefits" (Daniels & Sabin, 1998). Moreover, a few articles pose that "patients should be treated equally" regardless of the number of patients suffering from the specific type of cancer.

The main argumentation visible in this data subset poses that the need of the patient and the severity of the disease should be taken into account (Cookson et al., 2008; Linley & Hughes, 2013; Rocchi et al., 2008; Stafinski et al., 2014; Trowman et al., 2011). As cancer patients have urgent needs and a serious health condition, therefore therapy is, or should be, covered (Aggarwal et al., 2014; Brock, 2010; Cookson et al., 2008; Daniels & Sabin, 1998; Drummond et al., 2009; Drummond & Mason, 2007; D. Martin et al., 2001; Rocchi et al., 2008; Singer et al., 2000), heightened by concerns over dignity and impact on daily activities and family (Stafinski et al., 2014). The Rule of Rescue is the second main argumentation, where the necessity of covering a certain treatment is high specifically for identifiable patients who are in a life-threatening situation and are without alternative (Brock, 2010; MacKenzie et al., 2008; Singer et al., 2000). The unavailability of alternative treatments or therapies should thus be considered (Cookson et al., 2008; Daniels & Sabin, 1998; Drummond et al., 2009; Lim et al., 2014; Linley & Hughes, 2013), as visible in several coverage decisions (Chabot & Rocchi, 2010; Cookson et al., 2008; Daniels & Sabin, 1998; Drummond et al., 2009; Drummond & Mason, 2007; Lim et al., 2014; Linley & Hughes, 2013; D. Martin et al., 2001; Singer et al., 2000), but this argumentation is also sometimes questioned (Brock, 2010; Cookson et al., 2008). The 'identifiable individuals' element is also present in the 'Small Number of Patients' argumentation, which several authors pose is important (Cookson et al., 2008; Stafinski et al., 2014; Trowman et al., 2011) and as heightening justifiability (Chabot & Rocchi, 2010; Drummond et al., 2009), although others deny this (Cookson et al., 2008; Linley & Hughes, 2013; Singer et al., 2000). The third main argumentation holds that for some, like children, exceptions are (Lim et al., 2014; Rocchi et al., 2008), may (Cookson et al., 2008; Stafinski et al., 2014), may not (Linley & Hughes, 2013), or should be made (Brock, 2010). This is expanded by Brock, who argues that those who have had fewest QALYs in their lifetime are the worst off, which is most obviously the case

for paediatric patients (Brock, 2010). The final argumentation concerns the distribution of health care resources (Aggarwal et al., 2014; Drummond & Mason, 2007). E.g., Foy et al. emphasise that patients in different districts should be treated equally, thus arguing for more distributive justice (Foy et al., 1999).

2. Orphan drugs

For orphan drugs, we identified argumentations for both positive and negative coverage decisions. The structure and types of argumentations of positive decisions in this data subset greatly resemble those for cancer therapies and the use of a number of argumentations is questioned here as well.

Higher incremental cost-effectiveness ratios are generally found acceptable for orphan drugs so decisions reek of making exceptions to this rule. The fact that the severity of the illness does or should count as an argumentation is stressed by many (Bae et al., 2015; Clarke et al., 2001; Cohen & Felix, 2014; Denis et al., 2011; Dyfrig Hughes, 2006; Mentzakis et al., 2011; Nicod, 2016; Rosenberg-Yunger et al., 2011; Schlander & Beck, 2009; Winquist et al., 2012; Zelei et al., 2016), and so is need (Cohen & Felix, 2014; Nicod, 2016). Consequently, argumentations in favour of coverage include that orphan diseases are severe (Drummond & Towse, 2014; Dyfrig Hughes, 2006; Largent & Pearson, 2012; Paulden et al., 2015), that coverage only happens when they are (Clarke et al., 2001) and/or when patients are in need (Henschke, 2012). Patients are said to be vulnerable, which should be (Largent & Pearson, 2012; Winquist et al., 2012), and is (Bañón Hernández Antonio, 2015; Henschke, 2012), considered, and to score low on societal functioning, and orphan drugs should be covered because they maintain or restore these capacities (Largent & Pearson, 2012). As for cancer therapies, a common argumentation supportive of provision is that patients have "no alternative" (Drummond & Towse, 2014; Iskrov et al., 2013; Paulden et al., 2015), often generally considered important (Cohen & Felix, 2014; Denis et al., 2011; Rosenberg-Yunger et al., 2011; Zelei et al., 2016). The Rule of Rescue argues in favour of coverage in two cases (Bañón Hernández Antonio, 2015; Clarke et al., 2001), its relevance as a consideration is underlined by some (Bae et al., 2015; Rosenberg-Yunger et al., 2011; Zelei et al., 2016) but also regularly questioned (Burls et al., 2005; Arna S Desser et al., 2010; Gross, 2002; David Hughes et al., 2005; Juth, 2014; Largent & Pearson, 2012; Schlander & Beck, 2009). The small patient population is mentioned as an argumentation separately as well but primarily to say it should not be of effect on the coverage decision (Burls et al., 2005; Arna S. Desser, 2013; Arna S Desser et al., 2010; Drummond & Towse, 2014; David Hughes et al., 2005; Juth, 2014; McCabe et al., 2005; Mentzakis et al., 2011; Paulden et al., 2015), though not always (Dyfrig Hughes, 2006; Nicod, 2016; Zelei et al., 2016). Finally, the societal impact of treatment should be considered (Zelei et al., 2016). A new main argumentation in favour of coverage holds that the budget impact remains "sufficiently insignificant" given the low numbers of patients (David Hughes et al., 2005), as is the concurrent fact that the price is prohibitive to the individual (Denis et al., 2011). "Equity" and distributive justice are once again mentioned but the exact line of reasoning is not always explicated (Burls et al., 2005; David Hughes et al., 2005; Rosenberg-Yunger et al., 2011; Zelei et al., 2016), as is the case for the right to health care (David Hughes et al., 2005).

