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Good conduct in a context of rationing: A case study of how frontline professionals deal with distributive dilemmas of novel gene therapies

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Abstract

Classical dilemmas of how to distribute limited resources have been rekindled by the rise of advanced, high-cost therapies. Building on a case study of a novel gene therapy in neuropaediatric care, this article explores the dilemmas that explicit priority setting can create for frontline professionals and develops a typology of professionals' responses to these dilemmas. Despite political attempts to centralise priority setting and spare health professionals from having to consider treatment costs at the 'bedside', this study shows that concern for economic efficiency and budget control nonetheless need to be handled and balanced against other accountabilities in the daily work of frontline professionals. Contributing to the sociological debate on priority setting and rationing, this study develops an analytical perspective attuned to the relational aspects of frontline work and the challenges related to the balancing of diverging ideas of good conduct. Further, focussing on an empirical field at the forefront of genomic medicine, this study brings the sociological debate on

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priority setting and rationing up to date with current developments in precision medicine.

KEYWORDS
accountability, doctor-patient relationships, frontline professionals, gene therapy, priority setting, rationing, resource allocation

INTRODUCTION

Since the 1990s, sociological studies have engaged with the regulatory ideal that health-care rationing should be guided by explicit priority setting (Hauge et al., 2022). Informed by a vast literature in bioethics and health economics (Brookes et al., 2015; Cookson & Dolan, 2000; Daniels & Sabin, 2006[1997], 2008), the regulatory aim of explicit priority setting is to ensure objective and transparent decision-making through the application of formal criteria and standardised procedures. This is intended to overcome the alleged idiosyncrasies of localised and informal practices of rationing conducted by frontline professionals, often referred to as 'implicit rationing' or 'bedside rationing' (Lauridsen, 2009; Oei, 2016).

Sociological studies of priority setting and rationing do not start by privileging certain understandings of what constitutes an 'optimal' or 'fair' basis for resource allocation. Rather, priority setting and rationing is analysed as the outcome of situated negotiations among social groups (Light & Hughes, 2001). The scientific evidence that informs priority setting is often rife with uncertainty (Calnan et al., 2017; Hughes & Doheny, 2019; Moreira, 2011), and various actors may value treatment options differently (Wadmann & Hauge, 2021). Therefore, priority setting is typically not a straightforward application of decision criteria (Russell & Greenhalgh, 2014; Sjögren, 2008; Vifell & Sjögren, 2011). In practice, technical questions and political processes are often intertwined (Hunter, 1995; Moreira, 2011). By investigating how policy-makers, managers, health professionals and patients interact as they seek to justify or challenge allocative decisions (Garpenby & Nedlund, 2016; Moreira, 2005, 2011; Syrett, 2003), sociological studies of rationing demonstrate that economic transactions are embedded in social relations that are also guided by other forms of reasoning than calculative rationality, such as political discourse, social negotiation of eligibility criteria and practical problem-solving (Light & Hughes, 2001; Vassy, 2001).

In the sociological debate on priority setting and rationing, much discussion has centred on the advantages and disadvantages of centralised versus local decision-making (see Hauge et al., 2022). Yet, the dilemmas that can arise for frontline workers in relation to explicit priority setting have only received limited attention (e.g., Calnan et al., 2017; Carlsen & Norheim, 2005; Hedgecoe, 2006). When centralised priority setting implies that services are withheld or suspended from patients who request treatment, frontline professionals can be put in demanding situations where they need to balance concerns for economic efficiency and budget control against other ideas of good conduct (Angell, 1993; Hurst et al., 2006). We need to understand how frontline professionals handle the conflicting expectations of their accountability relations because the implications of centralised policies ultimately depend on the routines and responses developed at the frontline of public service provision, as Lipsky (1980) famously argued. This article investigates how the introduction of explicit priority setting can alter the accountability relations of frontline professionals, which tensions this gives rise to in their daily work and how they deal with these tensions.

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Building on a case study of novel gene therapy in neuropaediatric care in Denmark, this study makes three main contributions to the sociological literature on priority setting and rationing. First, by bringing this literature into conversation with studies of professional accountabilities and frontline work, we offer a new, relational perspective on how centralised priority setting affects professional work 'at the bedside'. Second, by developing a typology of professional responses to competing accountabilities, the study provides an analytical stepping stone for future research. While existing studies have provided examples of how frontline professionals seek to challenge budgetary limits and allocative decisions (e.g., Lapsley & Melia, 2001; Russell & Greenhalgh, 2014), the sociological literature on priority setting and rationing has yet to develop shared conceptualisations of how frontline professionals deal with distributive dilemmas. Third, the study brings the sociological debate on priority setting and rationing up to date with current developments in the medical field. Using an empirical case from the forefront of genomic medicine, the study identifies challenges that are likely to accompany the rise of advanced, high-cost therapies in precision medicine.

