A SOLID START FOR THE DUTCH FIRST THOUSAND DAYS-APPROACH

Insights into program adoption, monitoring and cross-sectoral collaboration

Joyce Marianne Molenaar

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General introduction

Importance of the first thousand days of life

The period from conception to a child's second birthday (i.e. the first thousand days of life) is crucial to children's further physical, mental and social development (1-3). During these first thousand days, the foundations for optimal later health and wellbeing are established (3, 4). The body grows, the immune system develops and all vital organs are formed. The pace of development far exceeds that of any other phase in life. Moreover, it is the period in which our developmental plasticity, the ability to adapt to environmental factors and exposures, is highest (5, 6). Although our experiences across the lifespan can still influence our development to some extent, the first thousand days form the basis for who we are and have lifelong effects (7, 8).

The well-studied Developmental Origins of Health and Disease concept (originally the 'Barker hypothesis') explains how early life experiences and exposures - both positive and negative - can influence later health and wellbeing (9, 10). There is an abundance of studies that show that many (chronic) health conditions such as diabetes, asthma, obesity, cardiovascular diseases, cancer and depression can trace their origins to early life (e.g. 11-18). This was demonstrated for instance by the findings of the Dutch Famine Birth Cohort study in which also timing of exposure appeared important (12, 19, 20). One of the described mechanisms of how early life exposures influence later health and wellbeing is through epigenetic programming. Epigenetic programming states that during critical times of development several factors can 'program' the bodily structures and functions to anticipate the environment it will face in the future (9, 10). Negative factors such as smoking during pregnancy, stress, pollutants and malnutrition can thereby lead to more susceptibility to diseases (21). For example, if a foetus is exposed to poor nutrition, it may adapt its metabolic system by storing more fat, which would be beneficial in case of food scarcity in adulthood, but can lead to obesity and other chronic diseases in an environment with abundant food (9, 10). These epigenetic changes can also be passed down from parents or grandparents to their offspring (8, 10).

A better physical, mental, and social development during early life can lead to various positive outcomes in the future for both the individual as well as society, and thus should be at the centre of investments (22, 23). Some of those positive outcomes include improved learning and behaviour, enhanced educational opportunities, better job prospects, more productivity and greater participation in the workforce or society later in life (8). The Nobel prize-winning economist James Heckman showed that the best return on investment that society can achieve is by focusing its efforts on these first few years (24, 25). Investing early means that the benefits can be enjoyed for longer, and have a compounding effect. These benefits result from both higher revenues as well as savings in costs related to social welfare, poverty, crime and negative health outcomes. Moreover, early interventions are generally less costly compared to later remedial programs (24). Hence, early life investments are the most efficient and effective public investments.

From a biomedical perspective to a focus on the social determinants of health

A wealth of research has indicated that our health and opportunities are not solely determined by our genetic, biological or medical characteristics, but rather depend on the direct and indirect impacts of social, economic, cultural and environmental conditions. These conditions in which people are born, grow, live, work, and age are called the Social Determinants of Health (SDOH) (26). Several studies have also proved the importance of (clustered) SDOH for outcomes during pregnancy or childbirth, being focused on factors such as area deprivation or socio-economic status with underlying concepts such as income, occupation and education (27-37). The SDOH framework provides an overview of the structural elements that shape the SDOH, their interrelatedness and the mechanisms by which social determinants generate health inequities (26). Although the medical sector also faces the consequences of unfavourable SDOH, many of the underlying elements and possible solutions fall outside their scope, posing a challenge to reduce health inequities within the medical sector alone (8).

The SDOH can have an impact at various stages of our lives: during the first thousand days, childhood, adolescence and adulthood. According to models of life course health development, our health development is a dynamic, complex and non-linear process that results from different exposures over the life course (7, 38, 39). Both negative (e.g. food or housing insecurity) and positive (e.g. positive school environment) contexts and experiences can lead to different health trajectories (8, 38). The life course theory emphasizes that health differences mainly result from exposures during critical periods in early development, which subsequently accumulate throughout the course of one's life.

The National Academies of Sciences, Engineering, and Medicine (NASEM) used the above insights in their report 'Vibrant and Healthy Kids: Aligning Science, Practice, and Policy to Advance Health Equity' (8). Their conceptual model shows various elements that shape children's health and development during the life course, from conception into adulthood (Figure 1). The development and health of children is presented in the inner purple circle. In the direct environment of children are the factors that directly influence their daily experiences and patterns, such as family cohesion, caregiver well-being and nurturing (dark pink circle). These factors are shaped by the SDOH (pink circle). These SDOH, in turn, are influenced by the outer level: the socioeconomic and political drivers such as policies and laws that distribute resources and opportunities among the population (grey circle). The distribution of resources and opportunities is often disproportionate, based on characteristics such as race, gender or social class. The model provides opportunities for interventions to enhance individual and population health, as well as health equity, from micro to macro levels. Moreover, the model provides a powerful call for practice and policy to prioritize investments in improving preconception and perinatal health, since early life experiences can shape health and well-being across an entire lifetime for parents themselves, but these risks and protective factors can also be transmitted to their children. As these children grow into adulthood and potentially become parents themselves, this can lead to new cycles of inequity or resilience. Because of the intergenerational aspect,

(future) parents or caregivers are a central focus in optimizing children's health and well-being and reducing health inequities (8, 40).

Early life health inequities are known as unjust, unnecessary and preventable differences in health between different (social) groups (41). This can be related to, for example, income, ethnicity, immigration status, education, living circumstances, gender or sexual orientation. The concept of health inequities is frequently used interchangeably with health disparities. Moreover, it is at times mixed-up with health inequalities, which refers more broadly to measurable differences in health between groups, without a moral judgement (41). One example of health inequities is seen in life expectancy (2019 – 2022): individuals with a higher educational level in the Netherlands live 5 years longer, and 14 years longer in good health, compared to individuals with a low educational level (42). Also for perinatal health outcomes during pregnancy and childbirth, inequities exist between and within high-income countries (43). For example, across Dutch municipalities, preterm birth rates ranged from 2.4% to 11.7% in 2021 (mean: 6.6%) (44). There are also large differences in perinatal outcomes between neighbourhoods with varying levels of socioeconomic status, as demonstrated well by the work of researchers from Rotterdam (28, 45-47).
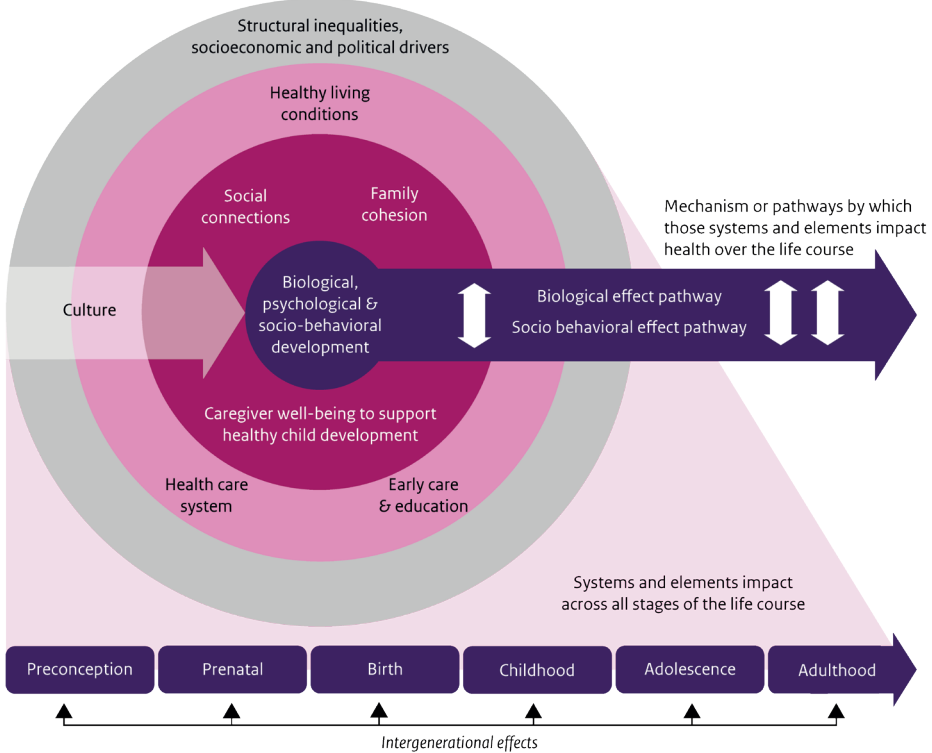

Figure 1. Conceptual framework 'Leveraging early opportunities to advance health equity across the life course' by the National Academies of Sciences, Engineering, and Medicine (2019) (8).

Despite accumulating evidence regarding the determinants of poor health, health inequities in birth outcomes seem to have persisted (48) and may even be widening in certain populations and for specific health outcomes (49, 50). These considerations of equity are also very important in investing in early life.

The concept of vulnerability in early life
Thousands of parents and children in high-income countries are exposed to adverse conditions such as poverty, violence, inadequate nutrition, substance abuse, and stress. This means that many face an increased risk or susceptibility to adverse health outcomes or decreased well-being, or they experience a lower access to care. Recent literature often uses the concept of 'vulnerability' when referring to these (future) parents and their newborn or unborn children (51-54), but terms such as 'disadvantaged', 'deprivation' or 'frailty' are also common in the scientific literature.

There are diverse and heterogeneous definitions and understanding of the concept of vulnerability around pregnancy. For example, de Groot and colleagues (2019) defined vulnerability as "a dynamic state that reflects converging effects of a set of interacting and amplifying personal and environmental factors" (p. 12), which increases a person's susceptibility to ill health and hamper their recovery (53). Scheele and colleagues (2020) referred to pregnant vulnerable women as being "threatened by physical, psychological, cognitive and/or social risk factors in combination with lack of adequate support and/ or adequate coping skills" (p. 4) (54). Various stakeholders in the city of Rotterdam (2020) explained how "vulnerability arises from an imbalance between risk factors and protective factors" (55). Their definition of vulnerability includes a distinction between highly vulnerable women for whom the risk factors require immediate action (e.g. domestic violence) and vulnerable women who have one or more risk factors (e.g. unhealthy lifestyle factors, unemployment) and insufficient protective factors (e.g. supportive social network, stable home situation). The Dutch national organization for midwives (Dutch abbreviation: KNOV) described how vulnerable pregnant women face several challenging circumstances, emphasizing different risk factors (56). Briscoe, Lavender and McGowan (57) described vulnerability in three main attributes: threat, barrier and repair. Whether potential biological, psychosocial or sociological threats lead to vulnerability, depends on both the existing recovery systems available (e.g. warm supporting relationships), as well as barriers that may impede access to healthcare (e.g. stigmatization, lack of compassion).

Taken together, most definitions of vulnerability acknowledge that vulnerability encompasses a dynamic, contextualized and complex process involving the interplay of risk and protective factors at different levels or life domains (51, 53-55, 58). In simplified terms, several stressors at either the individual or contextual level can function as risk factors contributing to vulnerability, whereas protective factors have the potential to diminish or prevent vulnerability. Whether risk factors increase vulnerability and hinder people from achieving their full potential, depends on the co-occurrence and balance of risk factors and protective factors (53, 55).

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When reviewing the previous literature about the influence of social factors and vulnerability on birth outcomes, it appears that most studies focus on a limited number of predetermined, single risk factors. Few authors have studied the clustering or interactions between risk factors (29, 30, 59-61). Moreover, protective factors are rarely considered in the studies. Hence, the influence of the co-existence of both protective and risk factors requires further study.

Need for improved collaboration across the social and medical sector to address vulnerability and inequity
Increased awareness of the influence of social factors has prompted further exploration of preventive strategies and interventions to address vulnerability and inequity during the first thousand days. While healthcare has a pivotal role in advancing health equity, it cannot effectively address health inequities on its own. Since many of the underlying determinants for health and well-being lie beyond the medical domain, addressing them requires collaboration with other domains as well. Recent literature widely acknowledges that cross-sectoral collaboration between the medical and social sector is necessary to provide children the best possible start in life (8, 62-64).

The urge for increased collaboration aligns with a wider movement in Western countries to maintain an accessible, affordable, safe and effective healthcare system. Our healthcare systems face increased pressure due to rising costs, ageing populations, changing disease patterns and care needs, and an alarming shortage of personnel (65-68). These challenges and the need to respond also applies to the maternity care population and system, with increasing maternal age, more co- and multimorbidity and unhealthier lifestyle among women of childbearing age, technological developments, and more diversity in cultural and ethnic groups (69, 70). These pressing issues also underscore the importance of implementing preventive measures and integrating medical and social care and support.

Previous research on collaboration during the first thousand days has predominantly focused on specific temporal windows within either the medical or social sector. For example, studies within the Netherlands (71-76) and other countries (77-81) explored collaboration between professionals and organizations during either pregnancy, childbirth or child service delivery. Collaboration in Dutch maternity care is often described as complex and not self-evident, as healthcare providers historically have worked relatively autonomous with separated organizational structures, education programs, protocols, cultures and practices (63, 74, 82). Few studies have devoted attention to the full period of the first thousand days within both the social and medical sectors (62, 63). Collaboration between sectors may present different challenges compared to collaboration within one sector, potentially due to larger differences in cultures and structures.

A nationwide first thousand days-approach: Dutch action program Solid Start
Yearly, approximately 170.000 children are born in the Netherlands (70). These children and their parents, especially those in vulnerable situations, could benefit from a more integrated and population health-based care and support system. In 2018, the nationwide

action program 'Solid Start' was launched by the Dutch Ministry of Health, Welfare, and Sport (Dutch abbreviation: VWS) to ensure that every child receives the best possible start during the first thousand days of life (83). It promotes collaborative efforts across the medical and social sector and focuses particularly on (future) parents and young children in vulnerable situations. The action program's strategic framework is built upon previous endeavours aimed at integrating medical and social services, such as the local 'Ready for a baby' program in Rotterdam (2008–2012) (84) and subsequent 'Healthy Pregnancy 4-All' programs implemented in various municipalities since 2011 (28, 62, 85). It is part of a wider movement in Dutch maternity care, which developed from a narrow focus to the mother's health during childbirth, to a more social and cross-sectoral approach for (future) parents and children in which pregnancy and early childhood is considered a window of opportunity to address health inequities and enhance overall well-being (63). Several key moments catalysed this movement. For example, the European Peristat reports showed relatively high perinatal mortality rates in 2004 and 2008 (86, 87) which created momentum for a cascade of activities (88). Activities including the establishments of maternity care networks in which midwives, gynaecologists and other maternity care providers collaborate (89), experiments with bundled payment (90) and the initiation of the Standard for Integrated Maternity Care (91).

The action program Solid Start employs a comprehensive and population-based strategy (83). It is conceptualized and implemented across three pillars: before pregnancy, during pregnancy, and after birth. At the start of the action program Solid Start, several aims were set, summarized as follows: prevent unintended pregnancies, prepare parents better for pregnancy, identify medical and non-medical issues earlier, and offer tailored support for (future) parents in vulnerable situations. The preventive and supportive measures aim to address the underlying determinants of health and well-being from an early stage, to prevent or mitigate health-related issues that may arise later in life. The program's backbone is the stimulation of cross-sectoral collaboration through local coalitions Solid Start. Municipalities are vital in creating local coalitions Solid Start, consisting of organizations and service providers spanning the medical, social and public health domain. Involved stakeholders can include midwives, obstetricians, maternity care assistants, youth healthcare providers, social workers, debt counsellors, municipal officials, experts-by-experience. Municipalities are stimulated to create their own approach that fits their local context, challenges and existing networks. Since the decentralization in 2015, municipalities were already given new responsibilities in youth care, long-term care and income-support that fuelled differences in their approach and services (92). Municipalities received financial support from the Ministry of Health, Welfare and Sport, and they were provided assistance in building or strengthening their coalition from Pharos, the Dutch Centre of Expertise on Health Disparities (93). Pharos' advisors have one-on-one meetings with municipalities, but the organization also provides shared training, webinars and informative webpages. Other support for local coalitions Solid Start included the availability of an analysis tool, data, a list of effective interventions and care pathways. Moreover, the Ministry of Health, Welfare and Sport facilitates and stimulates the action program Solid Start by striving for legal changes. Part of the action program also

includes the implementation of interventions, including 'Not Pregnant Now' that supports professionals in sustaining the autonomy of vulnerable groups in making informed choices regarding pregnancy and contraception (94).

Starting from 2019, the Ministry of Health, Welfare and Sport commissioned the National Institute for Public Health and the Environment (Dutch abbreviation: RIVM) to monitor the action program Solid Start. There are several reasons to monitor policy programs like the action program Solid Start. These reasons, for example, relate to accountability, learning and engagement (95, 96). Firstly, monitoring can be a tool to document actions and assess their alignment with predetermined plans or objectives. Secondly, monitoring for learning aims to provide insight into the approach (e.g. progress, facilitators, barriers) to allow reflection and make improvements. Thirdly, monitoring can facilitate the sharing of successes and small-wins, thereby keeping people engaged and enthusiastic. The monitoring efforts in relation to the action program Solid Start initially focus on gaining insight into how certain processes and outcomes develop over time, without determining causal effects.

In order to start monitoring the action program Solid Start, decisions had to be made on how to operationalize certain concepts (e.g. vulnerability) and which data and indicators are useful. Considering the cross-sectoral approach, a cross-sectoral data infrastructure was considered beneficial in the monitoring endeavours.

OBJECTIVE OF THIS THESIS

The main objective of this thesis is to provide insight into the adoption of the action program Solid Start, thereby focusing on monitoring and cross-sectoral collaboration. In this thesis, the monitoring aspect relates to both the what and how to monitor, as well as the developments and experiences with the action program Solid Start.

THESIS CONTEXT

This thesis constitutes the scientific basis for the monitoring of the Dutch action program Solid Start that is conducted by the National Institute for Public Health and the Environment. The National Institute for Public Health and the Environment is a knowledge institute that conducts independent scientific research for commissioning partners. In the case of the action program Solid Start, this is the Dutch Ministry of Health, Welfare and Sport (97). The acquired knowledge is shared with the government, professionals and the general public to support a healthy population and environment. The organization is an agency of the Dutch Ministry of Health, Welfare and Sport.