Arguing for denial of coverage, the most common assertion is that some orphan diseases may not be as severe as assumed, e.g. Gaucher's disease is said to be "minimally symptomatic" (Clarke et al., 2001; Gross, 2002). Hughes et al. state that orphan diseases may not "pose *sufficient imminent threat* to the life of patients to constitute a right to treatment" (italics mine) (David Hughes et al., 2005), see also (Juth, 2014). One coverage decision cites "equity" as a reason (Rosenberg-Yunger et al., 2011). Finally, denial of coverage for similar treatments may support another negative coverage decision (Burls et al., 2005; Winquist et al., 2012).

3. Infertility treatments

The infertility treatment data subset was the largest and the most diverse, containing positive coverage decisions, including those that set indication criteria, as well as negative coverage decisions.

The first main argumentation favouring coverage is that those seeking infertility treatment are in need (Blank, 1997; McMillan, 2001; McWhirter & McQueen, 2000; Rauprich et al., 2010; Redmayne & Klein, 1993; Shaw et al., 2002), resulting in a positive coverage decision once (Redmayne & Klein, 1993). Further, they are suffering morbidity (Brown, 2000; Chambers et al., 2013; E. G. Hughes & Giacomini, 2001; Lord et al., 2001; Redmayne & Klein, 1993; Shaw et al., 2002), explicated as mental distress and psychological harm in decisions by UK health authorities (Redmayne & Klein, 1993). Treatment may be medically necessary (Nachtigall et al., 2012) and patients therefore deserve compassion (Giacomini et al., 2000; Nachtigall et al., 2012). The second argumentation holds that the "cost of [coverage of] assisted conception would be surprisingly small to the NHS as a whole, although to an individual it is often prohibitive" (Lord et al., 2001) and (Chambers et al., 2013; Chambers et al., 2006; E. G. Hughes & Giacomini, 2001). Third, equity is mentioned again, but the underlying line of reasoning is more clearly explicated here, though not used in actual decisions. Lack of geographical equity as well as equity over the rich-poor divide is cause for concern (Brown, 2000; Chambers et al., 2006; Giacomini et al., 2000; E. G. Hughes & Giacomini, 2001; Lord et al., 2001; McWhirter & McQueen, 2000; Nachtigall et al., 2012; Redmayne & Klein, 1993; Shaw et al., 2002). Postcode and financial status should not affect coverage, and it would therefore be preferable to cover infertility treatment everywhere. Moreover, it is deemed inequitable to withhold treatment based on the environmental impact of having a child (Wilkinson & Williams, 2015). The fourth and fifth argumentations are not visible in the first two datasets, and both are only in use in hypothetical decisions, made by authors, patients, and the public. The fourth holds that infertility, or subfertility (E. G. Hughes & Giacomini, 2001), is an illness and its cure should therefore be covered (Brown, 2000; Chambers et al., 2013; Giacomini et al., 2000; E. G. Hughes & Giacomini, 2001; Lord et al., 2001; McMillan, 2001; McWhirter & McQueen, 2000; Mladovsky & Sorenson, 2010; Nachtigall et al., 2012; Neumann, 1997; Plomer et al., 1999; Rauprich et al., 2010; Redmayne & Klein, 1993; Wilkinson & Williams, 2015), especially because similar services are covered (E. G. Hughes & Giacomini, 2001; Lord et al., 2001; Wilkinson & Williams, 2015). Fifth, parenthood is part of the "right to reproduce" (Giacomini et al., 2000) or right to health (Mladovsky & Sorenson, 2010), required for societal functioning (Chambers et al., 2013; Lord et al., 2001; McMillan, 2001;

Mladovsky & Sorenson, 2010; Neumann, 1997; Redmayne & Klein, 1993) and part of "the basic opportunities every human should have" (Rauprich et al., 2010), also (Blank, 1997) and that those without children suffer stigmatisation (Chambers et al., 2013; Lord et al., 2001) and their dignity is impacted (E. G. Hughes & Giacomini, 2001). Lastly, the societal impact of children, in economic terms, should be considered (Connolly et al., 2010).

For the negative coverage decisions, we observe several argumentations, of which some are new. First, infertility is not an illness and therefore not covered (Neumann, 1997; Redmayne & Klein, 1993) or should not be (Wilkinson & Williams, 2015), also because it is not life-threatening (Neumann, 1997). In some cases, it is only covered in case of a diagnosed cause (Brown, 2000; McWhirter & McQueen, 2000). Other needs are thought "more pressing" (Giacomini, 2005; E. G. Hughes & Giacomini, 2001), the treatment risky and not medically necessary (E. G. Hughes & Giacomini, 2001), also the reason for exclusion from coverage in Ontario (Giacomini et al., 2000). Infertility has little emotional appeal (E. G. Hughes & Giacomini, 2001) and its treatment should therefore fall to personal, and not societal, responsibility (Blank, 1997; E. G. Hughes & Giacomini, 2001; Plomer et al., 1999). Other argumentations that support a hypothetical negative coverage decision include potential moral hazard (Blank, 1997; Mladovsky & Sorenson, 2010; Neumann, 1997) and that alternatives are available (Nachtigall et al., 2012; Wilkinson & Williams, 2015), of which the latter also used in an actual decision (Giacomini, 2005).