A SOCIOLOGICAL PERSPECTIVE ON PROFESSIONAL ACCOUNTABILITIES

Historically, the notion 'to account' was related to accounting in the literal sense of bookkeeping (Bovens, 2006). In contemporary social science, however, the concept of accountability has gained a broader meaning and refers to relationships that involve the giving and demanding of reasons for conduct (Laughlin, 1996, p. 225). Hence, accountability is about being answerable to some other who has a legitimate reason to ask for accounts. Accountabilities can be more or less formalised (Bovens, 2006). Yakel notes that accountability is 'more than formal reporting to a higher authority' as it can involve expectations of performance, responsiveness and morality to a broad range of people and institutions (2001, p. 234). Frontline professionals orient themselves not only hierarchically but also evaluate their work in relation to professional codes and their ability to help the individuals in front of them (Maynard-Moody & Musheno, 2000). In the words of Ewert, clinicians are required to be 'jacks-of-all-trade' (2020, p. 308) in navigating multiple accountability relationships where different conceptions of good conduct prevail, including loyalty towards colleagues, empathy in patient encounters, compliance with regulatory standards and economic efficiency. Failure to meet expectations can have different types of consequences, ranging from collaboration difficulties to formal sanctions (Hupe & Hill, 2007). At times, the concerns and ideas of good conduct associated with different accountabilities can appear incompatible or difficult to reconcile (Ewert, 2020), spurring reflection on what it means to be a 'good' professional (Navne & Svendsen, 2017). Often, it has been suggested frontline professionals may experience conflict because they are expected to act as both 'state-agents' and 'citizen-agents' (Maynard-Moody & Musheno, 2000) or because 'core' professional values and modes of reasoning are threatened by managerial values or economic rationales (e.g., Abernethy & Stoelwinder, 1995; Jackson et al., 2014; Lapsley, 2009). However, rather than presuming a binary opposition between 'managerial' and 'professional' values and rationales, we follow a recent strand of studies in the sociology of accounting (e.g., Gebreiter, 2022; Schrøder et al., 2022; Sjögren & Fernler, 2019) and explore how the explication of economic priorities may reconfigure professional work.

Frontline professionals' attempts at dealing with competing accountabilities are often analysed as 'coping strategies' (Tummers et al., 2015). As a psychological mechanism, the notion

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of coping focuses on cognitive or behavioural responses in individuals who experience conflict between external demands and internal values (Lazarus & Folkman, 1984). However, the notion of coping overlooks that attempts at navigating competing accountabilities do not happen in a social vacuum. Health professionals operate in a context where their treatment decisions are made in relation to both proximate and distant others (Navne & Svendsen, 2017; Timmermans & Mauck, 2005). Moreover, attempts at dealing with competing accountabilities may not be limited to cognitive or behavioural reactions but can also include new ways of organising work (Schrøder et al., 2022). Therefore, we regard the responses of frontline professionals to competing accountabilities as including not only their individual cognitive and behavioural reactions but also the social interactions and organisational efforts they engage in to reduce potential conflict. Such responses are not necessarily strategic and calculated but may be more or less improvised (Gebreiter & Hidayah, 2019).

Various typologies of professional responses to competing accountabilities have been developed (Gebreiter & Hidayah, 2019; Numerato et al., 2012; Pache & Santos, 2013; Tummers et al., 2015). While these typologies apply varying conceptualisations and framings, three overall types of professional responses can be synthesised. *First*, competing accountabilities may be dealt with by prioritising some ideas of good conduct over others. Pache and Santos (2013) and Gebreiter and Hidayah (2019) suggest that professionals may 'subscribe fully' to some ideas of good conduct and actively 'refuse' others (either due to moral commitment or fear of sanctions), implying that accountabilities are ordered hierarchically. Similarly, Tummers et al. (2015) distinguish between strategies that make professionals 'move closer' to either authorities (e.g. by expressing cynicism or aggression towards clients) or to the individuals in front of them (e.g. by bending or breaking rules). *Second*, conflict between competing accountabilities may be dealt with through 'compartmentalisation', that is, spatial or temporal separation of interactions where different expectations of good conduct prevail (Gebreiter & Hidayah, 2019; Pache & Santos, 2013). *Third*, attempts can be made to align various accountabilities (Pache & Santos, 2013). This can happen in various ways. For example, through 'translations' that are meant to align diverging ideas of good conduct (Gebreiter & Hidayah, 2019) or through 'co-optation', which refers to professionals' attempts at using regulatory techniques and rationales strategically to encompass other ideas of good conduct (Numerato et al., 2012). This study builds upon this synthesis of existing typologies and extends the existing work by identifying additional ways in which professionals seek to align various accountabilities.

STUDYING EXPLICIT PRIORITY SETTING IN DANISH NEUROPEDIATRIC CARE

Case description

Danish health professionals have played a prominent role in government attempts to ensure a fair and efficient distribution of public resources since the 1930s (Wadmann & Pedersen, 2020). However, only since 2017, with the establishment of the Danish Medicines Council (DMC) has economic priority setting in specialised care been made explicit. Like other national health technology assessment (HTA) agencies, the DMC holds a political mandate to include economic concerns in its recommendations about the use of new treatments. To explore how professional accountabilities in this setting may be altered by explicit priority setting, we examine the rationing of a novel, high-cost pharmacogenetic therapy, Spinraza (nusinersen), launched in 2017 by

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Biogen. Spinraza was the first active treatment for the rare disease 5q spinal muscular atrophy (SMA). SMA affects the nerves in the spinal cord and causes progressive muscle degeneration. For the most severe types, patients die before the age of two because they become too weak to breathe or eat. In the mildest cases, patients can expect a normal lifespan, but deteriorating muscle strength limits everyday activities and occupational choices (Wang et al., 2007). Spinraza is intended to stop disease progression by targeting the genetic mechanism that causes muscle degeneration. When launched, Spinraza was known as the world's most expensive pharmaceutical. Based on official list prices in Denmark in 2017, the incremental treatment cost per patient was DKK 4.4 million (EUR 580,000) for a 15-month period (Amgros, 2017). Moreover, documentation of its clinical effectiveness was marked by great uncertainty. In 2017, based on a cost-effectiveness evaluation, the DMC issued its first guidelines on Spinraza, stipulating that only a sub-group of the youngest and most severely affected patients were treated with Spinraza (DMC, 2017). While a re-evaluation in 2018 led to a second set of guidelines for Spinraza that expanded the treatment-eligible population, treatment access remains restricted (DMC, 2018).