In 2019, the National Institute for Public Health and the Environment started to monitor the Dutch action program Solid Start at national level. This national monitor has quantitative

and qualitative components. For the quantitative component, a Delphi study with experts from policy, practice and research was conducted to develop a set of fifteen indicators (98). Indicators reflect both processes (e.g. percentage of municipalities with a local coalition Solid Start) and outcomes (e.g. percentage of children born prematurely and/or with a low birth weight for gestational age). Together, these indicators provide insight in both the progress of program implementation, as well as developments or trends in health and its underlying factors for parents and children (98). Several data sources are used to quantify the indicators, including questionnaires among municipalities, inquiries among national or regional organizations (e.g. among those implementing interventions), and the nationwide population-based data infrastructure DIAPER (99). DIAPER (acronym for Data-InfrAstructure for ParEnts and childRen) combines routinely collected data from three major Dutch nationwide sources: 1) Perined - the Dutch perinatal registry that collects routine care data during pregnancy and childbirth on care use and health outcomes (100), 2) Vektis – the healthcare information centre that compiles data on medical spending under the Healthcare Insurance Act (101), and 3) Statistics Netherlands (Dutch abbreviation: CBS), which collects and publishes linkable data on societal aspects, including health, welfare, income, education, and employment (102, 103). DIAPER provides a suitable source to study the action program Solid Start and its related elements, because cross-sector data is considered essential in order to gain a comprehensive understanding of cross-sector collaboration. The qualitative component includes yearly focus group discussions and interviews with those involved in the action program Solid Start, including representatives from care and support organizations (e.g. managers and care providers), Solid Start project leaders and advisors, municipal officials, representatives of national knowledge institutes and professional associations, researchers, and experts-by-experience and clients. All results of the monitor are publicly available and presented in yearly factsheets or notes addressed to the Ministry of Health, Welfare and Sport (104-107). A scientific advisory committee oversees the monitoring activities.

In 2021, the National Institute for Public Health and the Environment was also commissioned to support municipalities in monitoring their local approach within the 'learning local monitor Solid Start'. The support program centralizes learning and knowledge sharing between and within local coalitions Solid Start. It aims to encourage both starting and more developed local coalitions Solid Start to use monitoring as a tool to reflect on and design their local Solid Start approach. To do so, the National Institute for Public Health and the Environment organizes regular learning sessions with eleven local coalitions Solid Start in which participants share best practices, challenges and needs for monitoring. Those needs are addressed in thematic sessions for a wider audience, open to all who are involved or interested in monitoring or implementing the action program Solid Start at the local level.

OUTLINE OF THIS THESIS

Chapter 2 and 3: monitoring vulnerability
The first two studies addressed the monitoring of vulnerability during pregnancy. The action program Solid Start specifically focuses on (future) parents and children in a vulnerable situation. Monitoring vulnerability at population-level requires more insight into the operationalization of vulnerability. We used various data-science techniques to gain insight into different vulnerability-classes with varying combinations of risk and protective factors (Chapter 2), and to identify if we could predict vulnerability at population-level using nationwide routinely collected data (Chapter 3). This led to the following overall research question: *What is vulnerability during pregnancy, and how to operationalize vulnerability for monitoring?*

Chapter 4: indicators for local monitoring
The action program Solid Start was quantitatively monitored at a national level right from the start of the program. Monitoring Solid Start for municipalities or coalitions at the local level may require different indicators, given the different context, informational needs and intended use. Therefore, Chapter 4 of this thesis describes how we used a Delphi approach in developing an indicator set to monitor the action program Solid Start on a local level. The research question was: *Which indicators can be used to monitor the action program Solid Start on a local level?*

Chapter 5: developments and experiences with Solid Start and cross-sectoral collaboration
The action program Solid Start was implemented at the end of 2018 with the aim to provide every child the best possible start in life. A key program element is to improve the collaboration between the medical and social sector by creating local coalitions Solid Start. Therefore, we aimed to describe the implementation of the action program Solid Start during the program's own first thousand days (2019, 2020 and 2021) with a specific focus on cross-sectoral collaboration. We used both quantitative and qualitative research methods to answer the following research question in Chapter 5: *What are the developments and experiences with the action program Solid Start, specifically regarding cross-sectoral collaboration?*

Chapter 6: general discussion
The separate and combined findings from the studies offer deeper insights into 1) the adoption of the action program Solid Start, 2) monitoring and 3) cross-sectoral collaboration. Chapter 6 discusses the main findings in light of these three elements, providing key lessons learned. The chapter proceeds with methodological considerations along with recommendations for research, a future outlook with recommendations for policy, practice and education, and concluding remarks.

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Defining vulnerability subgroups among pregnant women using pre-pregnancy information: a latent class analysis

Joyce M. Molenaar
Lindsey van der Meer
Loes C.M. Bertens
Eline F. de Vries
Adja J.M. Waelput
Marian Knight
Eric A.P. Steegers
Jessica C. Kiefte-de Jong
Jeroen N. Struijs

ABSTRACT

Background
Early detection of vulnerability during or before pregnancy can contribute to optimizing the first thousand days, a crucial period for children's development and health. We aimed to identify classes of vulnerability among pregnant women in the Netherlands using pre-pregnancy data on a wide range of social risk and protective factors, and validate these classes against the risk of adverse outcomes.

Methods
We conducted a latent class analysis based on 42 variables derived from nationwide observational data sources and self-reported data. Variables included individual, socioeconomic, lifestyle, psychosocial and household characteristics, self-reported health, healthcare utilization, life-events and living conditions. We compared classes in relation to adverse outcomes using logistic regression analyses.

Results
In the study population of 4172 women, we identified five latent classes. The largest 'healthy and socioeconomically stable'-class [n = 2040 (48.9%)] mostly shared protective factors, such as paid work and positively perceived health. The classes 'high care utilization' [n = 485 (11.6%)], 'socioeconomic vulnerability' [n = 395 (9.5%)] and 'psychosocial vulnerability' [n = 1005 (24.0%)] were characterized by risk factors limited to one specific domain and protective factors in others. Women classified into the 'multidimensional vulnerability'-class [n = 250 (6.0%)] shared multiple risk factors in different domains (psychosocial, medical and socioeconomic risk factors). Multidimensional vulnerability was associated with adverse outcomes, such as premature birth and caesarean section.

Conclusions
Co-existence of multiple risk factors in various domains is associated with adverse outcomes for mother and child. Early detection of vulnerability and strategies to improve parental health and well-being might benefit from focussing on different domains and combining medical and social care and support.

INTRODUCTION

The first thousand days of life, from preconception to the child's second birthday, are crucial to children's further physical, mental and social development. This critical and sensitive period is an important determinant of health and well-being in adulthood, as supported by the well-evidenced Developmental Origins of Health and Disease (DOHaD) concept (1, 2). The DOHaD concept explains how experiences and exposures during early life, such as stress and nutrition, influence susceptibility to disease in later life and across generations, arguably through epigenetic mechanisms of foetal programming (1, 2). Because of this intergenerational aspect, parents are the central focus to improve child health and advance health equity (3).

To indicate subgroups of parents and their unborn or newborn children who are at higher risk of poor health or have lower access to healthcare, the concept of vulnerability is often used (4-6). Vulnerability reflects a complex and dynamic process. Simplified, various stressors at individual or contextual level (e.g. unemployment or living in a deprived neighbourhood) can act as risk factors to vulnerability, while protective factors (e.g. stable social network) might reduce or prevent vulnerability (4, 5, 7, 8).

Whether the presence of risk factors increases vulnerability and thereby hinder achieving one's optimal health potential depends on the balance and interaction between risk and protective factors (4, 8). While research on perinatal health has traditionally focussed on risk factors of a medical nature, there is now indisputable evidence for direct and indirect influences of social factors as well (9-14). The social, economic, cultural and environmental living conditions (i.e. social determinants of health) that shape parents' and children's daily experiences and thereby influence their health and development, are embedded in larger systems and structures such as policies and laws (3, 15).

There is an international growing professional and political focus on early detection of vulnerability during the first thousand days and development of effective strategies to improve parental health and well-being (3, 16). For instance in the Netherlands, the government launched a nationwide 'Solid Start'-programme in 2018 with the aim of providing each child the best start in life by strengthening collaboration between medical and social services, with a specific focus on families in vulnerable situations (16). Detecting vulnerability during pregnancy with the preventive purpose of countering suboptimal child health is challenging and can benefit from in-depth knowledge into vulnerability.

However, currently, little is known about the combination of different risk and protective factors to vulnerability and its influence on health outcomes. There seems to be few studies that consider protective factors to vulnerability and there is limited insight into clustering and underlying interactions, while it is recognized that especially the co-existence of risk factors can lead to adverse birth outcomes (11, 17, 18). Previous studies frequently explored the association between a limited number of predetermined, single risk factors

and adverse birth outcome, but neglected co-existence of both protective- and risk factors that can influence outcomes (12, 18, 19).

The aim of this study was to identify classes of vulnerability among pregnant women based on a wide range of social risk and protective factors in a latent class analysis (LCA). We conducted the LCA using Dutch observational nationwide data sources and self-reported data prior to pregnancy. In addition, we validated these classes by studying the association between latent class membership and various maternal and perinatal health outcomes and care utilization.

METHODS

Data sources
This study utilized data from the nationwide population-based data infrastructure DIAPER (acronym for Data-InfrAstructure for ParEnts and childRen). DIAPER integrates routinely collected observational data from three Dutch nationwide data sources (Perined, Vektis and Statistics Netherlands) at individual level. The Dutch Perinatal Registry 'Perined' collects routine care data on pregnancy after 22 weeks of gestation, birth and the first 28 days after birth, as supplied by midwives, gynaecologists and paediatricians (20). Healthcare information centre 'Vektis' collects claims data under the Dutch Healthcare Insurance Act and provides data on healthcare utilization and spending (21). 'Statistics Netherlands' collects and publishes data on societal matters and provides access to data through their System of Social Statistical Datasets (SSD) (22, 23). This linkable SSD-data covers nearly 20 themes, including health, welfare, income, education and labour.

We enriched DIAPER with self-reported data on health, well-being and lifestyle of the Public Health Monitor 2016 (PHM-2016) (24). This is a health survey among a varying sample of the Dutch population aged 19 years and older, carried out every 4 years by the Community Health Services, Statistics Netherlands and the National Institute for Public Health and the Environment. The PHM-2016 had 457.153 participants and was mainly conducted from September-December 2016. Appendix A provides more information about the data sources.

Study population
To ensure that information was not influenced by pregnancy itself, women were eligible for inclusion if these criteria were met: (i) they participated in the PHM-2016 (pre-pregnancy), (ii) they gave birth (livebirth or stillbirth) or had a termination of pregnancy before 1 January 2019, and (iii) pregnancy data in 2017 or 2018 were recorded within Perined. In case women had multiple pregnancies or births during the study period, only data on the first observation was included, to avoid duplication of women's characteristics.

Variables

The selection of variables for the LCA started with compiling a list of all possible risk and protective factors to vulnerability based on the framework of the National Academies of Sciences, Engineering, and Medicine (3), other scientific studies and definitions of vulnerability (4, 5, 8), and expertise of the research team. Based on this list, 42 variables were available and selected in our data sources. These were divided into nine themes: individual characteristics, socioeconomic characteristics, lifestyle factors, household characteristics, self-reported health, healthcare expenditures and utilization, psychosocial characteristics, life-events and living conditions. The timing of the PHM-2016 was decisive in the choice for 1 October 2016 as baseline to include information. If data were available only on yearly basis, we included data from 2016. To increase interpretability, variables were categorized into two or three categories with the first category representing the risk factor to vulnerability. Appendix 1 provides a detailed overview of the variables, including definitions, categories and sources.

Outcomes

We studied the association between latent class membership and perinatal and maternal health outcomes and care utilization to validate classes. Perinatal health outcomes comprised: preterm birth (<37 weeks), small for gestational age (SGA, <10th percentile corrected for gestational age and foetal sex), preterm birth and/or SGA, and admission to a neonatal intensive-care unit (NICU) after birth. Maternal health outcomes comprised: primary and secondary caesarean section, pre-eclampsia/hypertension and postpartum haemorrhage (≥1000 ml). Outcomes regarding healthcare utilization included: not having the first antenatal care appointment (i.e., booking visit) before the 10th week of pregnancy and not receiving postpartum care (at home) after birth. Appendix 1 provides more information.

Statistical analyses

Latent class analysis

LCA is a data-driven analysis technique that aims to structure heterogeneity in a population by classifying individuals into unobserved – or latent – homogeneous classes (25). Structuring is based on included variables. Each class is denoted by conditional probabilities for each variable to take on a certain response value (e.g. 1 or 0), with the objective to categorize individuals into the smallest possible set of distinct and interpretable latent classes.

Using R version 3.6.2 (package poLCA), we estimated latent class models using all 42 variables with no prior assumptions about the optimal number of classes (26). Missing data were imputed through Multiple Imputation using Chained Equations (MICE) (Appendix 2). We started with a one-class model and stepwise increased to a 15-class model. Parameters of the latent class models were estimated by maximum likelihood. We considered both statistical fit as well as parsimony and interpretability to select the optimal model (25). To compare the competing models' relative fit, we used the Akaike Information Criterion (AIC) (27) and sample-size adjusted Bayesian Information Criterion (aBIC) (28). Lower values

indicate better fit of the model to the data. We also considered the fit-indices' relative decrease, as done in previous studies (29), because a continuous decrease in the AIC is common with large sample sizes and the aBIC also may indicate towards a model with more classes than useful (30). We additionally reviewed the models' entropy, which reflects how clearly the classes can be distinguished with scores ranging from 0 to 1 (optimum) (31). We selected three preferred models based on their fit statistics and compared their item-response probabilities. The final model was selected based on parsimony and interpretability and women were classified into one of the identified classes based on predicted class membership (largest posterior probability). Further, to evaluate the LCA's robustness, we performed two additional analyses. First, to unravel the impact of previous pregnancies, we excluded nullipara and conducted a LCA with additionally previous perinatal and pregnancy outcomes. Second, to evaluate whether similar vulnerability classes can be distinguished across women in the entire reproduction age, we repeated the LCA with a different study population consisting of all women between 19 and 44 years old.

Regression analysis
We studied the association between class membership and adverse outcomes by means of unadjusted logistic regression analysis. Results are reported as odds ratios (ORs) with 95% confidence interval (CI). A *p*-value of <0.05 was considered statistically significant.

RESULTS

The study population consisted of 4172 women, of whom 1129 had missing data (Table 1). A five-class model was considered best (see Appendix 3 for fit-indices). The aBIC reached a minimum in the 12-class model, but did not show considerable improvement after models beyond seven classes when reviewing the relative fit (elbow shape). The AIC continuously decreased as expected. Entropy values were regarded best for models with two to five classes. We compared the interpretation of models with four, five and six classes and chose the five-class model for its interpretative and distinctive classes.

Table 1. Characteristics of the study population (including missing data)

		n (%)
Individual characteristics		
Age	19-23	306 (7.3)
	24-35	3528 (84.6)
	>35	338 (8.1)
Ethnicity	Non-Western	420 (10.1)
	Western	343 (8.2)
	Native Dutch	3409 (81.7)
Parity[*]	Nullipara	1755 (42.1)
	Primipara, multipara	2410 (57.8)
	Missing	*<10 (<0.2)*
Asylum seeker status	Yes	39 (0.9)
	No	4133 (99.1)
Socioeconomic characteristics		
Educational level	Low	328 (7.9)
	Moderate	1513 (36.3)
	High	2303 (55.2)
	Missing	*28 (0.7)*
Household income	Low	202 (4.8)
	Moderate	3348 (80.2)
	High	591 (14.2)
	Missing	*31 (0.7)*
Socioeconomic position	No income/ receiving benefits	532 (12.8)
	Student	82 (2.0)
	Paid work	3502 (83.9)
	Missing	*56 (1.3)*
Debts and payment arrears	Yes	45 (1.1)
	No	4127 (98.9)
Insufficient financial resources	Yes	524 (12.6)
	No	3267 (78.3)
	Missing	*381 (9.1)*
Permanent contract	No	1929 (46.2)
	Yes	2243 (53.8)
Full-time contract	No	1925 (46.1)
	Yes	2247 (53.9)
Lifestyle factors		
Smoking	Yes	661 (15.8)
	No	3315 (79.5)
	Missing	*196 (4.7)*
Alcohol use	Yes (excessive)	418 (10.0)
	No	3503 (84.0)
	Missing	*251 (6.0)*
Physical activity	Less than recommended	1696 (40.7)
	As recommended or more	2158 (51.7)
	Missing	*318 (7.6)*
Body Mass Index (BMI)	Unhealthy BMI	1386 (33.2)
	Healthy BMI	2641 (63.3)
	Missing	*145 (3.5)*
Household characteristics		
Type of household	One-person/ parent household	353 (8.5)
	Other	3819 (91.5)
Marital status	Unmarried	2147 (51.5)
	Married	2025 (48.5)
Dissolution of marriage	Yes	58 (1.4)
	No	4114 (98.6)

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Table 1. Continued.

		n (%)
Household size	≥6 persons	93 (2.2)
	<6 persons	4079 (97.8)
Youth support uptake	Yes	102 (2.4)
	No	4070 (97.6)
Self-reported health		
Perceived health status	Negative	465 (11.1)
	Positive	3653 (87.6)
	Missing	*54 (1.3)*
Long-term illness	Yes	747 (17.9)
	No	3362 (80.6)
	Missing	*63 (1.5)*
Restricted by health	Yes	724 (17.4)
	No	3330 (79.8)
	Missing	*118 (2.8)*
Healthcare expenditures and utilization		
Overall healthcare expenditures	High	824 (19.8)
	Low-average	3297 (79.0)
	Missing	*51 (1.2)*
General practitioners' (GP) expenditures	High	827 (19.8)
	Low-average	3308 (79.3)
	Missing	*37 (0.9)*
Hospital expenditures	High	413 (9.9)
	Low or none	3708 (88.9)
	Missing	*51 (1.2)*
Medication use	High	428 (10.3)
	Low or none	3744 (89.7)
Addiction related care uptake	Yes	23 (0.6)
	No	4149 (99.4)
Psychosocial characteristics		
Mental healthcare uptake	Yes	228 (5.5)
	No	3907 (93.6)
	Missing	*37 (0.9)*
Risk of depression or anxiety disorders	Moderate – high risk	1716 (41.1)
	No or low risk	2256 (54.1)
	Missing	*200 (4.8)*
Loneliness	Feeling lonely	1100 (26.4)
	Not feeling lonely	2719 (65.2)
	Missing	*353 (8.5)*
Feelings of control over life	Low	144 (3.5)
	Moderate	2741 (65.7)
	High	1006 (24.1)
	Missing	*281 (6.7)*
Mild intellectual disability	Yes	13 (0.3)
	No	4159 (99.7)
Life-events		
Crime suspect	Yes	95 (2.3)
	No	4077 (97.7)
Crime victim	Yes	874 (20.9)
	No	3298 (79.1)
Having been detained*	Yes	not shown
	No	not shown
History of frequent moving	Yes	1250 (30.0)
	No	2900 (69.5)
	Missing	*22 (0.5)*

Table 1. Continued.

		n (%)
Loss of a family member	Yes	147 (3.5)
	No	4025 (96.5)
Living conditions		
Home ownership	Rented	990 (23.7)
	Owner occupied	3099 (74.3)
	Missing	*83 (2.0)*
Motorized vehicle ownership	No	494 (11.8)
	Yes	3678 (88.2)
Proximity to general practitioners' (GP) office	>3 km	265 (6.4)
	<3 km	3847 (92.2)
	Missing	*60 (1.4)*
Liveability neighbourhood	Low-mediocre	273 (6.5)
	High	3695 (88.6)
	Missing	*204 (4.9)*
Outcomes		
Preterm birth	Yes	277 (6.6)
	No	3895 (93.4)
Small for gestational age (SGA)	Yes	324 (7.8)
	No	3814 (91.4)
	Missing	*25 (0.6)*
Preterm birth and/or SGA	Yes	557 (13.4)
	No	3590 (86.0)
Admission to neonatal intensive care unit (NICU)	Yes	130 (3.1)
	No	4042 (96.9)
Primary caesarean section	Yes	318 (7.6)
	No	3854 (92.4)
Secondary caesarean section	Yes	303 (7.3)
	No	3869 (92.7)
Pre-eclampsia/hypertension	Yes	250 (6.0)
	No	3922 (94.0)
Postpartum haemorrhage	Yes	265 (6.4)
	No	3907 (93.6)
No postpartum care (at home)	No postpartum care	258 (6.2)
	Postpartum care	3914 (93.8)
No antenatal care before week 10	No antenatal care before week 10	563 (13.5)
	Antenatal care before week 10	3236 (77.6)
	Missing	*373 (8.9)*

* Following guidelines of Statistics Netherlands, the data of some variables were rounded (parity) or not shown (having been detained) to prevent disclosure of information about individuals.
Detailed definitions of variables and categories are provided in Appendix 1.
Missing data are shown in italic.