For the decisions that set indication criteria; Mladovsky and Sorenson describe how the necessity of coverage is lowered when doctors rely "on patients to ascribe infertility" (Mladovsky & Sorenson, 2010). Argumentations with concrete, micro-level indication criteria include, for example, "the number of children living in the home/from previous relationships" (Brown, 2000; McWhirter & McQueen, 2000; Plomer et al., 1999; Shaw et al., 2002).

4. Viagra

Viagra is almost never covered (Klein & Sturm, 2002; Stolk et al., 2002), and moreover, about half of the argumentations oppose coverage. The other half contains both positive decisions and those that set indication criteria.

Viagra is, in actual coverage decisions, considered not medically necessary (Klein & Sturm, 2002; Stolk et al., 2002), patients not in need (Klein & Sturm, 2002), and erectile dysfunction not serious enough to prevent societal functioning (Hoedemaekers & Oortwijn, 2003) or to be a normal part of ageing (Stolk et al., 2002). Moreover, the apparent subjectivity of this determination of need by the patient (rather than the doctor) may have further loosened the concept of 'medical necessity' for Viagra. This may well effectuate over-usage, as policy makers feel patients cannot be fully relied upon to make this kind of decisions (Klein & Sturm, 2002). Moreover, because Viagra is available "over-the-web," the doctor is thus conclusively unneeded for diagnosis and prescription, which decreases the necessity of coverage, further strengthened by fear of moral hazard (Klein & Sturm, 2002).

Others, primarily authors, do support coverage of Viagra, based on the severity of psychosocial problems (Hornbrook & Holup, 2011; Klein & Sturm, 2002; Manson, 2005), impeded societal functioning as sexual function is considered essential to quality of life (Hornbrook & Holup, 2011) and positive coverage of similar treatments (Hornbrook & Holup, 2011).

Indication criteria are set in several countries, generally holding that Viagra as a treatment for erectile dysfunction is covered in cases of a spinal cord injury only, an argumentation related to the definition of illness and morbidity (Klein & Sturm, 2002; Manson, 2005; Stolk et al., 2002). This use of the definition of illness argumentation is also questioned once (Manson, 2005).

5. Cosmetic surgery

Both the public and policy makers often regard cosmetic surgical procedures like tattoo removal or 'boob jobs' a prime example of a form of care that should not be funded (Polikowski & Santos-Eggimann, 2002; Russell et al., 2014); in several countries it is indeed excluded from coverage (Schreyögg et al., 2005). The decisions examined all set limits on eligibility through indication criteria or, in cases like circumcision, exclude the form of care altogether.

The main argumentation is offered in support of a decision setting indication criteria, that is, cosmetic surgery is generally reimbursed for certain indications only. All decisions, which included primarily actual and a few hypothetical decisions, use certain physical characteristics, evidence of physical or psychosocial morbidity, reduced social capacity, or a combination thereof, to argue for necessity of coverage *in certain cases*. Decision makers thus set physical or psychological patient eligibility criteria on the morbidity experienced that need to be applied by a physician (Adler, 2011; Benditte-Klepetko et al., 2007; Breuning et al., 2010; Cook et al., 2003; Goodson et al., 2011; Henderson, 2009; Horner, 2002; Kerrigan et al., 2002; Krieger & Lesavoy, 2001; McClean & Hanke, 1997; Mukherjee et al., 2014; Nguyen et al., 2008; Russell et al., 2014; Stevens et al., 2015; A. M. Wong, 1995; Wraight et al., 2007). Individual assessment providing evidence of morbidity here leads to a higher justifiability of surgery. In this way, surgery is performed only on those patients that fall on the 'right' side of the line between cosmetic and non-cosmetic care (Goodson et al., 2011; Krieger & Lesavoy, 2001) or outside a "range of normality" (Breuning et al., 2010; Cook et al., 2003; Goodson et al., 2011; Henderson, 2009; Kerrigan et al., 2002; McClean & Hanke, 1997; Mukherjee et al., 2014; Nguyen et al., 2008; Nicoletti et al., 2009; Russell et al., 2014; Schnur et al., 1991; Stevens et al., 2015) so that care is only provided if medically necessary (Benditte-Klepetko et al., 2007; True, 2012). Some argue that cosmetic surgery should be covered only in case of societal functioning problems (Breuning et al., 2010; Henderson, 2009; Mukherjee et al., 2014), which McClean and Hanke argue to be affected only in some cases (McClean & Hanke, 1997). Directly related is the illness definition (Adler, 2011; Stevens et al., 2015): what counts as illness needs treatment and what falls inside the 'range of normality' does not need treatment.

This is also reflected in the argumentation supporting a negative coverage decision, stating that because "newborns do not have a medical condition," circumcision should not be covered (Adler, 2011), see also (Darby, 2016; Krieger & Lesavoy, 2001). Other argumentations against coverage

include, for actual decisions, the availability of alternative treatments (True, 2012) and the lack of medical necessity (McClean & Hanke, 1997), the latter also in hypothetical decisions (Adler, 2011; Darby, 2016; Jacobs, 1980).