This case is illustrative for at least three reasons. *First*, with Spinraza, neuropaediatrics became a test bed for explicit priority setting in Denmark, as health economic rationales were used explicitly to limit access to a novel, specialised therapy. *Second*, health professionals were urged by multiple stakeholders to account for their treatment decisions, providing an opportunity for an empirical study of how competing accountabilities were dealt with in practice. *Third*, the case allows us to explore dilemmas that are likely to become more pressing in the near future, as neuropaediatrics is at the forefront of a growing field of advanced, high-cost therapies targeted at small patient populations. Horizon scans predict a substantial rise in the number of gene and cell therapies to be marketed from 2020 to 2030 (Quinn et al., 2019; Ronco et al., 2021).

Methodological and ethical considerations

Studying the treatment of rare diseases in a small country like Denmark involves both methodological and ethical challenges. By definition, rare diseases involve a small number of patients and, consequently, a small number of professionals, which reduces the number of potential informants and poses challenges to anonymisation. To address these challenges, we included all three treatment sites in Denmark, together serving a national population of about 200 patients with SMA. While several professions are involved in the diagnosis, treatment and monitoring of SMA, physicians and physiotherapists have specific responsibilities pertaining to decisions about the withholding or withdrawal of treatment. Table 1 provides an overview of the empirical material used in this article.

Our understanding of priority setting in Denmark is also informed by over 20 interviews with hospital managers, representatives from the DMC and families of SMA patients, which serve as background information in this study. We discussed the challenges of anonymisation with informants and agreed on which identifying information to omit. Further, a draft manuscript was sent to informants for approval of quotations and 'member checking' of the emergent analysis (cf. Birt et al., 2016). Consequently, we left out one empirical example that was considered too sensitive by an informant. The study was approved by the Human Subject Protection Review Board at The Danish Center for Social Science Research (VIVE).

The analysis builds on a process of analytical abduction that moves dialectically between empirical material and theoretical abstractions (Tavory & Timmermans, 2014). An initial open coding alerted us to the frontline professionals' efforts of maintaining relationships with many

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TABLE 1 Overview of empirical material gathered from May 2017 to June 2019

Sources	Materials
Semi-structured interviews	- 5 neuropediatricians[a] - 7 physiotherapists involved in monitoring of SMA patients - 1 clinical manager
Observations	- 5 consultations for SMA patients - 1 meeting with physicians and nurses where the implementation of DMC guidelines were - 1 network meeting for neuropediatricians from all treatment sites - 3 public meetings on medical priority setting
Documents	- Guidelines on the use of Spinraza published by the DMC - Parliamentary white paper on medical priority setting - Procedure for 'exceptional' treatment access issued by the regional authorities - Standard letters of rejection - Debate articles on Spinraza and priority setting published in national media

[a]A follow-up interview was made with one of the neuropediatricians in 2022.

parties from parents to managers. We noted that the explication of costs (only) sometimes gave rise to conflict and started to compare these situations systematically. Concurrently, we engaged in a process of 'casing' (Tavory & Timmermans, 2014) to explore possible conceptualisations through a series of workshops. Sociological studies of accounting and professional accountabilities propelled us to analyse the implications of explicit priority setting as reconfigurations of professional accountabilities and responses to competing concerns.

ANALYSIS: RECONFIGURATION OF ACCOUNTABILITIES IN NEUROPEDIATRIC CARE

While critical decisions on the withholding or withdrawal of treatment are an integral part of paediatric care (Navne & Svendsen, 2017; Svendsen, 2022), the explicit use of economic arguments to limit access to treatment constitutes what Mattingly calls a 'disruption of routines' (2012, p. 5). In our case, the efforts of frontline professionals to account for their decisions were challenged in at least three ways.

First, the ideas of good conduct expressed in hierarchical accountability relations became more ambiguous. On the one hand, the professionals were expected to 'follow suit': economic incentives were established by the regional authorities that made hospitals bear the treatment costs if frontline professionals did not comply with treatment guidelines from the DMC. Thus, the discretionary power of the professionals was delimited. On the other hand, professionals' ability to exercise discretion was reinforced. The Danish Parliament's white paper on medical priority setting (adopted unanimously just before Spinraza was evaluated by the DMC) stipulates that physicians should be able to prescribe therapies based on 'a medical assessment of individual cases', even if the therapies were not recommended for clinical use by the DMC (Danish Ministry of Health, 2016). The regional authorities, who bear political and administrative responsibility for the provision and financing of health care, translated the principle of the white paper into procedures to evaluate the 'exceptionality' of individual patient cases. These 'exceptionality procedures' can allow treatment for patients who do not meet the formal eligibility criteria if physicians can argue that a case is 'exceptional' and this argument is accepted by the Region's pharmaceutical committee (Danish Regions, 2018).

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Second, the importance of ensuring alignment of treatment strategies with professional peers was strengthened. As treatment decisions became subject to public scrutiny, variation in the reasons given for decisions became a potential problem because it could be used in allegations of unfair discrimination and could compromise welfare state ideals of fairness and impartiality. A physician likened the absence of common treatment standards to 'a cowboy country where everyone minds their own business' and explained: 'We are a small country, and it doesn't work well for our country as a whole if we don't follow the same standards; it doesn't work for us as professionals, and overall, patients will be worse off' (Interview, June 2019). Similarly, another physician explained, 'I may disagree with the criteria [in national guidelines] but I don't want to undermine our shared standpoint in Denmark' (Interview, June 2019).

Third, the professionals' relationship with patients and relatives was put under strain. One physician described how explaining to parents that treatment for their child had to be withheld due to uncertain treatment cost-effectiveness was a 'huge dilemma' (Interview, December 2018). Another explained: 'Some of the parents get angry with me because I keep telling them "no"' (Interview, June 2019). Hence, while explicit priority setting is often framed as a way of overcoming the challenges of 'bedside rationing', it can also add new complexity to frontline work. In this case, a number of tensions arose as expectations of good conduct in hierarchical relations became more ambiguous, closer coordination was required in peer-relationships, and the professionals' relationships with patients and relatives became more prone to conflict.