The five-class model divided the study population into one class characterized by vulnerability in various domains, three classes characterized by vulnerability predominantly in one specific domain and one class with mainly protective factors (see Table 2 for all class proportions and characteristics). Figure 1 provides a visual representation.

Table 2. Class proportions and descriptives of the final 5-class model

	Class	1	2	3	4	5
	Label	Multi-dimen-sional vulne-rability	High care utili-zation	Socio-economic vulne-rability	Psycho-social vulne-rability	Healthy and socio-economi-cally stable
	Class proportions	0.06 (n=250)	0.11 (n=485)	0.09 (n=395)	0.24 (n=1005)	0.49 (n=2040)
Individual characteristics						
Age	19-23	0.14	0.03	*0.16*	0.13	0.03
	24-35	0.74	0.84	0.75	0.76	*0.92*
	>35	0.12	*0.13*	0.09	0.10	0.05
Ethnicity	Non-Western	0.26	0.02	*0.44*	0.13	0.02
	Western	0.06	0.08	*0.13*	0.11	0.06
	Native Dutch	0.68	0.90	0.43	0.76	*0.91*
Parity	Nullipara	0.38	0.39	0.23	*0.55*	0.41
	Primipara, multipara	0.60	0.61	*0.77*	0.45	0.59
Asylum seeker status	Yes	0.00	0.00	*0.09*	0.00	0.00
	No	0.98	*1.00*	0.91	*1.00*	*1.00*
Socioeconomic characteristics						
Educational level	Low	*0.30*	0.04	*0.30*	0.09	0.01
	Moderate	*0.54*	0.31	0.39	0.50	0.29
	High	0.16	0.65	0.30	0.41	*0.70*
Household income	Low	0.16	0.00	*0.30*	0.05	0.00
	Moderate	0.82	0.75	0.66	*0.90*	0.80
	High	0.00	*0.25*	0.03	0.05	0.20
Socioeconomic position	No income/ receiving benefits	**0.62**	0.03	**0.87**	0.00	0.02
	Student	*0.06*	0.00	*0.06*	0.04	0.00
	Paid work	0.30	0.97	0.06	0.96	*0.98*
Debts and payment arrears	Yes	*0.12*	0.00	0.03	0.00	0.00
	No	0.88	*1.00*	0.97	*1.00*	*1.00*
Insufficient financial resources	Yes	***0.60***	0.07	0.35	0.27	0.02
	No	0.38	0.93	0.65	0.73	*0.98*
Permanent contract	No	**0.92**	0.32	**0.99**	0.45	0.34
	Yes	0.08	0.68	0.01	0.55	0.66
Full-time contract	No	**0.74**	0.45	**0.96**	0.31	0.40
	Yes	0.26	0.55	0.04	*0.69*	0.59
Lifestyle factors						
Smoking	Yes	*0.36*	0.16	0.14	0.25	0.12
	No	0.64	0.84	0.86	0.75	*0.88*
Alcohol use	Yes (excessive)	*0.14*	0.10	0.04	0.11	0.12
	No	0.86	0.90	*0.96*	0.89	0.88
Physical activity	Less than recommended	*0.52*	0.47	0.48	0.45	0.42
	As recommended or more	0.48	0.54	0.52	0.55	*0.58*
Body Mass Index (BMI)	Unhealthy BMI	***0.64***	0.30	0.49	0.42	0.26
	Healthy BMI	0.36	0.70	0.51	0.58	*0.74*
Household characteristics						
Type of household	One-person/ parent household	*0.38*	0.03	0.10	0.15	0.03
	Other	0.62	*0.97*	0.90	0.85	*0.97*
Marital status	Unmarried	***0.66***	0.45	0.30	0.47	0.46
	Married	0.34	0.55	*0.70*	0.42	0.54

Table 2. Continued.

	Class	1	2	3	4	5
	Label	Multi-dimen-sional vulne-rability	High care utili-zation	Socio-economic vulne-rability	Psycho-social vulne-rability	Healthy and socio-economi-cally stable
	Class proportions	0.06 (n=250)	0.11 (n=485)	0.09 (n=395)	0.24 (n=1005)	0.49 (n=2040)
Dissolution of marriage	Yes	*0.08*	0.02	0.00	0.02	0.00
	No	0.92	0.99	*1.00*	0.98	*1.00*
Household size	≥6 persons	0.04	0.02	*0.10*	0.01	0.01
	<6 persons	0.96	0.98	0.90	0.98	*0.99*
Youth support uptake	Yes	*0.18*	0.01	0.04	0.03	0.00
	No	0.80	0.99	0.96	0.97	*1.00*
Self-reported health						
Perceived health status	Negative	**0.70**	0.12	0.10	0.20	0.00
	Positive	0.30	0.88	0.90	0.80	*1.00*
Long-term illness	Yes	**0.68**	0.32	0.09	0.28	0.06
	No	0.32	0.68	0.91	0.72	*0.94*
Restricted by health	Yes	**0.76**	0.28	0.16	0.29	0.04
	No	0.24	0.72	0.84	0.71	*0.96*
Healthcare expenditures and utilization						
Overall healthcare expenditures	High	**0.66**	**1.00**	0.16	0.05	0.04
	Low-average	0.34	0.00	0.84	0.95	*0.96*
General practitioners' (GP) expenditures	High	**0.68**	0.33	0.23	0.21	0.10
	Low-average	0.30	0.67	0.77	0.79	*0.90*
Hospital expenditures	High	0.30	**0.69**	0.08	0.00	0.00
	Low or none	0.70	0.31	0.92	*1.00*	*1.00*
Medication use	High	*0.54*	0.23	0.06	0.10	0.03
	Low or none	0.46	0.77	0.94	0.90	*0.97*
Addiction related care uptake	Yes	*0.06*	0.00	0.00	0.00	0.00
	No	0.94	*1.00*	*1.00*	0.99	*1.00*
Psychosocial characteristics						
Mental healthcare uptake	Yes	*0.32*	0.12	0.01	0.06	0.01
	No	0.68	0.88	*0.99*	0.94	*0.99*
Risk of depression or anxiety disorders	Moderate – high risk	**0.86**	0.46	0.56	**0.71**	0.21
	No or low risk	0.12	0.54	0.44	0.28	*0.79*
Loneliness	Feeling lonely	**0.68**	0.22	0.56	0.57	0.14
	Not feeling lonely	0.32	0.78	0.44	0.43	*0.86*
Feelings of control over life	Low	*0.24*	0.03	0.11	0.10	0.00
	Moderate	0.72	0.75	0.76	*0.81*	0.63
	High	0.02	0.22	0.13	0.09	*0.37*
Mild intellectual disability	Yes	*0.02*	0.00	0.01	0.00	0.00
	No	0.98	*1.00*	0.99	*1.00*	*1.00*
Life-events						
Crime suspect	Yes	*0.14*	0.01	0.03	0.03	0.01
	No	0.86	*0.99*	0.97	0.97	*0.99*
Crime victim	Yes	*0.34*	0.24	0.11	0.23	0.20
	No	0.66	0.77	*0.89*	0.77	0.80
Having been detained	Yes	*0.02*	0.00	0.00	0.00	0.00
	No	0.98	*1.00*	*1.00*	*1.00*	*1.00*
History of frequent moving	Yes	*0.42*	0.36	0.20	0.30	0.29
	No	0.56	0.64	*0.80*	0.70	0.71

Table 2. Continued.

	Class	1	2	3	4	5
	Label	Multi-dimen-sional vulne-rability	High care utili-zation	Socio-economic vulne-rability	Psycho-social vulne-rability	Healthy and socio-economi-cally stable
	Class proportions	0.06 (n=250)	0.11 (n=485)	0.09 (n=395)	0.24 (n=1005)	0.49 (n=2040)
Loss of a family member	Yes	*0.06*	0.05	0.01	0.03	0.03
	No	0.92	0.96	*0.99*	0.97	0.97
Living conditions						
Home ownership	Rented	**_0.64_**	0.10	0.58	0.36	0.10
	Owner occupied	0.34	*0.90*	0.42	0.64	*0.90*
Motorized vehicle ownership	No	*0.32*	0.07	0.29	0.13	0.06
	Yes	0.66	0.93	0.71	0.87	*0.94*
Proximity to general practitioners' (GP) office	>3 km	0.02	*0.08*	0.05	0.04	*0.08*
	<3 km	*0.98*	0.92	0.95	0.96	0.92
Liveability neighbourhood	Low-mediocre	0.18	0.03	*0.22*	0.09	0.03
	High	0.82	*0.97*	0.78	0.91	*0.97*

Proportions of risk factors (first category) >0.6 are shown in bold to indicate the higher occurrence of certain risk factors per class. For each category, the class with the highest proportion is shown in italic. Totals may not add up to 1.0 because of rounding. Following guidelines of Statistics Netherlands, the observed numbers in each category were rounded to five before calculating proportions in order to prevent the disclosure of information about individuals.
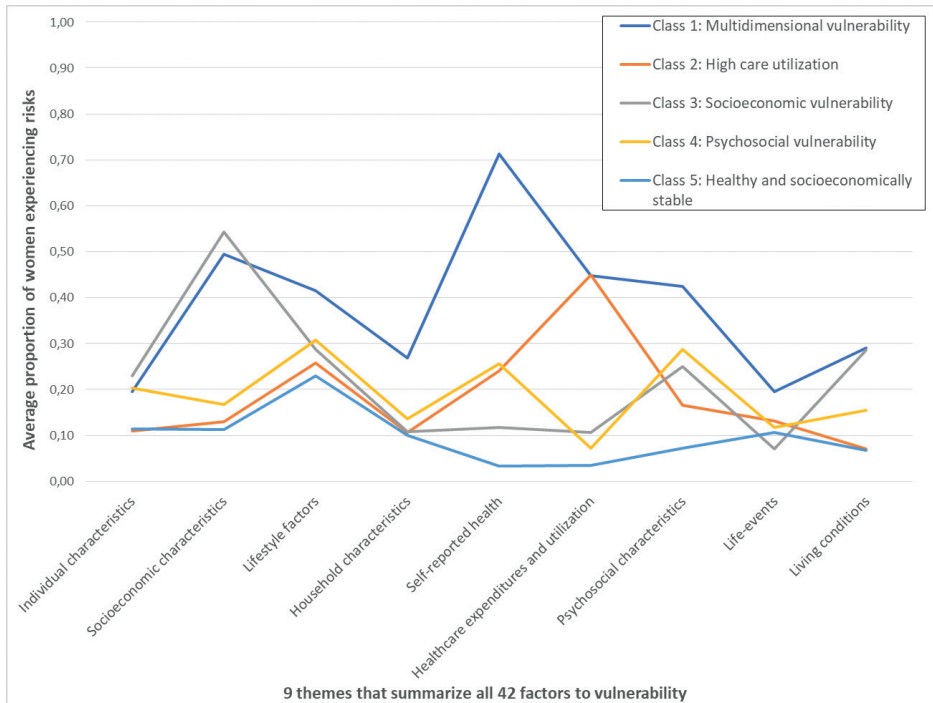
Figure 1. A visual representation of the five latent classes, described across the nine themes that summarize all 42 factors related to vulnerability. The vertical axis displays for each theme the average proportion of women within the categories that represent the risk factors (each first category in Table 2). A higher score means that a higher proportion of women in a class have risk factors to vulnerability. An example: the theme 'self-reported health' consists of three factors: perceived health, long-term illness and restriction by health. For Class 1 (multidimensional vulnerability), the average proportion of women with a negative perceived health (0.7), long-term illness (0.68) and feelings of being restricted by health (0.76), is 0.71. This average proportion is displayed.

Class 1 (n = 250; 6.0%), was characterized by high proportions of almost all risk factors to vulnerability. Women in this class were likely to receive social benefits or to have no income (proportion of 0.62) and to live in a rented house (0.65). Related to health, Class 1 was characterized by high GP healthcare expenditures (0.67), long-term illness (0.68) and negative perceptions of health (0.70). These women had a high probability of feeling lonely (0.87) and a moderate to high risk of depression or anxiety (0.87). Considering the vulnerabilities in different areas (including psychosocial, medical and socioeconomic risk factors), Class 1 was named 'multidimensional vulnerability'.

Class 2 (n = 485; 11.6%) was characterized by high healthcare expenditures. All women classified in this class had total healthcare expenditures in the highest quintile. Also, they frequently experienced high hospital care expenditures (0.69). Simultaneously, women in this class were likely to have protective factors including a healthy BMI (0.68), positive perception of health (0.87), high educational level (0.65), paid work (0.96), low probability of

feeling lonely (0.78) and an owner-occupied house (0.90). Based on the dominant features, Class 2 was named 'high care utilization'.

Class 3 (n = 395; 9.5%) was characterized in particular by high proportions of socioeconomic risk factors. Women in this class were likely to receive social benefits or have no income prior to pregnancy (0.87). They frequently lived in a rented house (0.58), had a non-Dutch background (0.56) and a low (0.30) or moderate (0.39) educational level. The probability of living in a neighbourhood with a low liveability score was highest in this class (0.22). When considering protective factors, these women were often married (0.70), had a positive perception of health (0.90) and low healthcare expenditures (0.83). Class 3 was named 'socioeconomic vulnerability'.

Class 4 (n = 1005; 24%) was characterized by psychosocial health issues. The majority had a moderate to high risk of depression or anxiety disorders prior to pregnancy (0.71). These women were likely to feel lonely (0.57) and nullipara were overrepresented (0.55). Regarding protective factors, the majority had a full-time contract (0.69), an owner-occupied house (0.64) and no high healthcare expenditures (0.95). Class 4 was named 'psychosocial vulnerability'.

Class 5 (n = 2040; 48.9%) was characterized by women with low probabilities of all risk factors to vulnerability before pregnancy. Instead, in general, these women had a positively perceived health (1.00), did not feel lonely (0.86), had a high educational level (0.70) and paid work (0.98). Women in Class 5 had the highest probability to experience high control over life (0.37). Class 5 was named 'healthy and socioeconomically stable'.

The analyses in the two additional study populations (women who gave birth before and all women aged 19-44 years) showed similar results. The five-class model was preferred and classes could be interpreted similarly.