Argumentations for a hypothetical positive decision were offered primarily for treatment of port-wine stains, with medical complications and psychosocial morbidity explicated (McClean & Hanke, 1997), also (Henderson, 2009). The (potential of a) 'postcode lottery' was brought up a number of times, to apparently argue in favour of coverage (Goodson et al., 2011; Henderson, 2009; Mukherjee et al., 2014; Stevens et al., 2015; Wraight et al., 2007).

6. Obesity treatments

Obesity treatments are often excluded from coverage (Polikowski & Santos-Eggimann, 2002). However, various argumentations in support of coverage are visible in the literature and a few in combination with indication criteria. In some cases, argumentations against coverage are given, but there is no evidence of public outrage over these negative decisions (Greer et al., 2012; L. F. Martin et al., 1998; Persson et al., 2010; Simpson & Cooper, 2009).

Articles mention the psychosocial burden of obesity (L. F. Martin et al., 1998; Persson et al., 2010) as supporting hypothetical coverage decisions, as well as the fact patients should be enabled to "conform to societal standards," which is linked to the range of normality mentioned for cosmetic surgery. For actual decisions, potential co-morbidities (Persson et al., 2010), as well as the lack of alternatives and simply "need and solidarity principles" (Persson et al., 2010) are brought up. Further hypothetical argumentations favouring coverage include societal responsibility for these patients (Greer et al., 2012), "fair treatment" (L. F. Martin et al., 1998), consideration of "human dignity" (Persson et al., 2010), and "social justice" (Greer et al., 2012).

Clear argumentations in support of decisions that set indication criteria are visible in decisions by American insurance companies, with positive decisions based on an expert opinion (L. F. Martin et al., 1998), the medical necessity as judged by a doctor (Simpson & Cooper, 2009) or a health risk assessment (Greer et al., 2012).

Conversely, the argumentations opposing coverage used by insurance companies in the USA hold that the individual patient is "to blame," and that obesity treatment should therefore not be included (Greer et al., 2012; L. F. Martin et al., 1998) but also that obesity is not an illness (L. F. Martin et al., 1998) or not severe enough (Simpson & Cooper, 2009).

7. Smoking cessation therapies

Smoking cessation therapy is covered in countries such as Australia, Canada, England, and the USA, though not in Argentina (Specogna, 2010), and it has been included and excluded several times in the Netherlands (Kroneman & de Jong, 2015). The subset contains negative and positive coverage decisions and focuses on the question of societal versus individual responsibility, only observed once before (in the obesity treatment subset).

For negative coverage decisions, two argumentations were elucidated. First, smoking or the cessation of it is considered in actual decisions to be a "personal" issue (Au-Yeung et al., 2010; Heath et al., 2002). Second, "tobacco consumption [is] not understood as an addiction or illness," which reduces the necessity of coverage.

For positive coverage decisions, the four argumentations include first, for actual decisions, the obligation to attempt "safe-guarding the wellbeing of fund participants" (Au-Yeung et al., 2010). Second, for hypothetical decisions, it is argued that distributive rather than retributive justice ought to prevail (Heath et al., 2002). Third, addiction is an illness (Heath et al., 2002; Woolf et al., 2006), also in use in actual decisions (Kroneman & de Jong, 2015). The fourth, though only used in hypothetical decisions, simply underlines the "need" for or "necessity" of smoking cessation services (Au-Yeung et al., 2010; Heath et al., 2002; Khalid, 1993; Specogna, 2010). The Dutch policy for coverage of smoking cessation therapy deserves special attention (Kroneman & de Jong, 2015). Smoking cessation therapy was excluded supported by the argumentation that the costs "could easily be paid from the savings that quitting smoking produced for individuals."

Analysis

In order to identify context-mechanism-outcome patterns (Pawson et al., 2005), we classified the twenty argumentation types per decision outcome: positive, negative, and conditional coverage (Table 3). First, seven argumentation types are generally used in the justification of positive coverage decisions (both hypothetical and actual), namely: Equity/Fairness/Justice, Societal Functioning, Individual Cost, the coverage of Similar Treatments, Human Rights, Dignity, and Societal Impact. Second, five argumentation types are solely in use in the justification of negative coverage decisions (both hypothetical and actual): Medical Necessity, Individual Responsibility, Moral Hazard, Small Number of Patients, and Patient-Diagnosis. Finally, eight argumentation types are employed for all types of coverage decision outcomes. This set comprises Morbidity/Severity, Range of Normality, Definition of Illness, Vulnerability/Compassion, Need, (No) Alternatives, Societal Responsibility, and the Rule of Rescue. Conditional coverage is, moreover, generally based on Morbidity/Severity, Definition of Illness, Range of Normality, Societal Functioning, and Medical Necessity – doctors are in these cases may make the final individual coverage decision. From Table 3, we thus observe that nearly two-thirds of the argumentation types is in use for either positive or negative decisions, from which we conclude the **first meta-level theory**: different argumentation types are generally used for different decision outcomes.