TENSIONS BETWEEN COMPETING IDEAS OF GOOD CONDUCT

Figure 1 illustrates the many ideas of good conduct facing frontline professionals. In the following, we outline how these ideas collided in their everyday work.

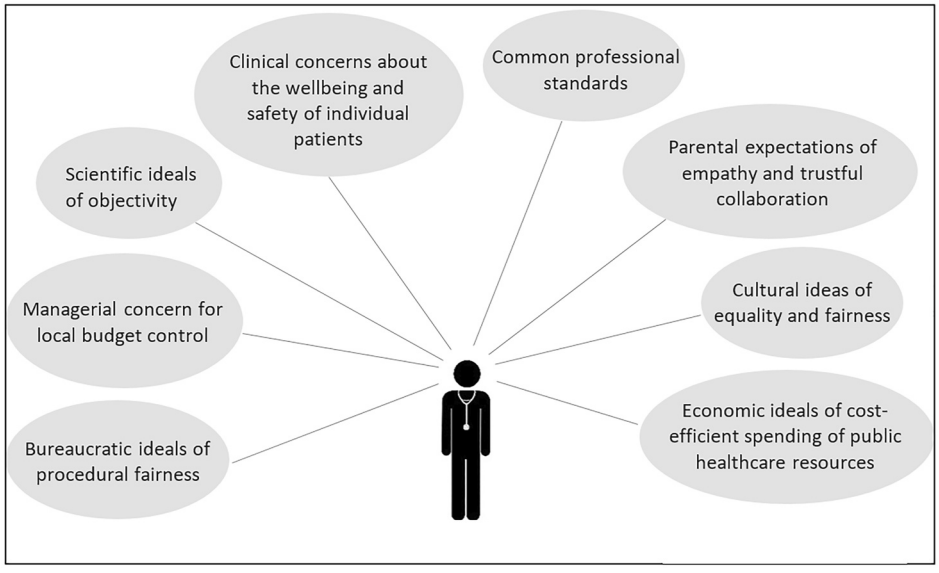


FIGURE 1 Competing ideas of good conduct that frontline professionals are to navigate

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Hierarchical accountabilities collide with accountability to patients and relatives

In several instances, the professionals found themselves split between the expectations of good conduct expressed in the DMC guidelines on Spinraza and the ideas of good and responsible care that prevailed in their interactions with SMA patients and their families. We illustrate this through three examples.

The first example centres on how the DMC guidelines sometimes conflicted with clinical evaluations of disease severity. The first guideline on Spinraza issued by the DMC required physicians to distinguish clearly between patients with two and three copies of the survival of motor neuron (SMN-2) gene, which is associated with disease severity (Calucho et al., 2018). However, this genetic marker of difference did not correspond to clinical experiences of severity: 'The condition can be just as severe, clinically speaking, if you have three SMN-2 copies [compared to two]', a physician explained and continued:

> We were put in a situation where we could imagine two patients [and their parents] sitting in the waiting room with the same condition, the same severity and the same burning hope of treatment. One of them, we would have to tell *no*; the other we could tell *yes*. So, it was really difficult to handle this decision in practice.
>
> (Interview, December 2018)

Hence, the DMC guidelines—that were meant to ensure procedural fairness and cost-efficient spending of public resources—required the physicians to differentiate the treatment of patients who were considered similar from a clinical point of view. It was a dilemma: physicians contested the validity of the formal eligibility criterion, yet if they went against the DMC guidelines, they would be in conflict with expectations of budget control expressed by superiors. In the words of a clinical manager, the professionals were well aware that they would 'bury their own department if they prescribe expensive treatments without coverage [from the Region]' because pressure would be put on staff resources if budgetary limits were exceeded (Interview, December 2018). Thus, tension arose between concerns for procedural fairness and cost-efficiency, local budget control and clinical perceptions of patient needs.

In a second example, we saw how the DMC guidelines did not fully correspond with clinical perceptions of treatment benefit. The DMC guidelines on Spinraza require that the treatment eligibility of children is evaluated regularly and that treatment should be stopped if the respiratory function or the motor function of the children deteriorate for a prolonged period (DMC, 2017, 2018). The professionals endorsed the need for standardised criteria and metrics when evaluating treatment benefits to avoid being 'misled by snapshots', as one physician expressed it (Interview, December 2018). However, the metric that was used in the evaluation of treatment effects (the Hammersmith scale) does not capture all aspects of treatment benefit that were considered important by the families of SMA patients and the professionals. 'There is much more to it than just those items we test', one of the physiotherapists noted, 'if they have the energy to attend kindergarten or stay awake a bit longer and play with friends, this is also progress and improved quality of life for these children' (Interview, May 2019). Furthermore, the professionals expressed concern that the tests might discriminate against children who were

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wheelchair users because the metric focus primarily on the ability of patients to stand or walk. A physician explained:

> The fine motor skills are extremely important for the children who use wheelchairs. Many of the items in the Hammersmith scale do not capture what's most decisive to them. It's important what they can do with their fingers. Their ability to control their wheelchair independently, do their hair or help themselves at the toilet. [...] That's probably more important for their quality of life.
>
> (Interview, December 2018)

As a validated scale to measure the functional ability of people with SMA, the Hammersmith scale is endowed with scientific credibility associated with test reliability and objectivity. Yet, from a clinical perspective, it was also regarded as a crude measure of treatment benefit with potential discriminatory effects. Hence, tension arose between concern for objectivity (associated both with scientific ideals of disinterestedness and bureaucratic ideals of impartiality) and concern for the everyday experience of individual patients, a dilemma that is also well-described in relation to clinical guidelines and other initiatives of evidence-based medicine (e.g. Timmermans & Mauck, 2005; Wadmann & Bang, 2015).