Figure 2 shows associations between classes and adverse outcomes. Class 5 (healthy and socioeconomically stable) was the reference-category. Women classified in Class 1 (multidimensional vulnerability) were more likely to have babies who were born prematurely, SGA or admitted to a NICU. These women were also more likely to have a caesarean section. There were no significant associations found for other maternal health outcomes including hypertension/pre-eclampsia and postpartum haemorrhage. Compared to Class 5 (healthy and socioeconomical stable), all other classes except Class 4 (psychosocial vulnerability) were more likely to not receive postpartum care (at home) and to not receive antenatal care on time. Adverse outcomes were quite similar in Class 2 (socioeconomic vulnerability) and Class 5 (healthy and socioeconomically stable), except from the odds of planned caesarean section. Appendix 4 shows prevalences of outcomes for each class.
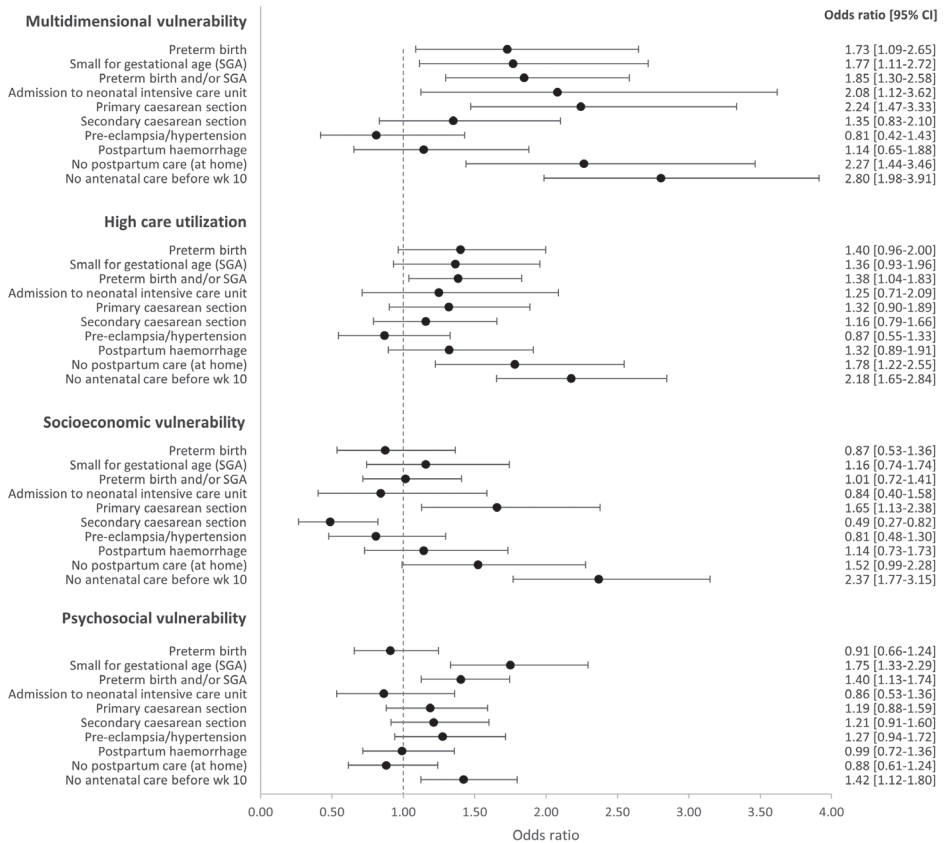
Figure 2. Likelihood of adverse perinatal and maternal health outcomes and healthcare utilization for four classes, compared to the reference-category 'healthy and socioeconomically stable'. The figure shows the OR and 95% CI (graphically and in text)

DISCUSSION

This study aimed to identify classes of vulnerability among pregnant women and to validate these classes by studying the association with adverse perinatal and maternal health outcomes and care utilization. The LCA procedure identified five classes with different combinations of risk and protective factors to vulnerability. Most women were classified into the 'healthy and socioeconomically stable' class with mainly protective factors. Women classified in the classes 'high care utilization', 'socioeconomic vulnerability' or 'psychosocial vulnerability' shared risk factors to vulnerability in one specific domain and protective factors in others. Women classified into the 'multidimensional vulnerability' class shared multiple risk factors in several domains (e.g., psychosocial, medical and socioeconomic) and were more likely to develop poor health outcomes such as premature birth, SGA, caesarean section and NICU admission.

Our study showed that multidimensional vulnerability leads to experiencing worse outcomes compared to vulnerability on a single domain or no vulnerabilities. This indicates the importance of co-existence or clustering of multiple risk factors (such as no income, high healthcare expenditures and feelings of loneliness) in increasing the probability of adverse outcomes for mother and child. Our findings strengthen results from previous studies that aimed to explain differences in adverse outcomes by interrelated individual or contextual risk factors (10, 11, 17). Previous LCA studies also led to classes of pregnant women with different health behaviours, psychosocial or socioeconomic characteristics that show differences in outcomes, although these studies included less factors and domains, and other populations in comparison to our study (17, 32, 33). The findings do not inform us on how risk factors interplay and lead to adverse health outcomes. The syndemic model provides a perspective on this interplay by describing how co-occurring health adversities are fuelled by different social and contextual factors that interact and increase the health burden of both mental and physical illness (34). This suggests the need to combine social and medical care and support, instead of focussing on the separate domains to combat multidimensional vulnerability.

We found that women with socioeconomic vulnerability generally did not experience worse outcomes. This finding is not in congruence with previous research indicating that adverse perinatal health outcomes are more prevalent among women with a low socioeconomic status (SES) (9, 10, 14). Previous studies often focussed on a limited number of risk factors or domains, or used more traditional (regression) techniques to study the relation between SES and outcomes. However, as the impact of risk factors can depend on other factors, it is important to step away from traditional independent 'ceteris paribus' linear effect assumption of social determinants. Therefore, we used LCA as analytical approach that considers the combination of both risk and protective factors, allowing a more comprehensive approach to study vulnerability. Protective factors (e.g. social support) can act as positive exposures or buffering mechanisms that promote resilience and improve health (3, 8, 35, 36). This indicates the importance of acknowledging both strengths and challenges in families to create a supportive environment for early development (37). Additionally, low SES may not necessarily be a risk factor for adverse outcomes unless it coincides with other hardships. The relation between SES and health can be described by processes such as social causation (adverse conditions of poverty impact health through, for example, stress and food insecurity) and health selection (people with worse physical or mental health outcomes fall into poverty through, for example, stigma, health expenditures and lower productivity) (38). This increases the importance for healthcare professionals to understand different domains of vulnerability and tailor the need for support to the individual (39, 40).

Our findings reveal a difference in care utilization patterns. The 'healthy and socioeconomically stable' class was most likely to receive early antenatal care and postpartum care (at home). This corresponds to findings of Grabovschi et al. (6) in their scoping review into vulnerability. People with higher vulnerability levels (i.e. multiple vulnerability aspects) have higher

healthcare needs, but less access to services and lower quality of healthcare. This raises questions about whether current support meets parents' needs.

The main strength of this study is that we linked routinely collected nationwide observational data sources to self-reported data on health, well-being and lifestyle. This offered the opportunity to include data on a wide range of medical and social factors for a large group of pregnant women to better understand vulnerability. While previous studies often had a unidimensional perspective to vulnerability (focussing on single risk factors such as individual SES, or neighbourhood SES on aggregated level), we could unravel the difference between unidimensional and multidimensional types of vulnerability due to our extensive dataset. Another strength is that we included protective factors, while most studies focus primarily on factors that increase the risk of adverse outcomes and less on protective factors that might counteract these effects (18, 19). Unfortunately, data on topics such as nutrition, stress, health literacy, preconception care and adverse childhood experiences were not available, while these factors could provide additional insights into vulnerability. Next, using largest posterior probability to assign women to classes is a limitation, because not all women are fully representative of one class only. Our study was moreover limited by not including the father or woman's partner, despite growing evidence of their importance in promoting healthy pregnancy, childbirth and child-outcomes. Another limitation relates to the representativeness of the study population due to using the PHM-2016. Compared to all other pregnant women in 2017/2018, women in our study less often had a low income (5% vs 8%), low educational level (8% vs 12%) and migration background (18% vs 32%). Since generally people with higher vulnerability less often participate in research, we assume that the size of the multidimensional vulnerability-class is an underestimation. Nevertheless, since we could identify classes of vulnerability and differentiate between single and multidimensional vulnerability, we expect that their characteristics are also applicable beyond the study population. Similar results from our additional analyses strengthen this expectation. Nevertheless, our approach and findings should be validated in other cohorts and countries and until then be interpreted with caution.

Our findings can have several implications for practice and research. We believe that screening instruments for vulnerability before and during pregnancy could benefit from including a balanced set of both risk and protective factors. In refining screening instruments, we have to consider the various criteria for responsible screening, such as the availability of associated care or support strategies (41). Greater consciousness among healthcare providers regarding the complexity of vulnerability in terms of risk and protective factors and personal perceptions could enhance the provision of person-centred care and support (6, 40, 42). Multiple studies argue that future strategies should also pay attention to underlying, root causes of vulnerability in policies, laws and governance (3, 15, 43). Advancing health equity requires both individual-level interventions targeted at vulnerable individuals as well as systemic-level change (3, 15, 43). Factors related to housing, education and social security for example, frequently lie upstream of individual lifestyle and behavioural factors modifiable through individual-level interventions. Findings of our study can be input for longitudinal monitoring of vulnerability at population level. Future

research is needed to identify if vulnerability classes can be identified using solely routinely collected population data, without using self-reported data. Additionally, more research is necessary regarding the role of the father or woman's partner in relation to vulnerability.

In conclusion, there is growing attention for early detection of vulnerability and implementing effective strategies to improve health and well-being of current and next generations. Results of this data-driven study suggest that several vulnerability classes can be distinguished among pregnant women in the Netherlands. The co-existence of risk factors in multiple domains leads to more adverse outcomes for mother and child. Effective strategies, starting preconceptionally, should include both medical and social care and support.

Data availability
We are unable to share the individual data used for this study as data linkage and analysis was conducted within the highly safeguarded Remote Access (RA) platform of Statistics Netherlands (23). All data within this platform are pseudonymised to ensure data safety and confidentiality. Access to the data from Perined, Vektis, Statistics Netherlands and the Public Health Monitor 2016 can be requested from the relevant parties.

Key points

- Previous evidence supports the influence of social factors on maternal and perinatal health, but few studies consider the combination of different social risk and protective factors to vulnerability.
- Pre-pregnancy data of 4172 women on a wide range of social risk and protective factors to vulnerability were used to identify latent vulnerability classes.
- Five classes could be distinguished: multidimensional vulnerability, high care utilization, socioeconomic vulnerability, psychosocial vulnerability and a healthy and socioeconomically stable-class.
- Multidimensional vulnerability, characterized by experiencing risk factors in different domains and few protective factors, was associated with adverse outcomes for mother and child, while experiencing risk factors solely in one domain was not necessarily associated with adverse outcomes.
- Public health programmes should start preconceptionally, include both medical and social care and support, and be attentive to systemic causes of vulnerability

2

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APPENDIX 1.
Description of data sources, variables and outcomes included

Data sources

The Dutch Perinatal Registry Perined collects routine care data on pregnancy after 22 weeks of gestation, birth and the first 28 days after birth, as supplied by midwives, gynaecologists and paediatricians.[1] Perined includes data on type of care used and health outcomes of mother and child. From 2000 onwards, Perined contains data on 96 – 99% of all births.

Healthcare information centre Vektis collects claims data under the Dutch Healthcare Insurance Act and provides data on healthcare utilization and spending.[2] Data on 99% of the Dutch (insured) population is available. DIAPER contains detailed claims data of Vektis as available from 2015.

Statistics Netherlands collects and publishes data on societal matters and provides access to data through their System of Social Statistical Datasets (SSD).[3,4] This linkable SSD-data covers nearly 20 themes, including health, welfare, income, education and labour. Data originate from various (governmental) organisations, including municipalities and the National Tax Authority.

We enriched DIAPER with self-reported data on health, well-being and lifestyle of the Public Health Monitor 2016 (PHM-2016). This is a health survey among a varying sample of the Dutch population aged 19 years and older, carried out every four years by the Community Health Services (GGD), Statistics Netherlands and the National Institute for Public Health and the Environment. The PHM-2016 had 457.153 participants and was mainly conducted from September-December 2016.

1   Perined. Over Perined [about Perined] n.d. Available from: https://www.perined.nl/over-perined.
2   de Boo A. Vektis 'Informatiecentrum voor de zorg' [Vektis 'Information center for healthcare']. Tijd-schrift voor gezondheidswetenschappen. 2011;89(7):358-9
3   Bakker BF, Van Rooijen J, Van Toor L. The system of social statistical datasets of Statistics Netherlands: An integral approach to the production of register-based social statistics. Statistical Journal of the IAOS. 2014;30(4):411-24.
4   Statistics Netherlands (CBS). About CBS. n.d. Available from: https://www.cbs.nl/en-gb/about-us/.

Variables in LCA

Variable	Definition	Categories	Data source*
Individual characteristics			
Age	Age of the woman in categories, based on an increased risk for pregnancy complications related to age[5,6]	19– 23; 24 – 35; >35	SSD
Ethnicity	Migration status based on birthplace of parents, following categorization of Statistics Netherlands.[7] A person with a migration background is a person of whom at least one parent was born abroad	Non-Western; Western; Native Dutch	SSD
Parity	The number of times that a woman has given birth after a gestational period of 24 weeks or more: 0 times (nullipara), 1 time (primipara), 2 or more times (multipara)	Nullipara; primipara, multipara	Perined
Asylum seeker status	Registered as asylum seeker, status holder or refugee and included in the municipal population register	Yes; no	SSD
Socioeconomic characteristics			
Educational level	Highest completed education, according to the International Standard Classification of Education and following categorization of Statistics Netherlands[8]	Low; moderate; high	PHM-2016 and SSD
Household income	Household income based on percentage groups of standardized disposable income of private households; low is <10th percentile, moderate is 10th – 90th percentile and high is >90th percentile	Low; moderate; high	SSD
Socioeconomic position	Source of income, if any	No income/ receiving benefits; student; paid work	SSD
Debts and payment arrears	Difficulty paying bills, based on registration of debt restructuring and/or delayed health insurance payments for more than six months	Yes; no	SSD
Insufficient financial resources**	Having insufficient means, based on the question: 'in the past 12 months, have you struggled to make ends meet on your household income?'	Yes; no	PHM-2016
Permanent contract	Having a permanent type of work contract	No; yes	SSD
Full-time contract	Being full-time employed	No; yes	SSD
Lifestyle factors			
Smoking**	Based on the question: 'do you ever smoke?'	Yes; no	PHM-2016
Alcohol use	Alcohol use, categorized as excessive based on the number of glasses of alcohol in a week (>7), following Dutch 'Guidelines for Good Nutrition'[9]	Yes (excessive); no	PHM-2016

5   Londero, A.P., Rossetti, E., Pittini, C. et al. Maternal age and the risk of adverse pregnancy outcomes: a retrospective cohort study. BMC Pregnancy Childbirth 19, 261 (2019).

6   FIOM. Jong ouderschap. Available from: https://fiom.nl/kenniscollectie/jong-ouderschap

7   Statistics Netherlands (CBS). Person with a migration background. Available from: https://www.cbs.nl/en-gb/our-services/methods/definitions/person-with-a-migration-background

8   Statistics Netherlands (CBS). Standaard onderwijsindeling 2016 (The Dutch standard classification of education). Available from: https://www.cbs.nl/nl-nl/onze-diensten/methoden/classificaties/onderwijs-en-beroepen/standaard-onderwijsindeling--soi--/standaard-onderwijsindeling-2016

9   Gezondheidsraad [Dutch Health Council]. Richtlijnen goede voeding 2006 [Guidelines good nutrition 2006]. Den Haag: Gezondheidsraad, 2006.

Variable	Definition	Categories	Data source*
Physical activity	Compliance with guidelines for physical activity (>150 minutes of moderate to vigorous intensive activities every week, as well as muscle and bone strengthening activities) as measured by the SQUASH-questionnaire[10]	Less than recommended; as recommended or more	PHM-2016
Body Mass Index (BMI)	BMI based on height and weight. According to the International Obesity Task Force, BMI <15 is underweight, BMI 18,5 – 25 is normal weight and BMI ≥25 overweight[11]. Underweight and overweight are categorized into 'unhealthy BMI'. Normal weight is categorized into 'healthy BMI'	Unhealthy BMI; healthy BMI	PHM-2016
Household characteristics			
Type of household	Type of household was categorized based on registrations of persons living at the same home address. Women without a registered partner were classified as a one-person household (without children) or a one-parent household (with children). All other types were categorized into 'other'	One-person/ parent household; other	SSD
Marital status	Marital status of the woman	Unmarried; married	SSD
Dissolution of marriage***	Having been divorced [2012-2016]	Yes; no	SSD
Household size	Number of persons registered at the same home address	≥6 persons; <6 persons	SSD
Youth support uptake***	Previously received youth support for children within the family [2015-2016]	Yes; no	SSD
Self-reported health			
Perceived health status	Based on the question: 'how is your health in general?' and categorized into negative (very bad or bad) and positive (very good, good or moderate)	Negative; positive	PHM-2016
Long-term illness	Based on the question: 'do you have one or more long-term (≥ 6 months) illnesses or disorders?'	Yes; no	PHM-2016
Restricted by health	Based on the question 'to what extent have you been limited in activities due to health for the last 6 months or longer?' into yes (severely limited or limited) and no (not limited)	Yes; no	PHM-2016
Healthcare expenditures and utilization			
Overall healthcare expenditures	Total healthcare expenditures in 2016 subdivided into quintiles and divided into high (quintile 5) and low-average (quintile 1 to 4). In case a woman was pregnant in 2016, total healthcare expenditures of the previous year (2015 or 2014) were included	High; low-average	SSD (Vektis)
General practitioners' (GP) expenditures	GP expenditures in 2016 subdivided into quintiles and divided into high (quintile 5) and low-average (quintile 1 to 4)	High; low-average	SSD (Vektis)

10 Gezondheidsraad [Dutch Health Council]. Beweegrichtlijnen 2017 [exercise guidelines 2017]. Den Haag: Gezondheidsraad, 2017.

11 Cole TJ, Bellizzi MC, Flegal KM, Dietz WH. Establishing a standard definition for child overweight and obesity worldwide: international survey. BMJ. 2000;320(7244):1240-3.

Variable	Definition	Categories	Data source*
Hospital expenditures	Hospital expenditures in 2016, categorized using a cut-off value corresponding to the highest 10% of expenditures among all women with hospital expenditures. In case a woman was pregnant in 2016, hospital expenditures of the previous year (2015 or 2014) were included, to avoid including pregnancy related health care costs	High; low or none	SSD (Vektis)
Medication use	Number of registered types of medication used in 2016 categorized into high (≥5 different types) and low (<5) according to the most common definition of polypharmacy[12]	High; low or none	SSD
Addiction related care uptake ***	The presence or history of a diagnosis-treatment combination for mental healthcare related to addiction [2011-2016]	Yes; no	SSD
Psychosocial characteristics			
Mental healthcare uptake	Expenditures that were made regarding mental healthcare services	Yes; no	SSD (Vektis)
Risk of depression or anxiety disorders**	Based on ten questions of the Kessler Psychological Distress scale[13], categorized into moderate to high risk and no or low risk	Moderate-high risk; no or low risk	PHM-2016
Loneliness**	Based on 11 statements of the De Jong – Gierveld scale[14], categorized into feeling lonely (moderate – severe) and not feeling lonely	Feeling lonely; not feeling lonely	PHM-2016
Feelings of control over life**	Based on seven statements[15] of the Pearlin Mastery scale and categorized into low (insufficient), moderate and high	Low; moderate; high	PHM-2016
Mild intellectual disability	Registered indication for a mild intellectual disability as recorded in data sources of occupational disabilities, sheltered employment and entitlement to the Long-Term care Act	Yes; no	SSD
Life-events			
Crime suspect***	Having been registered as a crime suspect [2009-2016]	Yes; no	SSD
Crime victim***	Having been a victim of a crime that was reported to the police [2011-2016]	Yes; no	SSD
Having been detained***	Having been detained [2004-2016]	Yes; no	SSD
History of frequent moving	A change of registered home address of more than five times in the past five years	Yes; no	SSD
Loss of a family member***	Loss of a parent and/or child [2011-2016]	Yes; no	SSD
Living conditions			
Home ownership	The home ownership of the woman's home address	Rented; owner-occupied	SSD
Motorized vehicle ownership	A registered motorized vehicle at the woman's home address, according to the Dutch vehicle registration authority	No; yes	SSD

12 Masnoon N, Shakib S, Kalisch-Ellett L, Caughey GE. What is polypharmacy? A systematic review of definitions. BMC geriatrics. 2017;17(1):1-10.