Table 4 gives a précis of those decisions where policy makers made a coverage decision responding to a patient (representative), the media, or the public, and it was explicitly analysed in the article as such. These actors were thus reported to be involved in setting the decision on the agenda, often reacting to a negative decision. We quickly detected that all coverage decisions in this table have a positive outcome. We first identified two decisions (Aggarwal et al., 2014; Burls et al., 2005) that use an argumentation type, in both cases Equity/Fairness/Justice, which is generally used in positive decisions (as visible in Table 3), meaning that the positive outcome was to be expected from the

Mechanism	Outcome					
Argumentation type	POSITIVE COVERAGE		NEGATIVE COVERAGE		CONDITIONAL COVERAGE	
	Actual	Hypothetical	Actual	Hypothetical	Actual	Hypothetical
Equity/Fairness/ Justice		Cancer; Cosmetic; Infertility; Obesity; Orphan; Smoking	Orphan			
Societal Functioning	Infertility	Cancer; Cosmetic; Infertility; Obesity; Orphan; Viagra	Viagra		Cosmetic	
Individual Cost		Infertility; Orphan	Smoking			
Similar Treatments		Infertility; Viagra	Orphan			
Human Right		Infertility				
Dignity		Cancer; Obesity				
Societal Impact		Cancer; Infertility				
Medical Necessity		Smoking	Cosmetic; Infertility; Viagra	Cosmetic; Infertility	Obesity	Cosmetic
Individual Responsibility			Infertility; Smoking	Obesity		
Moral Hazard				Infertility; Viagra		
Small Number of Patients	Cancer			Cancer; Orphan		
Patient-Diagnosis				Viagra		
Morbidity/Severity	Cancer; Infertility; Viagra	Cancer; Cosmetic; Infertility; Obesity; Orphan; Viagra	Cosmetic; Obesity; Viagra	Cosmetic; Infertility; Orphan	Cosmetic; Infertility; Orphan; Viagra	
Range of Normality		Cosmetic; Infertility	Viagra	Cosmetic	Cosmetic; Infertility	Cosmetic
Definition of Illness	Obesity; Smoking	Infertility; Smoking; Viagra	Infertility; Obesity; Smoking	Infertility	Cosmetic; Infertility; Obesity; Viagra	Cosmetic
Vulnerability/ Compassion	Cancer; Orphan	Cancer; Cosmetic; Infertility; Orphan	Cosmetic	Infertility		
Need	Cancer; Infertility; Obesity	Cancer; Cosmetic; Infertility; Smoking	Viagra	Infertility; Orphan		
[No] Alternative	Cancer; Obesity	Cancer; Infertility; Orphan	Cosmetic; Infertility	Infertility		
Societal Responsibility	Smoking	Obesity	Infertility	Infertility		Smoking
Rule of Rescue		Cancer; Orphan		Cancer; Orphan	Orphan	

Table 3 - Articles containing argumentations used to justify coverage decisions per treatments or therapies. 'Cancer' stands for cancer therapies, 'orphan' for orphan drugs, 'infertility' for infertility treatments, 'cosmetic' for cosmetic surgery, 'obesity' for obesity treatment and 'smoking' for smoking cessation therapy. NB: When a particular argumentation-outcome combination occurred only in one article, the cell is coloured light grey.

Context		Mechanism	Outcome
Country	Actor	Argumentation type(s)	Decision
UK	Politician	Equity/Fairness/Justice	Positive
Canada	Patient representative	Equity/Fairness/Justice	Positive
Canada	Patient representative	Rule of Rescue	Positive
Australia	Journalist	Rule of Rescue Small Number of Patients No Alternative	Positive
USA	Patient	Morbidity/Severity Medical Necessity Societal Functioning	Positive
UK	Patient	(not explicated, possibly Morbidity/Severity or Need)	Positive
Netherlands	Lobbyist	(not explicated, possibly Societal Responsibility or Societal Impact)	Positive

Table 4 - Overview of decisions where it was recorded that the public, a patient or a patient representative was involved in setting the coverage decision on the policy agenda, with the argumentation type and final decision

argumentation type used. However, we also found five cases that employ argumentation types also sometimes used to justify negative decisions, but that had, in fact, also a positive outcome (Burls et al., 2005; Clarke et al., 2001; Kroneman & de Jong, 2015; MacKenzie et al., 2008; McClean & Hanke, 1997). From this, we conclude that who set the decision on the agenda may have played a role in these cases. The **second meta-level theory** is therefore: when the public or a patient (representative) is recorded to have been involved in setting the decision agenda, regardless of the argumentation type used, the decision outcome is positive in this dataset. Indubitably, we would need a more extensive analysis of grey literature and media coverage to 'prove' this conclusively or to draw conclusions for unexamined coverage decision processes. It seems conceivable, however, that the working of the argumentation types depends upon the context. We conclude that an important context is who set the agenda; when a patient (representative), lobbyist, journalist, or politician was recorded to be involved, the resulting decision was always positive.

For the latter two meta-level theories, we utilise the distinction between 'actual' and 'hypothetical' coverage decisions. Actual decisions are those made by policy makers and insurance companies, that is, they concern coverage policies and therefore directly affect patients' access to medicine or therapy. Hypothetical decisions are those made by all other parties, which includes patients, authors (generalising, primarily ethicists and economists), the general public, and the media. We grouped the twenty necessity argumentation types into two broad categories, which are based on the type of decision (actual vs. hypothetical) they are used in. The categorisation is then made as follows: the first category is argumentations used in *both* actual and hypothetical decisions, the second category contains argumentations *only* used in hypothetical decisions.