Our third example shows how the DMC guidelines sometimes challenged conceptions of fairness expressed in informal accountability relationships. This was clear in the case of two siblings with SMA. While one sibling clearly met the formal criteria for treatment, the eligibility of the other sibling was questionable because the disease was more advanced. Their parents reacted strongly and went straight to the press when they learnt that their children might be placed on either side of a dividing line that defined their treatment prospects. When discussing the case, the professionals deemed the differential treatment of the siblings unfair. Yet, deviation from the DMC guidelines was not a viable option due to budget concerns. Discussing the prospect of increasing numbers of high-cost therapies becoming available, the professionals expressed a need to 'respect the guidelines' to ensure the economic sustainability of their clinics and the overall health-care system (Fieldnotes, May 2018). Hence, tension arose between concern for cost-efficient spending of public resources, local budget control and culturally ingrained ideas of equity and fairness in kinship relations.

Conflict between accountability to individual patients and accountabilities to parents

While families are often thought of as a unit, the professionals sometimes experienced that their effort to act responsibly to the individual child brought them into conflict with parents. Some physicians explained that treatment with Spinraza might not always be in the best interest of the child, because the clinical effectiveness was not firmly established for all patient groups and the treatment involved some risks. Alluding to the fact that the medicine was to be injected directly into the spinal cavity, a physician explained, 'The child has to be jabbed repeatedly, and the medicine can have adverse effects such as high intracranial pressure' (Interview, February 2019). Others mentioned the risk of infections. When physicians lacked convincing trial results, the withholding of treatment was considered correct 'out of concern for the child, as well as for scientific integrity', a physician explained (Interview, February 2019). However, this was difficult for some parents to accept. One physician expressed concern that a focus on 'numbers' and

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'statistics' might be considered 'unempathetic' by the families because positive stories about Spinraza and its effects were circulating on social media. The physician explained:

> Much of what is said is not backed by science. And it's really difficult for the patients to understand what this means to us. We cannot just prescribe medicine for DKK 800,000 [EUR 108,000] per dose if we don't know whether it works.
>
> (Interview, December 2018)

Advanced therapies for rare diseases typically come with high pricing and considerable uncertainty about clinical effectiveness, making the need for economic priority setting more pressing from a health-care system perspective (Jönsson et al., 2019). However, considerations about cost-effectiveness may threaten the relationship between health-care professionals and families according to the physicians. One physician explained: 'What is crucially important in all of this, and what is completely new, is that I lose the parents.' The physician continued:

> I won't say I lose their trust completely, but [you lose] the positive role you have as the family's or the child's doctor during a chronic disease.
>
> (Interview, February 2019)

As parents are the primary caregivers of the child, parental collaboration is key to paediatric care. Therefore, eroding trust not only affected the 'role' of the professionals but also their ability to help the child. Hence, tension arose between concern for the safety of individual patients, parental collaboration, scientific integrity and cost-efficiency.

The tensions between competing expectations of good conduct prompted the professionals to reflect on what it means to act in a 'good' and 'responsible' manner. In the words of a physician, you have 'to find [your] way in all of this because you feel for the family and want to do the best for them. And now you suddenly find yourself in the dark. No one can instruct [you] on how to do this' (Interview, February 2019). In the following, we outline what the professionals did to find their way through 'the dark' and handle conflict between competing accountabilities.

PROFESSIONALS' RESPONSES TO COMPETING ACCOUNTABILITIES

Overall, the professionals sought to navigate either by *separating* the enactment of competing ideas of good conduct or *combining* these. In the following, we unfold these responses, which are summarised in Table 2.

Compartmentalisation

Various measures were taken that separated the enactment of competing accountabilities in time or space, akin to what has been called *compartmentalisation* (Gebreiter & Hidayah, 2019). Front-line professionals were instrumental in this process but they did not act on their own. Their push for new, formal procedures activated managers and others in the hospital organisations and resulted in the establishment of local procedures. A clinical manager explained:

> The difficulties arise in borderline cases or when patients fall outside the criteria defined by the DMC. In the beginning, we didn't have a clear procedure to handle these cases. This was established later. When clinicians want to treat patients who don't meet the criteria, they now file a request, which is then evaluated by the

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TABLE 2 Frontline professionals' responses to competing accountabilities

Responses	Description	Examples	Rationing turns into...	Risks
Compartmentalisation	Spatial or temporal separation of interactions where different ideas of good conduct prevail	- Distribution of decision-making - Delegation of tasks - Separation of communication channels	Impersonal case handling	Complaints about the lack of individual assessments
Translation	Reframing what the conflict is about	- Justifications made in direct communication - Self-reflection	A matter of professional ethics	Distrust in the motives of frontline professionals
Co-optation	Strategic use of regulatory techniques and ideas of good conduct to meet other concerns	- Introduction of new evaluation tools - 'Pro forma' use of procedures	Strategic moves to influence the regulatory framework	Uncertain impact on treatment decisions
Situated mediation	Mediation of regulatory demands and individual needs on a case-by-case basis	- Mobilisation of uncertainties in guidelines - Professional coordination of treatment decisions	Situated interpretation of eligibility criteria in relation to individual patient traits	Allegations of unfair discrimination

> clinical pharmacological unit. [...] Before this, it was really difficult to find out how to manage these cases because there were no procedures.
>
> (Interview, December 2018)

These new procedures distributed decision-making within the hospital organisations. To reduce conflict between physicians and families, the making of rationing decisions was separated from the clinical encounter. Letters signed by hospital managers–not individual physicians-were created to inform families if their children were deemed ineligible for treatment. In one clinic, oral communication of treatment decisions was also delegated from physicians to a clinical manager (Fieldnotes, September 2018). While the professionals did not discard the concern for economic efficiency and budget control, which was expected in hierarchical accountability relations, they sought to disentangle economic concerns from their direct interaction with the families. 'You need distance to think in economic terms', as a physician noted (Interview, December 2018).