13 Kessler RC, Andrews G, Colpe LJ, Hiripi E, Mroczek D, et al. Short screening scales to monitor population prevalences and trends in non-specific psychological distress. Psychological medicine. 2002;32(6):959.

14 De Jong Gierveld J, Van Tilburg T. Manual of the loneliness scale 1999. Department of Social Research Methodology, Vrije Universiteit Amsterdam, Amsterdam (updated version 1801 02). 1999.

15 Pearlin LI, Schooler C. The structure of coping. Journal of health and social behavior. 1978:2-21.

Variable	Definition	Categories	Data source*
Proximity to GP office	Distance (by road) between registered home address and the nearest GP's office	>3 km; <3 km	SSD
Liveability neighbourhood	According to the Dutch Leefbaarometer[16], based on various elements including housing characteristics, residents, distance to services, safety and physical environment	Low-mediocre; high	SSD

* SSD and Perined are nationwide observational data sources, the Public Health Monitor 2016 (PHM-2016 contains self-reported data.

** The data of the PHM-2016 is collected through a combination of the regular Health Survey of Statistics Netherlands (98% of the study population) and the health surveys of the Municipal Health Services (2% of the study population). The variables indicated by the asterisks were not included in the regular Health Survey of Statistics Netherlands.

*** The number of included years was based on data-availability, which varied per item.

Outcomes

Outcome	Definition	Categories	Data source
Preterm birth	Birth occurring from 24 weeks of gestation and before 37 weeks of gestation	Yes; no	Perined
Small for gestational age (SGA)	Birth weight below 10th percentile, corrected for gestational age and fetal sex[17]	Yes; no	Perined
Preterm birth and/ or SGA	Baby born prematurely and/or SGA, following definitions above	Yes; no	Perined
Admission to neonatal intensive-care unit (NICU)	Admission to the Neonatal Intensive Care Unit after birth	Yes; no	Perined
Primary caesarean section	Birth by caesarean section, decision taken before the start of birth	Yes; no	Perined
Secondary caesarean section	Birth by caesarean section, decision taken during birth	Yes; no	Perined
Pre-eclampsia/ hypertension	Having pre-eclampsia or maternal hypertension (high blood pressure), chronic or pregnancy induced	Yes; no	Perined
Postpartum haemorrhage	Heavy bleeding after birth (>1000 ml blood loss)	Yes; no	Perined
No postpartum care (at home)	Woman does not receive postpartum care at home by a maternity care assistant[18]	No postpartum care; postpartum care	Vektis
No antenatal care before week 10	Not having the first antenatal care appointment (i.e., booking visit) before the 10th week of pregnancy	No antenatal care before week 10; antenatal care before week 10	Perined

16 Leidelmeijer, K., Marlet, G., Ponds, R., Schulenberg, R., van Woerkens, C., & van Ham, M. V. M. Leefbaarometer 2.0: instrumentontwikkeling. Rigo Research en Advies & Atlas voor de gemeenten. 2014

17 Hoftiezer, L., Hof, M. H., Dijs-Elsinga, J., Hogeveen, M., Hukkelhoven, C. W., & van Lingen, R. A. From population reference to national standard: new and improved birthweight charts. American journal of obstetrics and gynecology. 2019; 220(4): 383.

18 Laureij, L. T., van der Hulst, M., Lagendijk, J., Been, J. V., Ernst-Smelt, H. E., Franx, A., & Lugtenberg, M. Insight into the process of postpartum care utilization and in-home support among vulnerable women in the Netherlands: an in-depth qualitative exploration. BMJ open. 2021; 11(9): e046696.

APPENDIX 2.
Details on the multiple imputation process

The missing data (range missing values per variable: 0 to 9.1%) for the latent class analysis were imputed using multiple imputation[19]. We created five imputed datasets in R through the package Multiple Imputation using Chained Equations (MICE)[20]. Model development was done in each set and the outcomes were compared. The outcomes were highly similar: they let to the same conclusions for both fit-statistics and class-specific probabilities. Subsequent analyses and the presentation of results was done for one randomly chosen imputed dataset.

	Multiple Imputation
Software used	R version 3.6.2
Imputation method and key settings	Fully conditional specification (package mice version 3.13.0); maximum iterations: 5
Number of imputed datasets created	5
Analyses variables	Age; Ethnicity; Parity; Asylum seeker status; Educational level; Household income; Socioeconomic position; Debts or payment arrears; Insufficient financial resources*; Permanent contract; Full-time employment; Smoking*; Alcohol use; Physical activity; Body Mass Index (BMI); Type of household; Marital status; Dissolution of marriage; Household size; Youth support; Perceived health status; Long-term illness; Restricted by health; Total healthcare expenditures; General Practitioners' expenditures; Hospital expenditures; Medication use; Addiction related care uptake; Risk of depression or anxiety disorders*; Loneliness*; Feelings of control over life*; Mental healthcare use; Mild intellectual disability; Crime suspect; Crime victim; Having been detained; Frequent moving; Loss of a family member ; Home ownership; Motorized vehicle ownership; Proximity to the GP's office; Liveability of the neighbourhood
Auxiliary variables	11 variables for loneliness (statements + sum scores); 9 variables for control over life (statements + sum scores); 11 variables for depression or anxiety disorders (statements + sum scores); 2 variables for alcohol use; language of questionnaire; 7 variables for noise nuisance; 2 variables for socioeconomic status; 3 variables for crime victim; 2 variables for addiction related care uptake; 2 variables for loss of family member; 2 variables for insufficient financial resources; number of movements; number of life-events.
Treatment of continuous data	Predictive mean matching
Treatment of binary data	Logistic regression
Treatment of unordered categorical data	Polytomous logistic regression
Population	For the imputation we used additional data from the Public Health Monitor 2016 and Statistics Netherlands (SSD). There were 3043 complete cases and 1129 women with missing data. These women mostly had missing data on one variable (n = 641).

* The data of the Public Health Monitor 2016 is collected through a combination of the regular Health Survey of Statistics Netherlands (98% of the study population) and the health surveys of the Municipal Health Services (2% of the study population). The variables indicated by the asterisks were not included in the regular Health Survey of Statistics Netherlands and thus contain missing values.

19 Sterne JA, White IR, Carlin JB, et al. Multiple imputation for missing data in epidemiological and clinical research: potential and pitfalls. Bmj. 2009;338:b2393.
20 Buuren van S, Groothuis-Oudhoorn K. mice: Multivariate Imputation by Chained Equations in R. Journal of Statistical Software. 2011;45:1-67.

APPENDIX 3.

Fit results of LCA model with increasing number of classes

Model*	Akaike Information Criterion (AIC)	Relative decrease AIC	Size adjusted Bayesian Information Criterion (aBIC)	Relative decrease aBIC	Bayesian Information Criterion (BIC)	Relative decrease BIC	Entropy
Model 1	144228.5	*NA*	144380.1	*NA*	144532.6	*NA*	NA
Model 2	137476.2	*-0.047*	137782.5	*-0.046*	138090.8	*-0.045*	0.807
Model 3	135647.8	*-0.013*	136108.9	*-0.012*	136572.8	*-0.011*	0.781
Model 4	134443.5	*-0.009*	135059.5	*-0.008*	135679.1	*-0.007*	0.803
Model 5	133396.3	*-0.008*	134167	*-0.007*	134942.3	*-0.005*	0.784
Model 6	132491.5	*-0.007*	133417	*-0.006*	134348	*-0.004*	0.771
Model 7	131736.2	*-0.006*	132816.5	*-0.005*	133903.2	*-0.003*	0.774
Model 8	131271.7	*-0.004*	132817	*-0.002*	133749.1	*-0.001*	0.680
Model 9	130858.8	*-0.003*	132506.7	*-0.002*	133646.7	*-0.001*	0.757
Model 10	130494.3	*-0.003*	132248.6	*-0.002*	133592.7	*0*	0.645
Model 11	130226.1	*-0.002*	132038.8	*-0.002*	133635	*0*	0.625
Model 12	130006.5	*-0.002*	131925.4	*0*	133725.8	*0.001*	0.537
Model 13	129830.9	*-0.001*	131860.6	*0*	133860.7	*0.001*	0.524
Model 14	129699.4	*-0.001*	131863	*0*	134039.7	*0.001*	0.531
Model 15	129599.9	*-0.001*	131918.3	*0*	134250.7	*0.002*	NA

*The model number reflects the number of classes included in each model. For example, model 5 reflects a model with 5 classes.

APPENDIX 4.

Prevalence of outcomes for the five latent classes

	Multi-dimensional vulnerability	High care utili-zation	Socio-economic vulnerability	Psycho-social vulnerability	Healthy and socio-economi-cally stable
Preterm birth	10.4%	8.6%	5.6%	5.8%	6.3%
Small for gestational age (SGA)	10.5%	8.3%	7.1%	10.4%	6.2%
Preterm birth and/or SGA	19.4%	15.3%	11.7%	15.5%	11.6%
Admission to neonatal intensive care unit	6.0%	3.7%	2.5%	2.6%	3.0%
Primary caesarean section	13.3%	8.2%	10.1%	7.5%	6.4%
Secondary caesarean section	9.2%	8.0%	3.5%	8.4%	7.0%
Pre-eclampsia/hypertension	4.8%	5.1%	4.8%	7.4%	5.9%
Postpartum haemorrhage	6.8%	7.8%	6.8%	6.0%	6.0%
No postpartum care (at home)	11.2%	9.1%	7.8%	4.7%	5.3%
No antenatal care before week 10	25.3%	20.9%	22.3%	14.7%	10.8%

3

Predicting population-level vulnerability among pregnant women using routinely collected data and the added relevance of self-reported data

Joyce M. Molenaar
Ka Yin Leung
Lindsey van der Meer
Peter Paul F. Klein
Jeroen N. Struijs
Jessica C. Kiefte-de Jong

ABSTRACT

Background

Recognizing and addressing vulnerability during the first thousand days of life can prevent health inequities. It is necessary to determine the best data for predicting multidimensional vulnerability (i.e. risk factors to vulnerability across different domains and a lack of protective factors) at population-level to understand national prevalence and trends. This study aimed to 1) assess the feasibility of predicting multidimensional vulnerability during pregnancy using routinely collected data, 2) explore potential improvement of these predictions by adding self-reported data on health, wellbeing and lifestyle, and 3) identify the most relevant predictors.

Methods

The study was conducted using Dutch nationwide routinely collected data and self-reported Public Health Monitor data. First, to predict multidimensional vulnerability using routinely collected data, we used Random Forest (RF) and considered the Area Under the Curve (AUC) and F1-measure to assess RF-model performance. To validate results, sensitivity analyses (XGBoost and Lasso) were done. Second, we gradually added self-reported data to predictions. Third, we explored the RF-model's variable importance.

Results

The initial RF-model could distinguish between those with and without multidimensional vulnerability (AUC 0.98). The model was able to correctly predict multidimensional vulnerability in most cases, but there was also misclassification (F1-measure 0.70). Adding self-reported data improved RF-model performance (e.g. F1-measure 0.80 after adding perceived health). The strongest predictors concerned self-reported health, socioeconomic characteristics and healthcare expenditures and utilization.

Conclusions

It seems possible to predict multidimensional vulnerability using routinely collected data that is readily available. However, adding self-reported data can improve predictions.

INTRODUCTION

A strong foundation during the first thousand days of life, which span from conception till a child's second birthday, can positively impact health and development in later life and across generations (1, 2). Adverse experiences and exposures can influence the health of parents themselves, but can also be transmitted to their children, which, as these children grown into adulthood and potentially become parents themselves, leads to new cycles of adversity (2). In order to prevent health inequities and break the intergenerational cycle, it is important to recognize and address vulnerability during the first thousand days (1-3). This is also a focus in the Dutch nationwide action program Solid Start (in Dutch: Kansrijke Start) (4). The concept of vulnerability is often used to describe subgroups with increased risks to adverse health outcomes or limited access to healthcare. In short, vulnerability encompasses a multifaceted and dynamic process in which diverse stressors at the individual or contextual level can serve as risk factors, whereas protective factors have the potential to mitigate or prevent vulnerability (5-9). Examples of risk factors encompass unemployment or stress, while examples of protective factors include a strong social network or effective coping skills.

The concept of vulnerability and its scope has garnered increasing attention among providers and policymakers who strive to enhance the provision of care and support during the first thousand days of life (4, 9, 10). In daily care, a common understanding between professionals from the medical and social sector on the characteristics of high-risk individuals can foster mutual understanding and improve cross-sectoral collaboration (9). At national and local policy levels, drawing attention to the prevalence, geographical distributions and trends in vulnerability can support policy monitoring and prioritization. These insights not only foster a sense of urgency, but also enhance the conversation between different stakeholders, and facilitate vision formulation and intervention prioritization (11).

This study extends our prior research to predict population-level vulnerability among pregnant women. Our previous study highlighted the significance of considering both risk and protective factors, particularly in the context of adverse outcomes (12). Through Latent Class Analysis (a data-driven technique to identify subgroups with similar characteristics), we identified five groups of pregnant women with different social risk and protective factors to vulnerability prior to pregnancy. Women in the 'multidimensional vulnerability'-group shared multiple risk factors across several domains (e.g. psychosocial, medical, and socioeconomic), lacked protective factors and were most at risk of adverse outcomes such as premature birth and caesarean section. Having risk factors in a single domain (e.g. socioeconomic) was not necessarily associated with adverse outcomes. This study utilized both routinely collected observational data and self-reported data on health, wellbeing and lifestyle of the Public Health Monitor 2016 (PHM-2016) to predict multidimensional vulnerability (12). Using the PHM-2016 resulted in a subset of the total Dutch pregnant population. Hence, the prevalence of multidimensional vulnerability across the entire population of pregnant women at a national level remains unknown, and it is unclear

whether this can be assessed using solely routinely collected observational data and what the added value of self-reported data is. Moreover, we lack an understanding of the strongest predictors for population-level vulnerability.

Mapping out the percentage of multidimensional vulnerability among pregnant women in the Netherlands and its predictors is relevant for risk stratification. In population health management, this is an essential initial step to tailor (preventive) actions to the needs of specific risk-groups to enhance population health (13, 14). Such stratification commonly relies on routinely collected data (15), offering advantages such as widespread availability, reduced practitioner burden, time and costs (16, 17). Moreover, the longitudinal and systematic approach facilitates comparisons over time (16, 17). However, it is important to empirically evaluate whether routinely collected data is sufficient for risk-stratification for high-risk groups. In addition, there is a potential for improvement in predicting multidimensional vulnerability at population-level by incorporating self-reported health, well-being and lifestyle data. For example, studies indicate that self-reported health and vulnerability correspond to or complement clinical measures in predicting adverse health outcomes (18-22). Yet, the impact of adding self-reported data next to routinely collected data in predicting vulnerability remains unexplored.

This study has three objectives. First, to assess the feasibility of accurately predicting multidimensional vulnerability during pregnancy at population-level using solely routinely collected observational data. We use the predictions to report on the prevalence and spatial variation of multidimensional vulnerability during pregnancy at population-level in the Netherlands. Second, to identify whether self-reported data on health, wellbeing, and lifestyle could improve those predictions with routinely collected data. Third, to identify the predictors that have the most significant impact on the classification of multidimensional vulnerability.

METHODS

Data sources
This study employed data from DIAPER (Data-InfrAstructure for ParEnts and childRen) (17). DIAPER integrates individual-level, routinely collected observational data from various nationwide data sources in the Netherlands, including Perined and Statistics Netherlands. Perined collects routine care data on pregnancy, birth, and the first 28 days after birth from midwives, gynaecologists, and paediatricians (23). Statistics Netherlands collects data about social issues, including health, welfare, income, education, and labour (24, 25). To enrich DIAPER, self-reported data on health, well-being, and lifestyle of the PHM-2016 were included (26). The PHM is a health survey conducted every 4 years among a varying sample of Dutch adults aged 19 years and older (about 450.000 in 2016).

Study population

The study population consisted of 4172 unique women with a pregnancy and childbirth in 2017 or 2018 who participated in the PHM-2016 prior to pregnancy. Details on selecting the study population are described in Chapter 2 (12). To illustrate the prevalence and spatial variation of multidimensional vulnerability at national level, all unique registered pregnancies in Perined from 2017 to 2021 were considered (n = 807.904) (17). Missing data were imputed through Multiple Imputation using Chained Equations (MICE), leading to five imputed datasets (27).

Independent variables

Analogous to our previous study, we included 42 variables in the predictive models (12). Details on these variables have been described in Appendix 1 of our previous study (Chapter 2) (12). The first category in each variable denotes the risk factor to vulnerability.

Of those 42 variables, 31 variables concerned routinely collected data available for all pregnant women in DIAPER (n = 807.904). Those included individual characteristics (age, ethnicity, parity, asylum seeker status), socioeconomic characteristics (educational level, household income, socioeconomic position by occupational status, debts and payment arrears, permanent employment contract, and full-time employment contract), household characteristics (type of household, marital status, dissolution of marriage, household size, and youth support utilization), healthcare expenditures and utilization (total healthcare expenditures, General Practitioner's (GP) expenditures, hospital expenditures, medication use, and addiction related care utilization), psychosocial characteristics (mental healthcare utilization, mild intellectual disability), life events (crime suspect, crime victim, having been detained, frequent moving, loss of a family member), living conditions (home ownership, motorized vehicle ownership, proximity to GP office, liveability neighbourhood).

The other 11 variables were derived from the PHM-2016 and consequently only available for 4172 individuals. These variables included lifestyle factors (smoking, alcohol use, physical activity, Body Mass Index (BMI)), self-reported health (perceived health status, long-term illness, restricted by health), psychosocial characteristics (risk of depression or anxiety disorders, loneliness, feelings of control over life) and socioeconomic characteristics (insufficient financial resources).