The first category (as visible in the upper half of Table 5) contains ten argumentation types: Definition of Illness, Individual Responsibility, Medical Necessity, Morbidity/Severity, Need, (No) Alternative, Range of Normality, Societal Functioning, Societal Responsibility, and Vulnerability/ Compassion. The lower half of Table 5 holds the second category of necessity argumentations,

Mechanism		Outcome	
		Decision type	
Argumentation type		Actual decision [Policy makers and insurance companies]	Hypothetical decision [Patients, authors, the public, and the media]
Used in both actual and hypothetical decisions	Definition of Illness	Cosmetic; Infertility; Obesity; Smoking; Viagra	Cosmetic; Infertility; Smoking; Viagra
	Individual Responsibility	Infertility; Smoking	Obesity
	Medical Necessity	Cosmetic; Infertility; Obesity; Viagra	Cosmetic; Infertility; Smoking
	Morbidity/Severity	Cancer; Cosmetic; Infertility; Obesity; Orphan; Viagra	Cancer; Cosmetic; Infertility; Obesity; Orphan; Viagra
	Need	Cancer; Infertility; Obesity; Viagra	Cancer; Cosmetic; Infertility; Orphan; Smoking
	[No] Alternative	Cancer; Cosmetic; Infertility; Obesity	Cancer; Infertility; Orphan
	Range of Normality	Cosmetic; Infertility; Viagra	Cosmetic; Infertility
	Societal Functioning	Cosmetic; Infertility; Viagra	Cancer; Cosmetic; Infertility; Obesity; Orphan; Viagra
	Societal Responsibility	Smoking; Infertility	Infertility; Obesity; Smoking
	Vulnerability/Compassion	Cancer; Cosmetic; Orphan	Cancer; Cosmetic; Infertility; Orphan
Used in hypothetical decisions only	Dignity		Cancer; Obesity
	Equity/Fairness/Justice	Orphan	Cancer; Cosmetic; Infertility; Obesity; Orphan; Smoking
	Human Rights		Infertility
	Individual Cost	Smoking	Infertility; Orphan
	Moral Hazard		Infertility; Viagra
	Patient-Diagnosis		Viagra
	Rule of Rescue	Orphan	Cancer; Orphan
	Similar Treatments	Orphan	Infertility; Viagra
	Small Number of Patients	Cancer	Cancer; Orphan
	Societal Impact		Cancer; Infertility

Table 5 - Twenty argumentation sets organised per category, and the articles containing argumentations per treatment set per decision type of outcome (actual vs. hypothetical). 'Cancer' stands for cancer therapies, 'orphan' for orphan drugs, 'infertility' for infertility treatments, 'cosmetic' for cosmetic surgery, 'obesity' for obesity treatment and 'smoking' for smoking cessation therapy. NB: When a particular argumentation-outcome combination occurred only in one article, the cell is coloured light grey.

which are only used in hypothetical decisions. It consists of the following ten argumentation types: Dignity, Equity/Fairness/Justice, Human Rights, Individual Cost, Moral Hazard, Patient-Diagnosis, Rule of Rescue, Similar Treatments, Small Number of Patients, and Societal Impact. Furthermore, we observe that the first category contains the three most strongly acknowledged argumentation types, namely Morbidity/Severity (acknowledged thirteen times), (No) Alternative (eight times), and Need (seven times). In contrast, the second category holds the two most strongly questioned argumentations (that is, more than once), namely Small Number of Patients (six times) and Rule of Rescue (four times), though these were also as often acknowledged as valid (seven times and five times, respectively). Based on this analysis, we formulate the **third meta-level theory**: half of

Mechanism		Context				
	Argumentation type	Australia	Canada	USA	UK	France, Germany, Sweden, the Netherlands
Used in both actual and hypothetical decisions	Definition of Illness	-	4.9	19.2	5.6	13.5
	Individual Responsibility	-	-	5.8	1.1	-
	Medical Necessity	-	4.9	19.2	-	2.7
	Morbidity/Severity	25.0	19.5	19.2	28.1	16.2
	Need	-	7.3	5.8	7.9	18.9
	(No) Alternatives	25.0	12.2	5.8	7.9	16.2
	Range of Normality	-	-	7.7	16.9	5.4
	Societal Functioning	-	2.4	3.8	5.6	2.7
	Societal Responsibility	-	-	1.9	-	2.7
	Vulnerability/Compassion	16.7	12.2	3.8	4.5	10.8
Used in hypothetical decisions only	Dignity	-	2.4	-	-	2.7
	Equity/Fairness/Justice	-	7.3	5.8	13.5	-
	Individual Cost	-	-	-	-	2.7
	Moral Hazard	-	-	1.9	-	-
	Rule of Rescue	25.0	12.2	-	1.1	-
	Similar Treatments	-	2.4	-	2.2	-
	Small Number of Patients	8.3	9.8	-	5.6	5.4
	Societal Impact	-	2.4	-	-	-
	Total number of argumentations	12	41	52	89	37

Table 6 - Percentages of use of argumentations per country (for which a minimum of five argumentations was found). The argumentations are shown in alphabetical order per category. Human Rights and Patient-Diagnosis are absent as they were not used in country-specific argumentations.

the argumentation types are used by policy makers and insurance companies as well as patients, authors, the public, and the media, whereas the other half are only used by the latter group. The argumentation types that are used by policy makers and insurance companies are, moreover, more frequently acknowledged as valid, whereas some that are only used by patients, authors, the public, and the media are strongly questioned.

In an effort to understand whether and how the geographical location (the second context that we observed the impact of) affected the use of argumentation types, we plotted the usage frequency per country in Table 6. We identified the following parallels between the countries. From the first necessity category (the upper half of Table 6, categorised by use as described above), most countries employ most argumentation types at some point, and all countries employ the argumentation types Morbidity/Severity, No Alternatives, and Vulnerability/Compassion. In contrast, Medical Necessity is constricted to the USA, and Need is most frequently visible for 'mainland Europe' (France, Germany, Sweden, and the Netherlands). For the second necessity category (the lower half of Table 6), the pattern is much less congruent. Canada and Australia have the highest percentage of use

of argumentations grouped into this category. Moreover, only these countries utilise the more controversial argumentation types, namely Rule of Rescue and Small Number of Patients, whereas this is not the case for the other countries. This is to be expected, as the Rule of Rescue is part of the official criteria of the PBAC (cf. Cookson et al., 2008). In addition, the UK has a high percentage of the Equity/Fairness/Justice argumentation type, which is primarily due to the concern and outrage about 'postcode rationing' (Breuning et al., 2010; Goodson et al., 2011; Henderson, 2009; Mukherjee et al., 2014; Stevens et al., 2015; Wraight et al., 2007).