The documentation of treatment benefit was also separated from the direct encounter between the families and the physiotherapists. During the tests (that were to inform decisions about treatment continuation or withdrawal), parents sometimes questioned whether the actual motor abilities of the child were reflected in the test situation, for example, by showing physiotherapist videos of the child. One physiotherapist also explained about an episode when parents started to negotiate the validity of the registered test results and noted: 'that's not how it's supposed to be'. In the informal encounters with parents, the physiotherapists would

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acknowledge the experience of the parents, for example, by encouraging them to save the videos and by counselling parents at length about how to stimulate their children in everyday life and access proper assistive devices. Yet, objectivity remained a key concern, and the physiotherapists developed 'scripts' to standardise test procedures and introduced various ways of documenting and validating test results after patient visits (Interview, June 2019). Acknowledging that even standardised tests do not preclude subjective assessment, the physiotherapists cross-validated their observations of children to make the scores 'as correct as possible', as one physiotherapist explained:

> Afterwards, we sit down and discuss. [...] You see, there's not a 100% clear answer to every test item. It might be pulled a bit in this direction or the other. We usually register very similar scores, and if we don't agree, then we discuss.
>
> (Interview, May 2019)

Others used video recordings: 'We don't really have the time, but we try to do it anyway - you know, repeat the rating based on the video' (Interview, June 2019). In striving to accommodate the needs of individual children and their families while also catering for objectivity, the physiotherapists established a spatial and temporal distance between the clinical encounter and the rating.

Fostering what Porter (1996) calls 'mechanical objectivity', the formalisation of procedures turned the evaluation of treatment eligibility into a matter of impersonal case handling that was done separately from the direct encounter between frontline professionals and the families. In this way, the professionals strived to maintain collaboration with parents while also living up to formal expectations of procedural fairness, objectivity and cost-efficiency expressed in hierarchical relationships. While these responses embedded difficult rationing decisions in organisational routines and made them easier to handle in everyday work, they did not eliminate the risk of conflict. Some parents drew on the national white paper on medical priority setting (Danish Ministry of Health, 2016) when making complaints that the standardisation of procedures deprived their children of the right to have an individual medical assessment (Skeem, 2020). Hence, the ambiguous notion of good conduct expressed in hierarchical relations was used by the parents to challenge the response of the frontline professionals.

Translation

To deal with the competing accountabilities, the frontline professionals also responded by reframing what the conflict was about. For example, this *translation* happened when physicians justified the withholding of treatment by casting it as a matter of best care for the child. One physician likened the treatment of children who did not meet the formal eligibility criteria to a type of experimentation:

> If the treatment has no effect, I will not let a disabled child – note this – I will not let a *disabled child* undergo experimental treatment if I personally don't believe it's effective for this child.
>
> (Interview, February 2019)

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As the physician continued, the question of access limitation was reframed as a matter of professional ethics rather than a matter of cost-effectiveness:

> To show my respect for the child [...], I try to state this in terms of ethics [...] without thinking about the costs. It's an exercise I've had to do for myself every day when I cycle home from work: Am I really certain about this?
>
> (Interview, February 2019)

To the physician, costs constituted an illegitimate basis for the justification of rationing decisions in encounters with patients and families. Likewise, a clinical manager stated that health professionals would 'give their right arm' to avoid declining a treatment request with reference to costs: 'You just can't [say this] to someone's face' (Interview, December 2018). By framing the withholding of treatment as a matter of respect for the child, the professionals sought to navigate the difficult situation by translating it into a question about 'good' SMA care that was cast in terms of patient respect instead of economic terms.

This response enabled the professionals to render their care for individual patients compatible with the formal guidelines that catered for the economic sustainability of the health-care system. Yet, it did not eliminate the risk of conflict, as parents could react with distrust:

> Before they saw you as their helper and guardian against the enemy, that is the DMC, now you are seen as someone who shifted loyalty and became an ally of the official system.
>
> (Interview with physician, February 2019)

While the translation enabled physicians to focus on the safety and wellbeing of the child 'without thinking about the costs', the motives of the physicians were questioned by parents, who brought up the issue of cost. 'Costs come to play a strange role,' a physician noted, and continued: 'The parents *keep* telling me: it's just because of the high costs. *This* is the reason you cannot allow it' (Interview, February 2019). According to this physician, parental collaboration suffered: 'They won't look me in the eyes. I can be honest with them, but they won't listen' (Interview, February 2019). Hence, the explication of health economic rationales at the national level instilled new forms of doubt in the interactions between frontline professionals and the families of SMA patients because it became visible for the parents that other concerns than the wellbeing of their individual child could be at stake.

Co-optation

A third type of response was one of *co-optation* (Numerato et al., 2012): regulatory techniques and rationales expressed in hierarchical relations were used strategically by frontline professionals to better encompass the ideas of good conduct that prevailed in informal relationships with peers and the families of SMA patients.

Physiotherapists identified and implemented additional measurement scales to make the regulatory standards more inclusive. To better capture any functional improvements of the children who were wheelchair users, two of the physiotherapists explained that they had convinced

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their superiors to supplement the Hammersmith scale with tests of fine motor skills, such as finger mobility:

> We thought it would be very relevant to add a test called RULM [the Revised Upper Limb Module] because the children might actually demonstrate progress on other parameters even if they stagnate on our tests or – in the worst case – start losing functional abilities again.
>
> (Interview, May 2019)

By expanding the range of parameters that could be used to evaluate treatment benefit, the physiotherapists acknowledged the concerns for objective measurement and procedural fairness that were expected in the relationships with superiors. However, they also used these rationales to encompass concerns for individual patient needs and subtly expand the opportunities for patients to stay on treatment.