Outcome: multidimensional vulnerability

The outcome measure is multidimensional vulnerability, as derived from our previous study (Chapter 2) (12). Women classified into the 'multidimensional vulnerability'-class share a combination of multiple risk factors to vulnerability in several domains and lack protective factors. It is not a straightforward equation and risk factors vary across individuals. Most present risk factors include not having an income or receiving benefits, rental housing, high GP healthcare expenditures, long-term illness, negative self-perceived health, and elevated risks of feeling lonely, depressed or anxious.

We added the variable multidimensional vulnerability to the dataset of 4172 individuals. All women who were previously assigned to the multidimensional vulnerability-class were classified as 'yes' (n = 249) and women in all other classes as 'no' (n = 3923).

Statistical analyses
To assess whether it is feasible to predict multidimensional vulnerability during pregnancy using solely routinely collected data at population-level (objective 1), we employed Random Forest (RF). RF is a machine learning method for regression and classification that operates through the construction of multiple decision trees (28). The method makes no assumptions about data distribution and works well with the number of individuals in our dataset relative to the number of variables. Sensitivity analyses were conducted using XGBoost and Lasso for validation (see Appendix 1).

We sought for the optimal model using the Area Under the Curve (AUC) and F1-measure (29). The AUC, ranging between 0.5 (random) to 1.0 (perfect model), illustrates the ability of the model to distinguish between those with and without multidimensional vulnerability. Due to our imbalanced dataset with relatively few cases of multidimensional vulnerability, we calculated F1-measures to focus on correct predictions of vulnerability (29). The F1-measure balances precision, also known as positive predictive value (i.e. proportion of correct predictions out of all predicted as vulnerable) and recall/sensitivity (i.e. proportion of individuals with vulnerability correctly predicted as vulnerable by the model). We treated both elements as equally important. A perfect score means the model can identify *all* positive cases while also identifying *only* positive cases (instead of assigning those without vulnerability incorrectly to the vulnerability-class). We additionally report on specificity (i.e. proportion of correct negative predictions out of all without vulnerability) and the confusion matrices showing true/false positives and true/false negatives. In model development, we used default hyperparameters settings in the R-packing 'ranger' (30), as these typically perform well. We used nested cross-validation to choose the threshold probability for classifying multidimensional vulnerability into 'yes' and 'no' and to assess model performance (31). This involved splitting the dataset in an outer loop (six folds of train-test combinations) and inner loop (five train-validate combinations), detailed in Appendix 1. The final RF-model can be utilized for predicting outcomes on new datasets. Being the best performing model, it was also used to report on the prevalence and spatial variation of multidimensional vulnerability during pregnancy from 2017 to 2021. We computed percentages for both national and municipality levels in the five imputed datasets and we conducted an additional complete cases analysis at national level for comparison. Municipality level results were visualized on a map of the Netherlands.

Next, to identify if self-reported data on health, wellbeing and lifestyle could improve predictions with solely routinely collected data (objective 2), we gradually added self-reported data from the PHM-2016 to the RF-model. Using the previous six train-test combinations, we calculated average F1-measures for different variable sets; 1) solely routinely collected data (baseline, 31 variables); 2) baseline combined with one varying PHM-2016 variable (comprising 32 variables); 3) baseline combined with two varying

PHM-2016 variables (comprising 33 variables); and 4) baseline combined with all PHM-2016 variables, representing a potential optimum (42 variables). Comparing average F1-measures for each combination helped identify which PHM-2016 variables enhanced model performance.

To identify which variables were most important in model predictions (objective 3), we assessed variable importance in the final RF-model with and without PHM-2016 data. Variable importance was measured using out-of-bag (OOB) observations, explained in Appendix 1. This process yields a ranking of variable importance (32). As sensitivity analyses, we checked the permutation importance and Partial Dependence Plots (PDPs), explained in Appendix 1.

Ethics approval

The Clinical Expertise Centre of the National Institute for Public Health and the Environment confirmed that our study was not subject to the Dutch Medical Research involving Human Subjects Act (WMO) (reference number: VPZ-574).

RESULTS

Study population

The study population comprised 4172 women (Appendix 2). Approximately 42.1% of these women were nullipara, 4.6% had a low income and 6.0% a low educational level. In comparison to all women with unique pregnancies between 2017 and 2021 (n = 807.904), the distribution regarding most variables was comparable, but differences were found for variables such as income, educational level and ethnicity. Among the 4172 women, there was generally a lower incidence of the risk factors.

Predictions with routinely collected data

The RF-model which included the routinely collected data obtained an average AUC of 0.98 (see Table 1). Such a high AUC implies that the model sufficiently distinguishes between those with and without multidimensional vulnerability. The F1-measure had an average of 0.70, indicating that the model is able to correctly predict cases of multidimensional vulnerability, but that there are also cases missed as well as women incorrectly assigned to the vulnerability-class. Appendix 2 presents the selected hyperparameters and thresholds and the results of the separate folds. Results were consistent with those of XGBoost and Lasso (Appendix 2).

The percentage of individuals with multidimensional vulnerability during pregnancy in the Netherlands was 8.1 in 2017 and decreased to 7.2 in 2021, as derived from the RF-model (Figure 1). The percentages were slightly higher for XGBoost and lasso (respectively 8.0% and 9.1% in 2021), but showed a similar decreasing trend, as printed in Appendix 2. Appendix 2 additionally shows the complete case analysis.

Figure 2 visualizes the geographical distribution of multidimensional vulnerability during pregnancy in the Netherlands over the years 2017 to 2021, based on predictions of the RF-model. There are differences between municipalities, with percentages ranging from 1.8 to 17.5%.

Table 1. Results of the RF and the sensitivity analyses

	Metrics Mean from five-fold cross validation (SD)					**Confusion matrices for best fold** Number in each category	
	AUC	F1-measure	Precision	Recall/ sensitivity	Specificity		
Random Forest	0.98 (0.00)	0.70 (0.03)	0.74 (0.06)	0.66 (0.04)	0.98 (0.00)	30 (TP) 6 (FP)	14 (FN) 645 (TN)
XGBoost	0.98 (0.00)	0.68 (0.04)	0.70 (0.02)	0.67 (0.08)	0.98 (0.00)	34 (TP) 10 (FP)	13 (FN) 638 (TN)
Lasso regression	0.98 (0.01)	0.68 (0.04)	0.67 (0.07)	0.70 (0.07)	0.98 (0.01)	32 (TP) 12 (FP)	11 (FN) 640 (TN)

AUC = Area Under the Curve, TP = True Positive, FN = False Negative, FP = False Positive, TN: True Negative
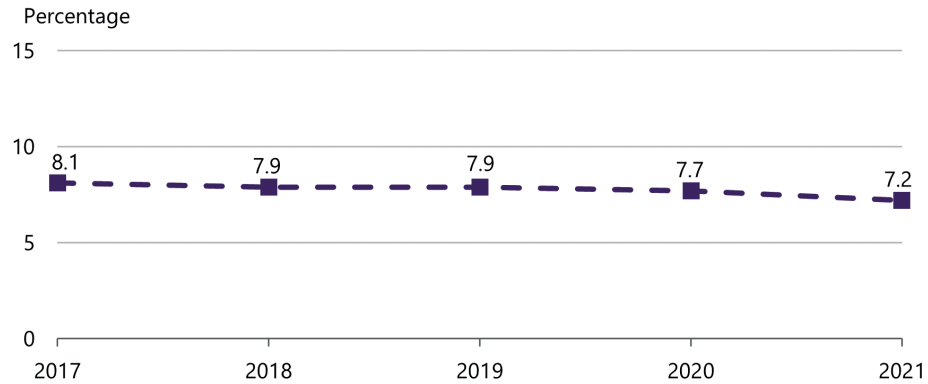Results based on analyses among study population of 4172 women

Figure 1. Percentage of multidimensional vulnerability during pregnancy in the Netherlands during the years 2017 to 2021, based on the RF-model using routinely collected data prior to pregnancy.
Results based on analyses among all unique pregnancies from 2017 – 2021 (n = 807.904)
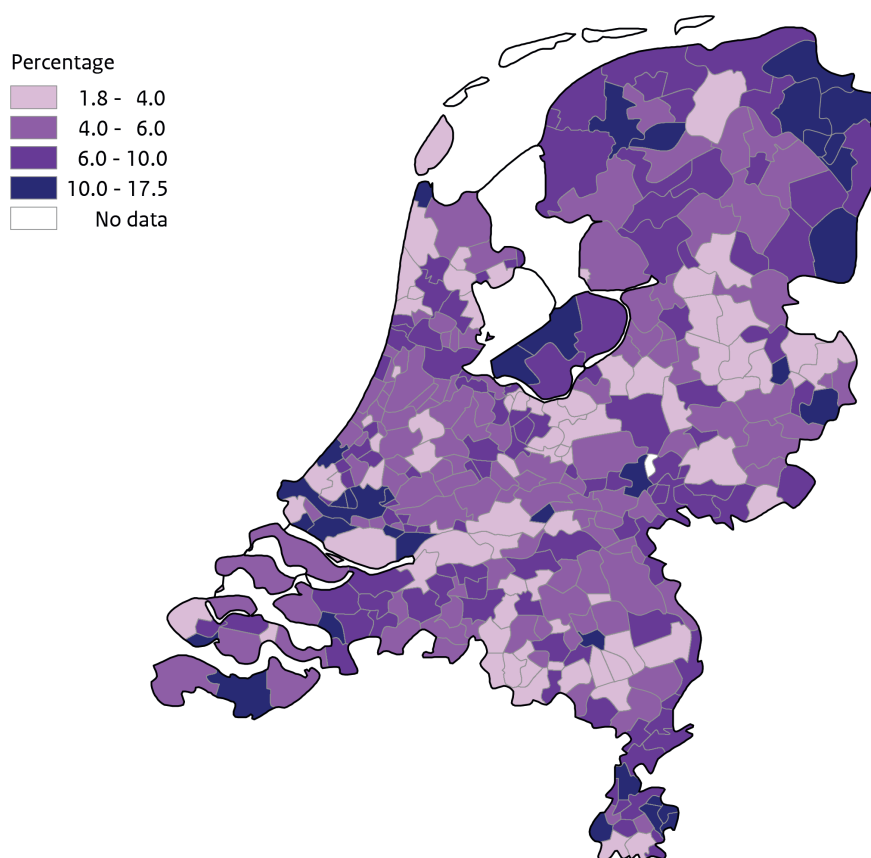
Figure 2. Heatmap visualizing the geographical distribution of multidimensional vulnerability during pregnancy in the Netherlands, at municipality level, for all pregnancies from 2017 to 2021. A darker color indicates a higher percentage of vulnerability.
Results based on analyses among all unique pregnancies from 2017 – 2021 (n = 807.904)

Adding self-reported data to predictions

The baseline F1-measure (using routinely collected data; 31 variables) was 0.70 and the potential optimum (using both routinely collected data and all self-reported data of the PHM-2016; 42 variables) was found to be 0.83, shown as vertical lines in Figure 3. Including self-reported variables improved the performance of the RF-models with solely routinely collected data. Especially self-reported data on 'perceived health status' (average 0.80) and 'restricted by health' (0.79) improved the model's performance, but also 'long-term illness' (0.77) and 'risk to depression or anxiety disorders' (0.74). Others had little impact or slightly decreased performance, such as physical activity. Appendix 2 presents the results of adding two varying self-reported variables. This further improved the performance of the model.
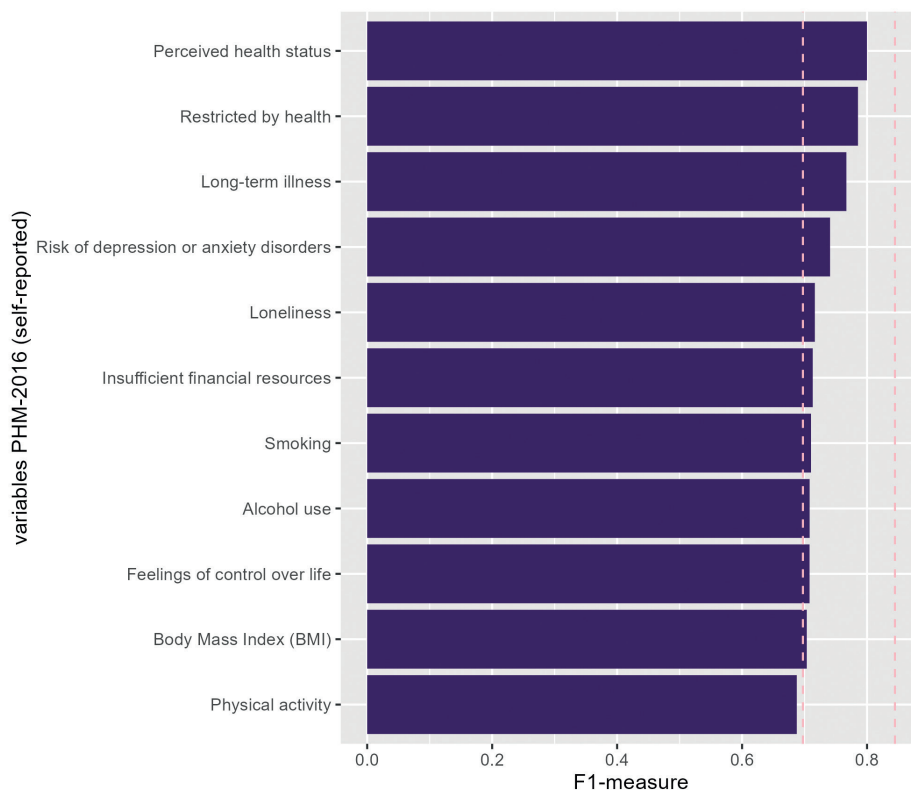
Figure 3. Variables on self-reported health, wellbeing and lifestyle added to the RF-model with solely routinely collected data. The vertical lines show the average F1-measures.
Results based on analyses among study population of 4172 women

Variable importance

Figure 4 shows the variable importance of all 42 variables. Top seven predictors for multi-dimensional vulnerability during pregnancy were: 'socioeconomic position (occupational-status)', 'perceived health status', 'restricted by health', 'permanent employment contract', 'medication use', 'long-term illness' and 'total healthcare expenditures'. Out of these seven variables, which represent both risk and protective factors, three concern self-reported health, two concern socioeconomic characteristics and two relate to healthcare expenditures and utilization. Related to financial status, self-reported 'insufficient financial resources' was ranked higher compared to the routinely collected 'household income' and 'depts and payment arrears'. Likewise, self-reported 'perceived health status' and 'feeling restricted by health' was ranked higher than 'medication use' and 'total healthcare expenditures'. We found the opposite for psychological characteristics: routinely collected 'mental healthcare utilization' was ranked higher than self-reported 'risk of depression or anxiety disorders' or 'loneliness'. However, differences were small.

The permutation importance ranking (Appendix 2) yielded comparable results, although 'mental healthcare utilization' and 'GP expenditures' were ranked slightly higher. Appendix 2 additionally shows the rankings without self-reported data, using solely routinely collected data (31 variables).
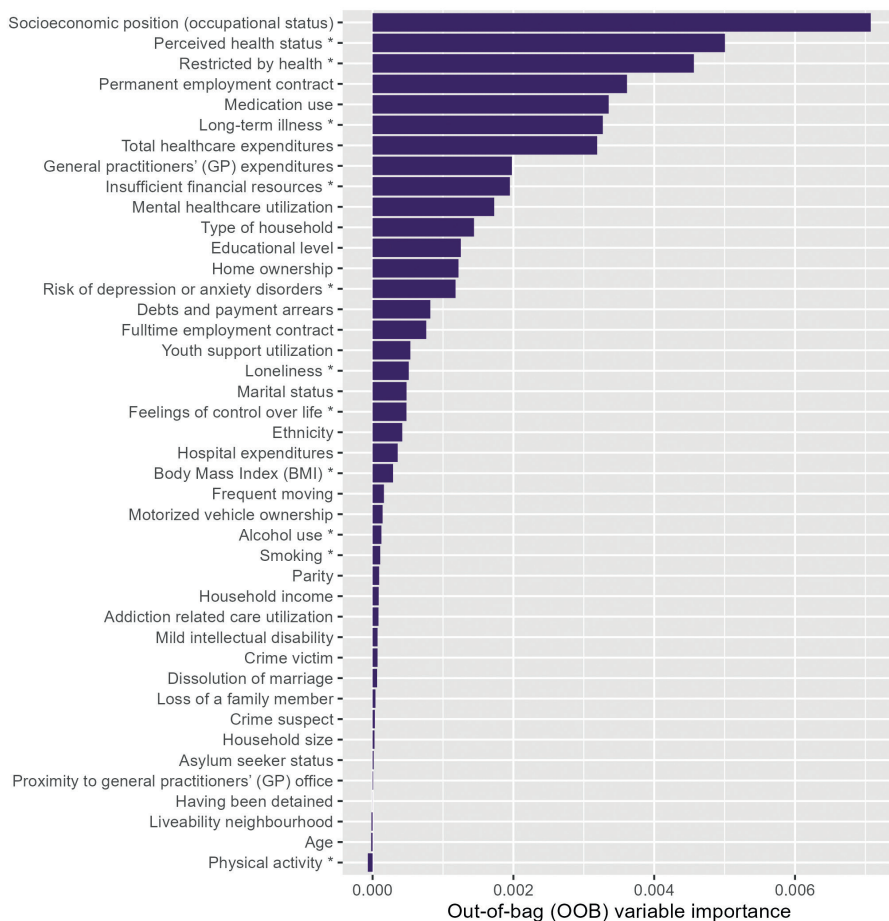

Figure 4. Variable importance ranking of the RF-model for 'multidimensional vulnerability'. The set of 42 variables used for classification are ordered from high to low importance. The length of a line represents the importance of a particular variable on the model's predictions. Self-reported data of the PHM-2016 is indicated with an asterisk (*).
Results based on analyses among study population of 4172 women

DISCUSSION

This study provides insight into predicting multidimensional vulnerability during pregnancy at population-level in the Netherlands using pre-pregnancy routinely collected data and the relevance of additional self-reported data on health, wellbeing, and lifestyle. Based on our results, it seems reasonably feasible to predict multidimensional vulnerability using solely routinely collected data, since the RF-model could distinguish between those with and without multidimensional vulnerability and was able to correctly predict multidimensional vulnerability in many cases. However, we found that adding self-reported data improved model performance. Out of the seven strongest predictors to multidimensional vulnerability in our dataset, three concerned self-reported health, two concerned socioeconomic characteristics, and two related to healthcare expenditures and utilization.