Accordingly, we formulated the **fourth and final meta-level theory**, which holds that the argumentations that fall under the first necessity category appear to be applied in most of the (predominantly Western) countries the dataset reports on, though the countries do appear to have their own argumentation type preferences. For the second category the patterns are much more varied: many argumentation types have a specific pattern (that is, used in two or three countries), which we conclude as depending on the local context.

DISCUSSION

In this chapter we unpack the question of necessity of coverage by reviewing the argumentations used and proposed to be used in coverage decision making. In seven treatment sets (cancer therapies, orphan drugs, infertility treatments, Viagra, cosmetic surgery, obesity treatment, and smoking cessation therapy) from 98 reviewed articles, we identified twenty different argumentation types employed to argue for or against the necessity of coverage of a certain therapy or treatment.

The argumentation types may be typified by primary decision outcome. The following eight argumentation types are used primarily in favour of coverage: equity, fairness and justice, a patient's societal functioning being impeded, the cost being too high to bear for an individual, the coverage status of similar treatments, considerations concerning human rights and dignity of the patient, and wider societal impact. Reversely, limited medical necessity, the individual being held responsible, potential moral hazard or over-usage, a small patient population, and a doctor needing to rely on the patient to set the diagnosis are argumentations solely employed to decrease the necessity of coverage. Finally, the morbidity experienced or the severity of the disease, whether the ailment falls within a defined 'range of normality' or is considered an illness, compassion for vulnerable patients, the need of the patient, the (un)availability of alternatives, society having to take responsibility for coverage, and the rule of rescue are used both in favour of and opposing coverage. The most important context affecting the argumentations in use in coverage decisions is the way the decision has come onto the agenda: when this is recorded in the article to be by a patient, a patient organisation, or the public, this always facilitates a positive decision, even when the argumentation type employed is also in use in negative coverage decisions. Others have stressed much the same point (Booth et al., 2007); in a cross-country comparison of hepatitis C coverage, Kieslich et al. describe experiences that are "as much a tale of challenges that arise when making difficult prioritization

decisions as they are a tale of agenda-setting" (Kieslich et al., 2016). Overall, it is important to note that no wide consensus is needed to let a consideration function as an argumentation in coverage decisions. This makes it doubly interesting to investigate what exactly happens during the processes of coverage decision making.

Necessity is also broadly categorised by two different types of usage. Under the first necessity category, indicating use in both actual and hypothetical decisions, fall the definition of illness and range of normality, the individual's own responsibility, the medical necessity, the morbidity (or the severity of the disease) and need experienced, the presence or absence of alternatives, the societal functioning of the patient, the societal responsibility for coverage, and compassion towards vulnerable patients. The second necessity category is not used in actual coverage decisions, that is, by policy makers and insurance companies but rather only by patients, authors, the public, and the media. For this second necessity category, we observed dignity, equity, fairness and justice considerations, human rights, the individual cost of the treatment, moral hazard, whether the patient sets the diagnosis him- or herself, the rule of rescue, coverage of similar treatments, the size of the patient population, and the societal impact of coverage. Interestingly, the use of the argumentation types that fall under this category is more localised, that is, country-specific, and more frequently debated. The argumentation types belonging to the first necessity category, however, are much more universally applied.

Comparison with existing literature

Our search string yielded seven studies containing literature reviews that were included in our review (Aggarwal et al., 2014; Drummond et al., 2014; Mladovsky & Sorenson, 2010; Nguyen et al., 2008; Paulden et al., 2015; Rocchi et al., 2008; Zelei et al., 2016), as well as a number of reviews that specified general qualitative criteria for coverage decisions. Cromwell, Peacock, and Mitton have conducted a literature review of 'real-world' decision criteria as found in 33 articles in peer-reviewed and grey literature (Cromwell et al., 2015). They searched for articles with an explicit acknowledgement of a "finite resource pool." Therefore, their review focuses on resource allocation, i.e., choosing one thing over another, in different settings such as hospitals and health authorities. They found that disease impact (burden) was an influential criterion in resource allocation decisions but did not observe other necessity-based argumentations.

Several other studies, e.g., Cerri et al., examined technology appraisals as conducted by macro-level institutions and commonly find it to be a "complex process involving numerous clinical, disease, and affordability considerations" (Cerri et al., 2014). Shah examined popular preferences in using severity of illness in economic evaluation as part of priority setting, showing that "people are, on the whole, willing to sacrifice aggregate health in order to give priority to the severely ill," which may further legitimise the use of individual burden of disease as a coverage criterion (Shah et al., 2014). Fischer conducted a broad review of decisions where quantitative methods were employed and identified several clinical criteria that strengthen our conclusions: availability of treatment alternative; condition is life threatening; condition caused by patients own behaviour; and end

of life considerations (Fischer, 2012). Hasman, McIntosh, and Hope, finally, interviewed decision makers on what they considered relevant in a coverage decision on a hypothetical drug (Hasman et al., 2008). The interviews highlighted agreement over cost effectiveness, clinical effectiveness, equality, and gross cost as important reasons for coverage. Interestingly, a lack of agreement was elicited over using 'absence of alternative treatments' as a criterion. Our study, rather than aiming to outline general criteria in use in coverage decisions, specifically gives an analysis of the tonal variety in the use of necessity in actual and hypothetical coverage decisions, and the effect contexts may have on the final coverage decision.