In another instance, a physician used the Region's procedure for 'exceptionality' to demonstrate to patients and families that decisions about treatment eligibility were made elsewhere and not by the physician. 'You may call it a kind of activism', the physician pondered and explained:

> I applied for exceptional treatment access for all our SMA patients [who were not eligible for treatment according to the national guidelines] to the regional pharmaceutical committee. They turned them all down. [...] It was not that surprising. But I needed to levy some pressure on the system to make it clear where the decisions were made. Now I can tell the patients: I would like to give you the treatment, but I cannot – because somebody else said no.
>
> (Conversation with physician, August 2022)

Even though expecting a negative outcome, the physician still applied for 'exceptional' treatment access to illustrate the constraints on professional discretion and ease the collaboration with parents.

Neither the physiotherapists nor the physicians violated the regulatory standards. Instead, they sought to navigate competing concerns by using the regulatory techniques and rationales in slightly new ways.

Situated mediation

A fourth response from the professionals was to use the interpretive flexibility of the DMC guidelines to provide treatment options for individual patients on the verge of treatment eligibility. Again, the professionals were careful not to violate the formal expectation of good conduct expressed in hierarchical relations but engaged in situated interpretation of the formal eligibility criteria to mediate between national standards and individual patient traits. Hence, we call this response *situated mediation*.

Such mediation happened, for example, when physicians argued that some patients could not be classified according to the formal criteria defined in the DMC guidelines. A physician explained:

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> We don't treat anyone without permission. But sometimes exceptions can be made if you can argue that you're actually in doubt about which type [of SMA] it is. [...] Even though the criteria are meant to be very precise, it's difficult to be absolutely certain whether a patient fits the criteria or not.
>
> (Interview, June 2019)

Some of the formal eligibility criteria for Spinraza were modelled over a particular classification of SMA subtypes based on a combination of genetic and clinical traits. However, some children were difficult to place unequivocally within a given subtype, for example, because genetic and clinical markers of disease pointed in slightly different directions. The frontline professionals mobilised this diagnostic uncertainty to include borderline cases into the group of treatment-eligible patients. Similarly, uncertainties about the onset of disease were mobilised by physicians when patients bordered on formal eligibility criteria related to disease duration. For example, in the previously mentioned case of the two siblings, the physicians took advantage of the fact that 'treatment onset' was not clearly defined in the national guidelines, allowing them to count the disease duration in a way that qualified both children for treatment. Moreover, the interpretive flexibility on the criteria for respiratory function was mobilised by physicians to postpone the time of treatment withdrawal when 'the parents weren't ready' (Fieldnotes, August 2019). In the clinical encounter, ending treatment is not merely a question of adequate resource allocation, but also a matter of navigating liminal states between life and death (Anspach, 1993; Navne & Svendsen, 2017). Treatment withdrawal marked a transition from heroic efforts of saving a seriously ill child to acceptance of the child dying. Offering an 'extra dose' served to prolong this transition and literally buy the parents time while also 'sustaining the collaboration with the family' (Interview, December 2018).

So, even if the centralised priority setting constrained the discretionary power of frontline professionals, they also found ways of exercising agency. Through situated mediation between formal standards and individual patient traits, frontline professionals strive to meet informal expectations of empathy and individualised care in relations with families of SMA patients without compromising the concerns about efficiency and budget control expressed in hierarchical relations. This response reduced conflict with some families. However, it was not without risks, as other families might perceive it as unfair discrimination. A physician explained how a given case of 'exceptional' treatment access had 'generated considerable noise' in the SMA community:

> You know, it generated dissatisfaction over why other patients couldn't get access. It really did. [Parents asked:] How come? They're the same age, so why [are they treated differently]?
>
> (Interview, December 2018)

In the small and well-organised SMA community, inconsistencies in the application of eligibility criteria were quickly exposed, which enabled the families of other patients to challenge the fairness of these treatment decisions. Accordingly, the frontline professionals took measures to coordinate clinical decision-making more closely. For instance, neuropediatricians who were involved in SMA treatment created an e-mail list and established a national network to provide for ongoing communication about difficult treatment decisions and the interpretation of eligibility criteria across treatment sites. Hence, the situated mediation between national standards and individual patient needs did not happen in a social vacuum. The ongoing interaction among the SMA families fostered a need for tighter professional coordination (note also how the physiotherapists emphasised the need for collegial coordination, see the section 'Compartmentalisation').

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DISCUSSION AND CONCLUSION

The classical dilemma of how to distribute limited health-care resources has been revitalised by the development of advanced, high-cost therapies, underlining the need for sociological debate about priority setting and rationing. The present study contributes to this debate in three ways.

First, the study links the sociological debate on priority setting and rationing with literature on professional accountabilities and frontline work. By analysing the implications of explicit priority setting for frontline work as a question of how professional accountabilities are reconfigured, we shed light on the relational aspects of frontline work, the dilemmas related to balancing competing ideas of good conduct and how the frontline professionals respond to these dilemmas. While sociological debate on priority setting and rationing has traditionally been occupied with discussion of pitfalls and possibilities related to centralised priority setting *versus* local rationing decisions (see Hauge et al., 2022), recent studies show that the centralisation of allocative decisions does not alleviate the need for professional judgement (Hedgecoe, 2006; Hughes & Doheny, 2011; Russell & Greenhalgh, 2014). Advancing this view, the analytical lens developed in this study invites attention to the *interplay* of centralised and local decision-making. For example, it allowed us to show how an ambiguous regulatory framework that both restricts and encourages professional discretion in rationing decisions gave rise to professional attempts to push responsibility up the hierarchical system. Meanwhile, the families of SMA patients could mobilise the ambiguities in the regulatory framework to challenge local clinical assessments. The present study also broadens the scope of contemporary sociological discussions on health-care rationing. In contemporary debates, much emphasis has been given to the ways in which epistemic uncertainties are dealt with by regulators or professionals (Calnan et al., 2017; Hughes & Doheny, 2019; Moreira, 2011; Sjögren, 2008). This study provides a broader view by unfolding the conflicting concerns and expectations of good conduct that professionals need to navigate—of which epistemic quarrels constitute only one issue among others.