Using solely routinely collected data to predict multidimensional vulnerability appears feasible, but several women were wrongly assigned to the vulnerability class, and other cases were missed. The crucial concern is whether the model achieved adequate performance, prompting consideration of using this readily available routinely collected data versus acquiring self-reported data on experienced health. Both data sources have advantages and disadvantages, and may be used for different purposes. Using routinely collected data is relatively easy, accessible and time efficient. This pragmatic approach recognizes that not all data are available and can be utilized, analysed and interpreted. However, it is less accurate which might mainly affect those missed by the model. Considering all relevant factors by using additional self-reported data leads to better predictions. However, this has numerous implications and inherent challenges, including increased burden to practitioners, time and costs. Based on our study, we consider routinely collected data sufficient for policy monitoring of multidimensional vulnerability at population-level. It can offer insight into its scope and development over the years and help identify municipalities and neighbourhoods characterized by increased vulnerability, enabling tailored (preventive) measures for efficient budget allocation. Simultaneously, we agree with previous scholars that applying vulnerability in a dichotomous way is challenging as the concept is multi-layered, contextualized and dynamic, requiring caution to avoid over-inclusion or exclusion of individuals (33, 34). Our previous study (12) revealed a greater array of vulnerability groups, with women having risk factors within one specific domain and protective factors in others. We must not overlook these and other intermediary and personal, contextual forms of vulnerability. Our predictive RF-model was not intended for application in individual predictions and individual decision-making but meant for risk-stratification on a population-level. Because risk assessment is not straightforward, we consider routinely collected data by itself unsuitable for individual predictions, given that it insufficiently accounts for protective factors and coping strategies at an individual level, among others. We believe that an open conversation with (future) parents about their experienced health and well-being is indispensable to better understand their context and needs. It is essential that this is accompanied by a trusting relationship, and appropriate follow-up steps, preventing stigmatization, simplification

and harm (22, 33, 35). Given the added value of self-reported data however, we suggest to explore how perceived health can be systematically included into screening guidelines and care registries for professionals, to enhance the provision of personalized care and support while further improving population-level predictions in the future.

In our study, adding self-reported data led to better model performance and self-reported health indicators were found as important predictors to multidimensional vulnerability. Consistent with the psychosocial literature, several subjective measures (e.g. self-reported 'insufficient financial resources') outweighed objective measures (e.g. registered 'income' and 'debts and payment arrears') as predictors in our study. For example, multiple studies reported a stronger link between people's subjective SES and wellbeing and physical health compared to objective SES based on income or education (36-39). Arguably, perceiving your circumstances through the lens of limited resources impacts decision-making and behaviour (e.g. favouring short-term over long-term considerations), increases uncertainties and stress, and thus exacerbates pre-existing vulnerabilities (40-43). Other studies reported how self-reported health or vulnerability correspond to, outperform or complement clinical measures in predicting physical health and mortality (18-20). However, using self-reported health also has its challenges. For instance, it provides little guidance regarding what respondents consider when reporting 'poor health' and whether they refer to physical pain, mental wellbeing, less vitality or other factors (21). Additionally, people can have diverse perceptions of their health influenced by cultural contexts, social positions, and personal health experiences (e.g. people suffering from the same illness for a longer time may report better levels of health due to various coping and self-management strategies) (22, 44). Nevertheless, self-reported health seems to be an important measure which can capture components of health or vulnerability that other measures alone cannot.

Strengths, limitations and future research
The availability of nationwide data on a wide range of risk and protective factors to vulnerability in many different domains was an important strength of this study. The outcome 'multidimensional vulnerability' was also based on 42 variables (12). Additionally, we conducted several sensitivity analyses, all of which yielded similar results, underscoring the robustness of our model. However, this study also had several limitations, mostly related to the data. One limitation concerns the representativeness of the dataset used to construct and evaluate the predictive models. It is possible that some factors (e.g. asylum seeker status) did not emerge as primary predictors because they were less present among the 4172 women, despite their association with vulnerability and adverse outcomes in the literature (45, 46). This may have also led to a slight underestimation of the actual percentage of multidimensional vulnerability. Additionally, we missed data on important topics that can contribute to vulnerability such as stress, health literacy, coping skills, and adverse (childhood) experiences including violence. Another limitation is that we insufficiently considered the dynamics around pregnancy in relation to vulnerability, since we merely incorporated data prior to pregnancy that can be subject to change. Future research should take into account that vulnerability can exist prior to pregnancy, but also

arise or change during pregnancy, childbirth or after birth (9). Also a consideration of the role of the father or woman's partner and wider social network could contribute to more insights into vulnerability and better predictions.

CONCLUSIONS

This study shows that it is feasible to predict multidimensional vulnerability at population-level using solely routinely collected data. Routinely collected data is readily available for the entire population, thereby providing a robust foundation for longitudinal monitoring and policy formulation at population-level. Nevertheless, while predictions are fairly accurate, adding self-reported data is of added value.

Data availability
We are unable to share the individual data used for this study as data linkage and analysis was conducted within the highly safeguarded Remote Access (RA) platform of Statistics Netherlands (25). All data within this platform are pseudonymised to ensure data safety and confidentiality. Access to the data from Perined, Statistics Netherlands, and the Public Health Monitor 2016 can be requested from the relevant parties.

Key points

- Considering the combination of both social risk and protective factors related to vulnerability is needed to identify pregnant women at risk of adverse outcomes
- Multidimensional vulnerability at population-level can be predicted using solely routinely collected data
- Using self-reported data in addition to routinely collected data can be relevant to further improve the prediction of multidimensional vulnerability
- The strongest predictors to multidimensional vulnerability are related to self-reported health, socioeconomic characteristics and healthcare expenditures and utilization
- Without additional data collection, routinely collected data could provide insight in the prevalence, geographical distribution and trends in multidimensional vulnerability at population-level, which can be used for longitudinal monitoring and the formulation of policies.

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APPENDIX 1.
Methodology

Sensitivity analyses: XGBoost and Lasso regression

XGBoost (extreme gradient boosting) is a machine learning technique that iteratively builds multiple shallow decision trees (1). Similar to RF, it is a flexible algorithm without assuming a functional form. Logistic regression, on the other hand, does assume a strong functional form, i.e. a linear relation between the independent variables and log odds. Logistic regression is a standard approach for binary classification with a long history in literature. The logistic regression analysis was conducted with lasso penalty to shrink coefficients towards zero such that less important variables are left out the model (2).

Nested cross-validation

The three techniques RF, Lasso and XGBoost each have their own set of hyperparameters that need to be chosen for the models. For RF, the default hyperparameter settings in the R-package 'ranger' (3) were used, as these default settings generally yield good performance. The parameter to choose for Lasso (R-package 'glmnet' (4)) was the lambda, which defines the penalty, and for XGBoost (R-package 'xgboost' (5)) the number of trees and tree-depth. For Lasso and XGBoost we used cross-validation to choose the hyperparameters. In addition, as the models predict the probability of multidimensional vulnerability, we need to choose the threshold at which all predicted probabilities above that threshold are classified as multidimensional vulnerable 'yes' (and as 'no' below that threshold). To choose the hyperparameters and threshold probability, and finally to assess the performance of the models, we used nested-cross validation. Firstly, the dataset of 4172 women was split into six folds: 5 parts train-set, 1 part test-set (outer loop). Secondly, in the nesting step (inner loop), each train set from the outer loop was again split into five folds: 4 parts train-set, 1 part validation-set. During each split, we made sure that the percentage of multidimensional vulnerability was approximately equal in each part.

Firstly, using the cross-validation of the inner-loop, we chose the hyperparameters: for Lasso, we chose the average lambda across the five validation folds and for XGBoost we selected the hyperparameters for which the average AUC over the five folds was highest. Secondly, using the defined hyperparameters, and the same inner loop, we selected the threshold probability that yielded the highest F1-value on the validation set (averaged over five validation folds). Thirdly, by utilizing both the selected hyperparameters and average optimal thresholds, we calculated the F1-value on the test set of the outer loop that has not been used in selecting hyperparameters and threshold. This three-step process was repeated for the 6 folds of the outer loop, yielding the F1-measure of the model averaged over the 6 test-sets. Using the average optimal thresholds (as well as the defined hyperparameters), we fitted the model one last time on all data. The final model can be utilized for predicting outcomes on new datasets. We used the final RF-model in our next analyses.

Variable importance with OOB-observations, including sensitivity analyses

Variable importance was measured with RF in the following way. RF takes a bootstrap sample for every tree that it constructs. The data that are not used in the bootstrap sample are called the out-of-bag (OOB) observations. RF makes a prediction for these OOB-observation based on the tree that is constructed on the bootstrap-sample, leading to an OOB-error. Next, to determine the tree-specific importance of a variable, a variable is randomly shuffled (permuted) in the bootstrap sample. In this new variant of the bootstrap sample, a new tree is grown which gives a new OOB-error. This OOB-error is then compared to the original OOB-error. If permuting a variable increases the error, it is considered important as the model relied on it for prediction. Consequently, by permuting a variable and comparing the OOB-error rates of the predictions before and after permutation (6), we obtain a measure of variable importance for each variable for a single tree. The OOB-errors increase for each variable are averaged over all trees and compared. The average of all these tree importance values yields the ranking of variables for the model (7).

As sensitivity analyses, we also checked the permutation importance and Partial Dependence Plots (PDP) (8-10). Permutation importance permutes the values of a specific variable in the full dataset (rather than individual trees) to measure the impact on the model's performance. The PDP of each variable provides insight into the direction and strength of the relationship with the dependent variable while holding all other predictors constant. We checked whether the direction of the important variables aligned with their categorization as risk or protective factors.

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APPENDIX 2.
Results

Table 1. Characteristics of the study populations (including missing data)

		Original study population *(objective 1, 2 and 3)*	All unique pregnancies **2017 – 2021** *(objective 1)*
	Total n =	4172	807.904
		n (%)	n (%)
Individual characteristics			
Age	19-23	306 (7.3)	43797 (5.4)
	24-35	3528 (84.6)	624292 (77.3)
	>35	338 (8.1)	135882 (16.8)
	Missing	-	*3933 (0.5)*
Ethnicity	Non-Western	420 (10.1)	170968 (21.2)
	Western	343 (8.2)	89970 (11.1)
	Native Dutch	3409 (81.7)	546624 (67.7)
	Missing	-	*342 (<0.1)*
Parity[a]	Nullipara	1755 (42.1)	355979 (44.1)
	Primipara, multipara	2410 (57.8)	449416 (55.6)
	Missing	*<10 (<0.2)*	*2509 (0.3)*
Asylum seeker status	Yes	39 (0.9)	19582 (2.4)
	No	4133 (99.1)	788322 (97.6)
Socioeconomic characteristics			
Educational level	Low	328 (7.9)	80849 (10.0)
	Moderate	1513 (36.3)	289681 (35.9)
	High	2303 (55.2)	340669 (42.2)
	Missing	*28 (0.7)*	*96705 (12.0)*
Household income	Low	202 (4.8)	60682 (7.5)
	Moderate	3348 (80.2)	613979 (76.0)
	High	591 (14.2)	103122 (12.8)
	Missing	*31 (0.7)*	*30121 (3.7)*
Socioeconomic position (occupational status)	No income/receiving benefits	532 (12.8)	132780 (16.4)
	Student	82 (2.0)	24988 (3.1)
	Paid work	3502 (83.9)	632685 (78.3)
	Missing	*56 (1.3)*	*17451 (2.2)*
Debts and payment arrears	Yes	45 (1.1)	19357 (2.4)
	No	4127 (98.9)	788547 (97.6)
Insufficient financial resources	Yes	524 (12.6)	
	No	3267 (78.3)	
	Missing	*381 (9.1)*	
Permanent contract	No	1929 (46.2)	433393 (53.6)
	Yes	2243 (53.8)	374511 (46.4)
Full-time contract	No	1925 (46.1)	653069 (80.8)
	Yes	2247 (53.9)	154835 (19.2)
Lifestyle factors			
Smoking	Yes	661 (15.8)	
	No	3315 (79.5)	
	Missing	*196 (4.7)*	
Alcohol use	Yes (excessive)	418 (10.0)	
	No	3503 (84.0)	
	Missing	*251 (6.0)*	
Physical activity	Less than recommended	1696 (40.7)	
	As recommended or more	2158 (51.7)	
	Missing	*318 (7.6)*	

Table 1. Continued.

		Original study population (objective 1, 2 and 3)	All unique pregnancies 2017 – 2021 (objective 1)
	Total n =	4172	807.904
		n (%)	n (%)
BMI	Unhealthy BMI	1386 (33.2)	
	Healthy BMI	2641 (63.3)	
	Missing	*145 (3.5)*	
Household characteristics			
Type of household	One-person/parent household	353 (8.5)	123697 (15.3)
	Other	3819 (91.5)	684207 (84.7)
Marital status	Unmarried	2147 (51.5)	433383 (53.6)
	Married	2025 (48.5)	374521 (46.4)
Dissolution of marriage	Yes	58 (1.4)	14928 (1.8)
	No	4114 (98.6)	792976 (98.2)
Household size	≥6 persons	93 (2.2)	20412 (2.5)
	<6 persons	4079 (97.8)	757300 (93.7)
	Missing	-	*30192 (3.7)*
Youth support utilization	Yes	102 (2.4)	79473 (9.8)
	No	4070 (97.6)	728431 (90.2)
Self-reported health			
Perceived health status	Negative	465 (11.1)	
	Positive	3653 (87.6)	
	Missing	*54 (1.3)*	
Long-term illness	Yes	747 (17.9)	
	No	3362 (80.6)	
	Missing	*63 (1.5)*	
Restricted by health	Yes	724 (17.4)	
	No	3330 (79.8)	
	Missing	*118 (2.8)*	
Healthcare expenditures and utilization			
Overall healthcare expenditures	High	824 (19.8)	151443 (18.7)
	Low-average	3297 (79.0)	627116 (77.6)
	Missing	*51 (1.2)*	*29345 (3.6)*
GP expenditures	High	827 (19.8)	145166 (18.0)
	Low-average	3308 (79.3)	633247 (78.4)
	Missing	*37 (0.9)*	*29491 (3.7)*
Hospital expenditures	High	413 (9.9)	76523 (9.5)
	Low or none	3708 (88.9)	731381 (90.5)
	Missing	*51 (1.2)*	*0 (0.0)*
Medication use	High	428 (10.3)	92295 (11.4)
	Low or none	3744 (89.7)	715609 (88.6)
Addiction related care utilization	Yes	23 (0.6)	4466 (0.6)
	No	4149 (99.4)	803438 (99.4)
Psychosocial characteristics			
Mental healthcare utilization	Yes	228 (5.5)	50630 (6.3)
	No	3907 (93.6)	739093 (91.5)
	Missing	*37 (0.9)*	*18181 (2.3)*
Risk of depression or anxiety disorders	Moderate – high risk	1716 (41.1)	
	No or low risk	2256 (54.1)	
	Missing	*200 (4.8)*	
Loneliness	Feeling lonely	1100 (26.4)	
	Not feeling lonely	2719 (65.2)	
	Missing	*353 (8.5)*	

Table 1. Continued.

		Original study population (objective 1, 2 and 3)	All unique pregnancies 2017 – 2021 (objective 1)
	Total n =	4172	807.904
		n (%)	n (%)
Feelings of control over life	Low	144 (3.5)	
	Moderate	2741 (65.7)	
	High	1006 (24.1)	
	Missing	*281 (6.7)*	
Mild intellectual disability	Yes	13 (0.3)	7187 (0.9)
	No	4159 (99.7)	800717 (99.1)
Life-events			
Crime suspect	Yes	95 (2.3)	35393 (4.4)
	No	4077 (97.7)	772511 (95.6)
Crime victim	Yes	874 (20.9)	171564 (21.2)
	No	3298 (79.1)	636340 (78.8)
Having been detained[a]	Yes	not shown	2592 (0.3)
	No	not shown	805312 (99.7)
History of frequent moving[b]	Yes	53 (1.3)	14081 (1.7)
	No	4119 (98.7)	793823 (98.3)
Loss of a family member	Yes	147 (3.5)	32839 (4.1)
	No	4025 (96.5)	775065 (95.9)
Living conditions			
Home ownership	Rented	990 (23.7)	264519 (32.7)
	Owner occupied	3099 (74.3)	531526 (65.8)
	Missing	*83 (2.0)*	*11859 (1.5)*
Motorized vehicle ownership	No	494 (11.8)	162579 (20.1)
	Yes	3678 (88.2)	645325 (79.9)
Proximity to General Practitioners' (GP) office	>3 km	265 (6.4)	62040 (7.7)
	<3 km	3847 (92.2)	740876 (91.7)
	Missing	*60 (1.4)*	*4988 (0.6)*
Liveability neighbourhood	Low-mediocre	273 (6.5)	87013 (10.8)
	High	3695 (88.6)	714817 (88.5)
	Missing	*204 (4.9)*	*6074 (0.8)*

a: Following guidelines of Statistics Netherlands, the data of some variables were rounded (parity) or not shown (having been detained) to prevent the disclosure of information about individuals.
b: Erratum: in the original paper of Molenaar et al. (12), there were inaccuracies in the reported percentages of the variable 'history of frequent moving', which consequently deviate from the values presented here. Missing data are shown in italic.