Strengths, limitations, and future research directions

To study, like we have, the way contexts influence coverage decision outcomes through the use of necessity as a criterion will aid our understanding of both coverage decision making as well as necessity as a concept as used in practice. In addition, the wide variety of argumentations described will aid further ethical reflection on the content and processes of actual and hypothetical coverage decisions.

Our search strategy in particular provided a challenge, as when one searches for "need" in the title/abstract, the articles that will be found are likely to discuss coverage decisions that use "need" as a criterion. As such, certain necessity-based argumentations were in that sense already to be expected from the wording of the search strategy. We have engaged two expert librarians to aid us in order to widen and strengthen our search terms and have thus acquired a relatively 'broad but clean' dataset.

The use of the realist review method has sharpened our review by forcing us to both broaden our choice in articles (opinion pieces, ethical and economic analyses, and reviews were analysed on top of document analyses and interviews/surveys) and take the defined contexts, rather than the practical set-up of the research, as primary quality attribute for inclusion and scoring. Although not always easy to accomplish, this study shows that the realist review method may successfully be applied to a wider variety of interventions than classical policy interventions such as patient portals (Otte-Trojel et al., 2014) or internet-based medical education (Wong et al., 2010). Necessity as a criterion is a policy 'product' that interacts with a variety of contexts to produce certain outcomes, and to frame our question thus has elicited exciting data on the varied use of necessity as a criterion in coverage decisions from peer-reviewed literature. It must, however, also be stressed that we have not effectuated a different 'weighing' of actual versus hypothetical decisions and that ethical discussions and public attitude surveys are of a wholly different substance than coverage decisions made by policy makers as they directly affect patients' lives. The inclusion of hypothetical decisions has not been self-evident, but it has been crucial to giving an overview of as wide a selection of criteria-in-action as possible.

It is, however, debatable to what extent we have grasped the full context of these criteria-in-action due to the way 'context' has been defined in this study. For example, on the micro level in Sweden, despite the national-level guidance not to treat smokers differently, "physicians are more inclined to

treat a non-smoking patient" (Björk et al., 2015). The element of weighing personal responsibility in coverage decisions is visible here but rarely expressed as such in health policy (Tinghög et al., 2010). This shows that criteria may be exhibited in other argumentations at the bedside (e.g., (Breuning et al., 2010; Rooshenas et al., 2015) or in court (e.g., (Giacomini, 2005; McIver & Ham, 2000). We have also omitted the characteristics of the exact form of care under consideration as being potentially influential, insofar as they were not described as part of the argumentations (cf. Lowi in (Kieslich et al., 2016). Additionally, one could argue that the way contexts have been defined in this study obscures the role of the decision maker or the structure of the decision-making process, as these also have not been considered as 'contexts'. Furthermore, the 'agenda setting' context is a short consideration of decisions that may have been made for political, rather than explicitly patient-focused, reasons (Gordon, 2006; Kroneman & de Jong, 2015; Specogna, 2010). Lastly, the data contained few articles on non-Western countries, which, upon becoming available, may well nuance the homogeneity in use of necessity considerations between countries. This, as well as additional research on the contexts mentioned earlier, may prove fruitful lines of further inquiry.

Finally, it would appear that using the realist review method predisposes one to think in terms of causality: a certain mechanism in a certain context leads to a certain outcome; argumentation type M, when used in country C leads to (hypothetical or actual) inclusion of treatment T. This, in itself, is problematic: the fact that a combination occurred 'out there' does not necessarily mean it *needed* to happen that way; it underplays the agency of the decision makers in this important sense. It also assumes that the outcome 'comes last'; it leaves no space for (additional) justifications that may have been brought in after the decision was made. Further investigation, for example through an ethnography, of the coverage decision making process and the role and use of necessity as a criterion in coverage decisions would be a useful exercise to shed more light hereon.

CONCLUSION

Necessity as a criterion in coverage decisions has lacked operationalisation: this chapter provides a handle on the wide variety of argumentation types that fall under this umbrella term by reviewing over 400 argumentations described in 98 peer-reviewed articles. These argumentations are grouped into twenty different necessity-based argumentation types that are used in coverage decisions, both 'actual' decisions (made by policy makers and insurance companies) as well as 'hypothetical' decisions (made by actors such as patients, authors, the public, and the media).

Eight of these twenty necessity-based argumentations are used to support both positive and negative coverage decisions; twelve are, however, only employed for either positive or negative decisions. When patients or the public were recorded to have aided in setting the decision on the agenda this always resulted in a positive decision, even when an argumentation type was used that could have resulted in a negative decision.

The argumentation types heightening or lowering necessity of coverage of a certain form of care may also be distinguished by their users. Half of the argumentation types is used in both actual and hypothetical coverage decisions, that is, by policy makers and insurance companies as well as patients, authors, the public, and the media. The second half is, in contrast, only used by the latter group. Argumentation types used in actual as well as hypothetical decisions are more strongly acknowledged and used in a greater number of countries. Conversely, argumentation types used in hypothetical decisions only are much more strongly questioned, and their use is much more country specific.

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