Second, the typology of professional responses presented in this study constitutes an analytical resource for future studies of how frontline professionals deal with conflicting concerns. Drawing on and adding to existing literature on professional responses to competing accountabilities, the conceptualisation of frontline professionals' responses developed in this study broadens the analytical focus from an emphasis on the cognitive and behavioural reactions of individual professionals to also encompass the social relationships and organisational conditions that shape professional work. The ability of frontline professionals to minimise conflict and align competing accountabilities relies not only on their own efforts, but also on the mobilisation—and sometimes resistance—of patients, relatives, colleagues and superiors. Overall, in our analytical framework, we distinguished between three types of responses: ones that (1) order accountabilities hierarchically as some ideas of good conduct are prioritised; (2) separate the enactment of competing accountabilities spatially or temporally; and (3) seek to align competing accountabilities by combining differing ideas of good conduct. A characteristic of the responses we identified in the empirical analysis was that frontline professionals strived to encompass informal expectations of good conduct expressed in relationships with the families of SMA patients and professional peers without violating formal regulations and expectations of good conduct expressed in hierarchical relations. They found ways of exercising discretion at the 'margins' (cf. Hedgecoe, 2006) of regulatory standards. In particular, effort was put into sustaining trusting relationships with parents and patients—more or less successfully. In addition to the responses of 'compartmentalisation', 'translation' and 'co-optation' identified in earlier studies (Gebreiter & Hidayah, 2019; Numerato et al., 2012;

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Pache & Santos, 2013), we found that the professionals mediated between national standards and individual patient traits to align different concerns on a case-by-case basis; a response we call 'situated mediation'. In concordance with Gebreiter and Hidayah (2019), we found that the attempts of frontline professionals to handle competing accountabilities were not mainly a result of strategic considerations. Their responses often emerged as situated attempts at 'muddling through' when confronted with demanding situations (cf. Calnan et al., 2017; Mechanic, 1997).

Third, by focussing on an empirical field at the forefront of genomic medicine, this study brings the sociological debate on priority setting and rationing up to date with current developments related to precision medicine (Prainsack, 2017). Hence, some of the challenges we identify are likely to increase and to be relevant beyond the field of neuropaediatrics. In particular, this study suggests that rationing criteria that are modelled on genetic markers of disease do not necessarily correspond to what frontline professionals conceive of as clinically relevant similarities and differences. The correspondence between phenotypic and genotypic markers of disease and related uncertainties are therefore important for authorities to take into account when delineating access to novel gene therapies. Moreover, our study suggests that it may be particularly difficult for frontline professionals to handle rationing decisions when it requires them to differentiate between patients who are linked through kinship relations. In such instances, regulatory ideals of procedural fairness and economic efficiency can collide with culturally ingrained ideas about fairness and family ties. In the realm of precision medicine, the genetic basis of diseases is increasingly being explored and targeted by new therapies. Consequently, disease aetiology and therapeutic options are likely to become more entangled with kinship relations, underlining the need for authorities and frontline professionals to consider how differential treatment of family members is to be dealt with in a context of priority setting. Furthermore, even if frontline professionals appear to acknowledge the legitimacy of rationing criteria based on scientific documentation of patients' potential treatment benefit, such criteria can still be difficult to handle in practice if consensus is not reached about the interpretation of trial results. Along with the development of precision medicine, the documentation of clinical effectiveness tends to become rife with uncertainties as study populations diminish in size and modelling may be needed to estimate long-term effects (Eichler et al., 2021). Accordingly, questions about how requirements on documented treatment benefit are to be balanced against 'the benefit of doubt' are likely to become more central. It has been beyond the scope of the present article to propose possible solutions to these dilemmas. While we find it unlikely that any absolute resolution can be identified, we believe that a first important step is to create awareness of the dilemmas and provide a basis for reflection about professional responses.

In conclusion, this study demonstrates that even if centralised priority setting may be a way of overcoming the challenges of 'bedside rationing', it can also generate dilemmas in frontline work—much like early rationing scholars foresaw (Mechanic, 1997; Syrett, 2003). We have shown that professional accountabilities can become more complex to navigate and prone to conflict, setting in motion a variety of professional responses. In the Danish case, we found that concern for economic efficiency and budget control (expected in hierarchical relations) sometimes collided with conceptions of good conduct expressed in relations with professional peers, patients and relatives. In addition, tension arose between concern for the safety and wellbeing of the patient and concern about maintaining good collaboration with relatives. Frontline professionals can respond to such dilemmas in quite ingenious ways as they strive to align competing concerns or otherwise reduce conflict between them. However, such efforts are unlikely to succeed if regulatory frameworks invite disputes over the discretionary power and responsibilities of profes-

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sionals, and if decision-makers at the top of the hierarchy hesitate to acknowledge their own responsibility for economic priorities.

AUTHOR CONTRIBUTIONS

Sarah Wadmann: Conceptualisation, methodology, investigation, formal analysis, writing—original draft preparation, writing—review and editing. **Amalie Martinus Hauge**: Conceptualisation, methodology, investigation, formal analysis, writing—review and editing. **Laura Emdal Navne**: Conceptualisation, methodology, investigation, formal analysis, writing—review and editing.

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CONFLICT OF INTEREST

As part of their employment at the Danish Center for Social Science Research—VIVE, all authors undertake commissioned research for public authorities and private institutions, including pharmaceutical companies and patient organisations.

DATA AVAILABILITY STATEMENT

Due to confidentiality agreements with the informants, we are unable to share the empirical data of this study. The publicly available documents that are referred to in the article, can be retrieved from the corresponding author upon request.

ETHICS STATEMENT

The study has been approved by the Human subject protection review board at The Danish Center for Social Science Research—VIVE.

PATIENT CONSENT STATEMENT

All observations and interviews that have involved patients have been conducted upon consent from patients or their relatives (when the patients were children).

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