Objective 1

Table 2. Hyperparameters and thresholds used for RF, XGBoost and Lasso regression

	Hyperparameters and thresholds			
	Threshold (average optimal)	Lambda	Tree depth	Number of trees
Random Forest	0.37 (0.01)	NA	default	default
XGBoost	0.36 (0.04)	NA	2	51
Lasso regression	0.26 (0.04)	0.01 (0.003)	NA	NA

Results based on analyses among study population of 4172 women

Table 3. Results of the RF and sensitivity analyses for the six separate folds

		Metrics				
		AUC	F1-measure	Precision	Recall/ sensitivity	Specificity
Random Forest	**Average**	**0.98 (0.00)**	**0.70 (0.03)**	**0.74 (0.06)**	**0.66 (0.04)**	**0.98 (0.00)**
	Fold 1	0.98	0.68	0.69	0.67	0.98
	Fold 2	0.98	0.68	0.68	0.69	0.98
	Fold 3	0.98	0.70	0.74	0.67	0.98
	Fold 4	0.98	0.75	0.83	0.68	0.99
	Fold 5	0.98	0.72	0.74	0.69	0.98
	Fold 6	0.97	0.66	0.75	0.59	0.99
XGBoost	**Average**	**0.98 (0.00)**	**0.68 (0.04)**	**0.70 (0.02)**	**0.67 (0.08)**	**0.98 (0.00)**
	Fold 1	0.98	0.68	0.68	0.69	0.98
	Fold 2	0.98	0.68	0.68	0.69	0.98
	Fold 3	0.98	0.61	0.70	0.55	0.98
	Fold 4	0.98	0.75	0.72	0.77	0.98
	Fold 5	0.98	0.66	0.74	0.60	0.99
	Fold 6	0.97	0.70	0.71	0.70	0.98
Lasso regression	**Average**	**0.98 (0.01)**	**0.68 (0.04)**	**0.67 (0.07)**	**0.70 (0.07)**	**0.98 (0.01)**
	Fold 1	0.97	0.64	0.54	0.78	0.96
	Fold 2	0.98	0.71	0.65	0.77	0.98
	Fold 3	0.97	0.65	0.71	0.60	0.98
	Fold 4	0.98	0.74	0.74	0.73	0.98
	Fold 5	0.98	0.66	0.68	0.64	0.98
	Fold 6	0.97	0.69	0.70	0.67	0.98

Results based on analyses among study population of 4172 women

Table 4. Average proportion of multidimensional vulnerability in the Netherlands over the years 2017 – 2021 for the three regression models

		2017	2018	2019	2020	2021
RF	Average proportion	0.081	0.079	0.079	0.077	0.072
	SD	8.21E-05	2.65E-05	6.7E-05	5.93E-05	0.000105
XGBoost	Average proportion	0.090	0.088	0.088	0.085	0.080
	SD	0.000105	0.000151	9.21E-05	7.95E-05	5.55E-05
Lasso regression	Average proportion	0.100	0.098	0.098	0.097	0.091
	SD	0.000116	8.69E-05	7.05E-05	5.66E-05	9.52E-05

Results based on analyses among all unique pregnancies from 2017 – 2021 (n = 807.904)

Table 5. Complete cases

		2017	2018	2019	2020	2021
RF	Proportion	0.086	0.084	0.083	0.080	0.075
XGBoost	Proportion	0.095	0.092	0.091	0.087	0.081
Lasso regression	Proportion	0.106	0.103	0.102	0.099	0.093

Results based on analyses among all unique pregnancies from 2017 – 2021 with complete data on all variables

3
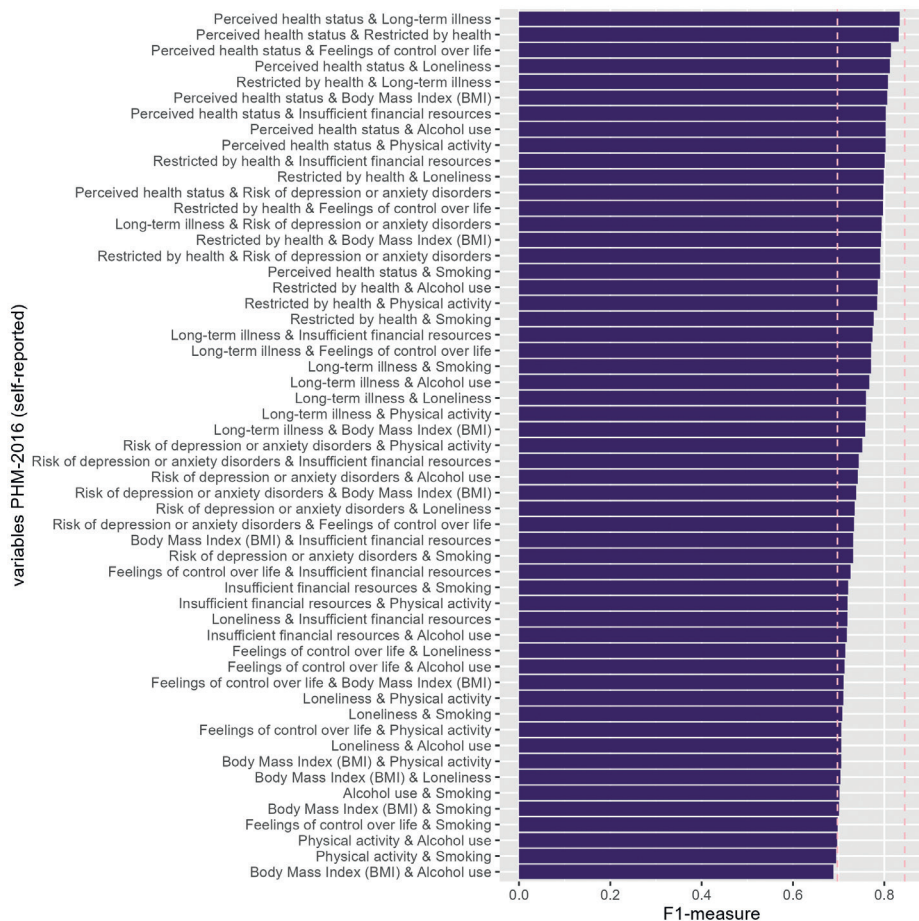
Objective 2

Figure 1. Two variables on self-reported health, wellbeing and lifestyle added to the RF-models with solely routinely collected data.
Results based on analyses among study population of 4172 women
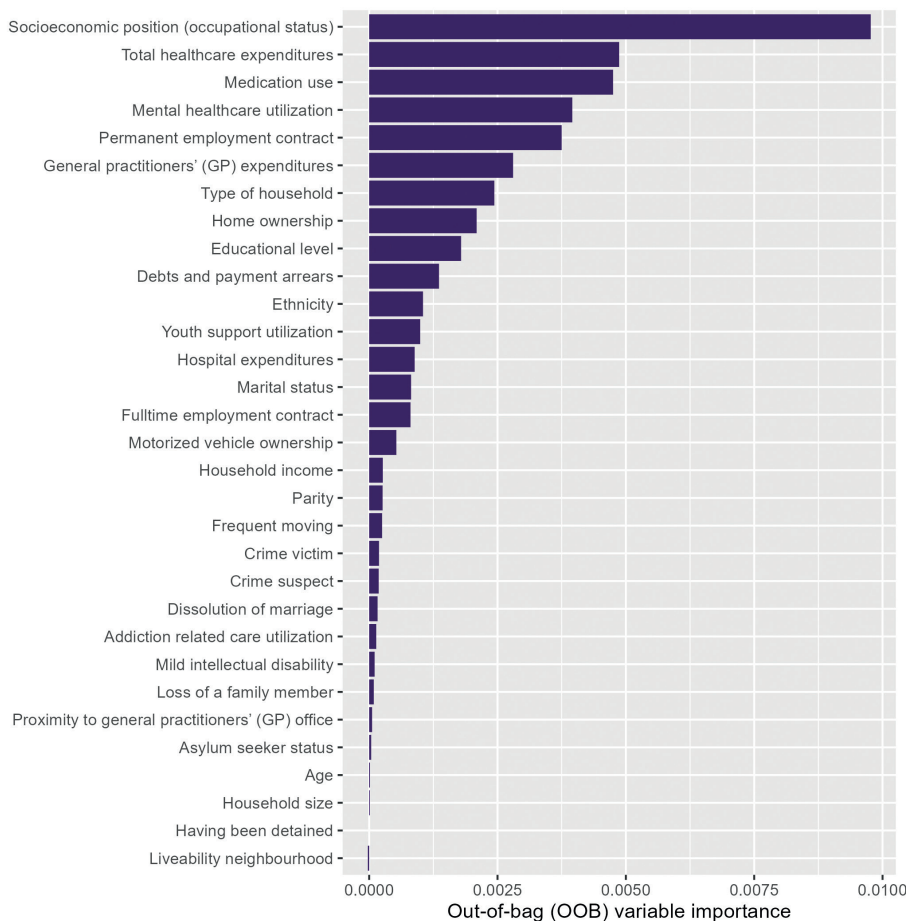
Objective 3

Figure 2. Variable importance ranking of the RF-model for 'multidimensional vulnerability', using 31 variables (solely routinely collected data).
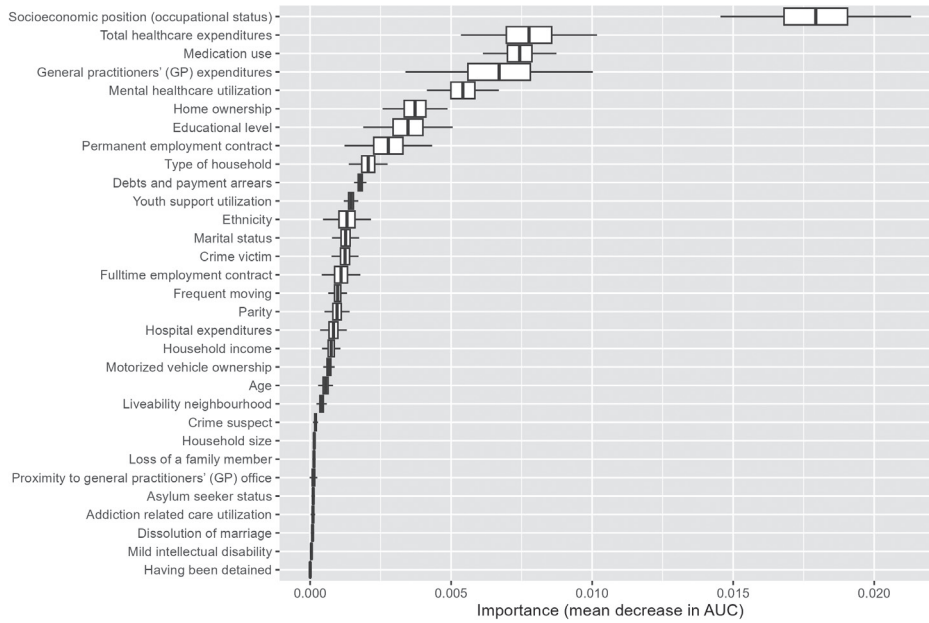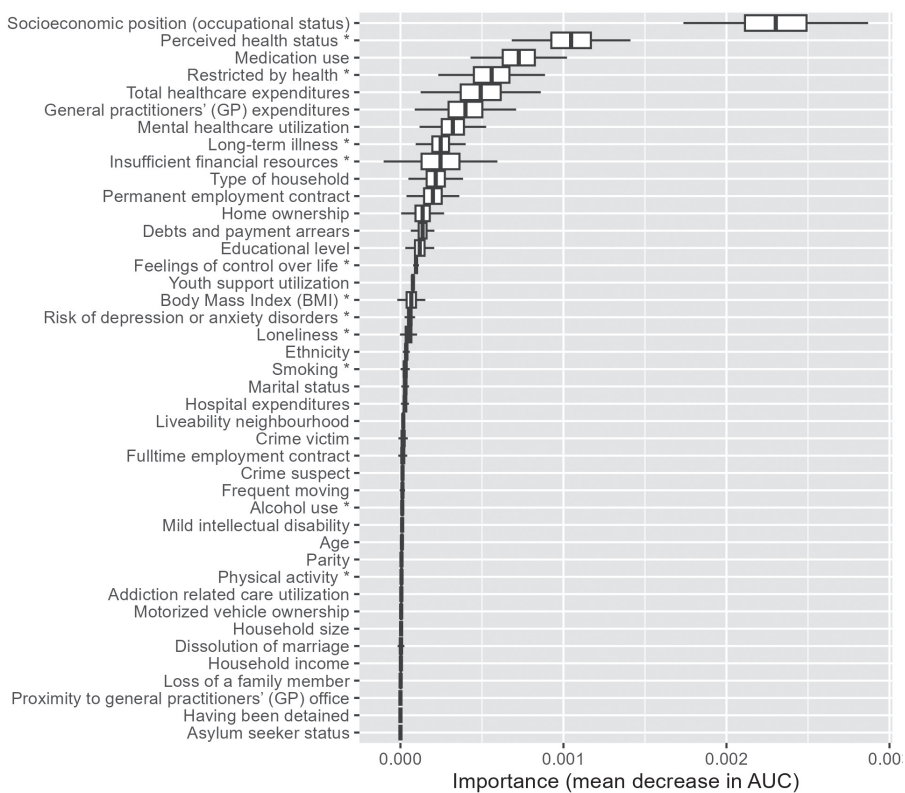Results based on analyses among study population of 4172 women

Figure 3. Permutation importance ranking of the RF-model for 'multidimensional vulnerability', using 31 variables (solely routinely collected data)
Results based on analyses among study population of 4172 women

Figure 4. Permutation importance ranking of the RF-model for 'multidimensional vulnerability', using the set of 42 variables (both routinely collected data and self-reported data)
Results based on analyses among study population of 4172 women

4

Monitoring the Dutch Solid Start program: developing an indicator set for municipalities to monitor their first thousand days-approach

Joyce M. Molenaar
Inge C. Boesveld
Jessica C. Kiefte-de Jong
Jeroen N. Struijs

ABSTRACT

Introduction

The Dutch Solid Start program aims to improve the collaboration between the medical and social sector to offer every child the best start in life. Municipalities form local coalitions of partners within the medical and social sector to support parents and children during the first thousand days. The aim of this study was to develop an indicator set for coalitions to monitor their local Solid Start program.

Methods

A modified Delphi study with three rounds was carried out among Dutch experts in Solid Start practice, policy and research (n = 39) to reach consensus.

Results

The indicator set included 19 indicators covering the three phases of the Solid Start program: preconception, pregnancy and after birth (up to two years). Prioritized indicators included both social and medical topics, among which poverty, psychological/psychiatric problems, stress, smoking, cumulation of risk factors, preconception care, low literacy, premature birth, intellectual disability. Additionally, a development agenda was established with topics and indicators that lacked data or clear operationalization (e.g. stress, unintended pregnancy, loneliness).

Discussion and conclusion

The developed indicator set enhances the conversation between policymakers, managers, professionals and other stakeholders about the local situation and developments in order to prioritize interventions and policies. Next, the indicator set needs evaluation to assess its usefulness.

INTRODUCTION

Reducing perinatal health inequities and improving health outcomes for parents and children are high on the Dutch policy agenda since the early 2000s. Following alarming perinatal mortality and morbidity figures (1, 2), several policy measures were taken to improve maternity care, including the establishment of maternity care networks (3), experiments with bundled payment for maternity care (4) and the development of the 'Standard for Integrated Birth Care' (5). Over the years, the focus of the programs shifted from the medical sector more towards the social and public health care sector, as perinatal and maternal health is strongly influenced by the wider social, economic and cultural contexts of families (6, 7). For instance, a regional cross-sectoral approach to perinatal and maternal health, integrating the medical and social sector, was taken in the local 'Ready for a baby' program (8) and subsequent 'Healthy Pregnancy 4-All' programs (9-11). These programs laid the foundation for the nationwide 'Solid Start' action program.

The Solid Start program was launched by the Dutch Ministry of Health, Welfare and Sport in September 2018 with the aim to give every child the best start in life by focusing on the first thousand days (12). This period from preconception to the child's second birthday is crucial for children's further physical, mental and social development and is therefore regarded as a window of opportunity to improve population health (6, 13, 14). The integrated approach of the Solid Start program combines medical and social services to offer better support during the first thousand days, specifically for parents in vulnerable situations. Consequently, the scope of integrated service delivery within the program is not limited to the health sector alone, but rather expanded to coordinate care and support also between the health and social sector (including public health) with its various organizations and providers (among which midwives, social workers, gynaecologists, youth healthcare providers, debt counsellors, and municipal officials). The Solid Start program is conceptualized and implemented over three phases: before pregnancy, during pregnancy and after birth (up to two years). Municipalities receive additional subsidies from the Ministry of Health to form local coalitions of partners within the medical and social sector, in order to tackle the region-specific challenges. Examples of region-specific challenges are unintended pregnancies, housing problems, domestic violence, and loneliness. This approach fits with the decentralization tendencies of social care in the Netherlands. Since 2015, the government has given municipalities new responsibilities in youth care, long-term care and income support, which cause local differences in policy implementation and outcomes (15). Next to the subsidies, supportive methods were developed and offered to local coalitions. Examples include an analysis tool to map the current and desired situation and an overview of effective interventions (e.g. prenatal home visits and 'Centering Pregnancy™': group care during pregnancy). Moreover, local coalitions receive support to develop and implement their local coalition and related programs by Pharos, which is the Dutch Centre of Expertise on Health Disparities.

The Ministry of Health commissioned the National Institute for Public Health and the Environment (Dutch abbreviation: RIVM) to monitor the implementation of the Solid

Start program. To this end, an indicator set including fifteen indicators was developed in a Delphi study with experts in 2019 (16) and reported annually in order to monitor the implementation of the nationwide program and to identify whether health outcomes improve. The indicator set reflects both processes (e.g. percentage of municipalities in which youth healthcare offers prenatal home visits) and outcomes (e.g. percentage of children born prematurely or with a low birth weight). In addition, the RIVM conducted a process evaluation to collect the experiences of those involved in the Solid Start program in order to provide further insight into factors that promote and hinder the implementation. The Ministry of Health uses the results of the monitor in combination with other data sources and expert opinions to determine whether goals are being achieved and to timely adjust policies. The results of the national Solid Start monitor showed that local coalitions evolve and formalize and that the majority of them also plan to monitor their local program, or have started to do so (17-19). However, the local coalitions generally experienced a lack of insight into which indicators to include in their local setting, where to find the data for their municipality and how to make optimal use of it. Because the national indicator set was considered less suitable for monitoring on a local level, they expressed a need for a uniform indicator set to use within their local coalition. In 2021, the RIVM started a support program that is focused on monitoring Solid Start on a local level (for additional information about the support program and its relation with the Solid Start program and national monitor see Appendix 1). Key elements of this support program include learning from and with other stakeholders (both within and between local coalitions) and sharing best practices within learning communities. The local coalitions that participated in the monitoring support program considered the development of a suitable indicator set the essential first step to stimulate monitoring on a local level.

In this paper, we describe our approach in developing an indicator set to monitor the Solid Start program in Dutch local coalitions and we present this indicator set. The indicator set can be used by local coalitions to enhance the conversation between policymakers, managers, professionals and other stakeholders about the local situation and developments in order to prioritize interventions and policies. This can help to strengthen and promote integrated service delivery.

METHODS

Design and procedure
Within this mixed-methods study, we used a modified Delphi technique as a structured method to reach consensus on an indicator set to monitor Solid Start on a local level (20). This commonly used approach in health research is suitable to synthesize knowledge from various experts with a different background or geographical location (21). Our study had several iterative rounds of self-administered questionnaires and expert meetings (Figure 1). The study was conducted between March and June 2021.
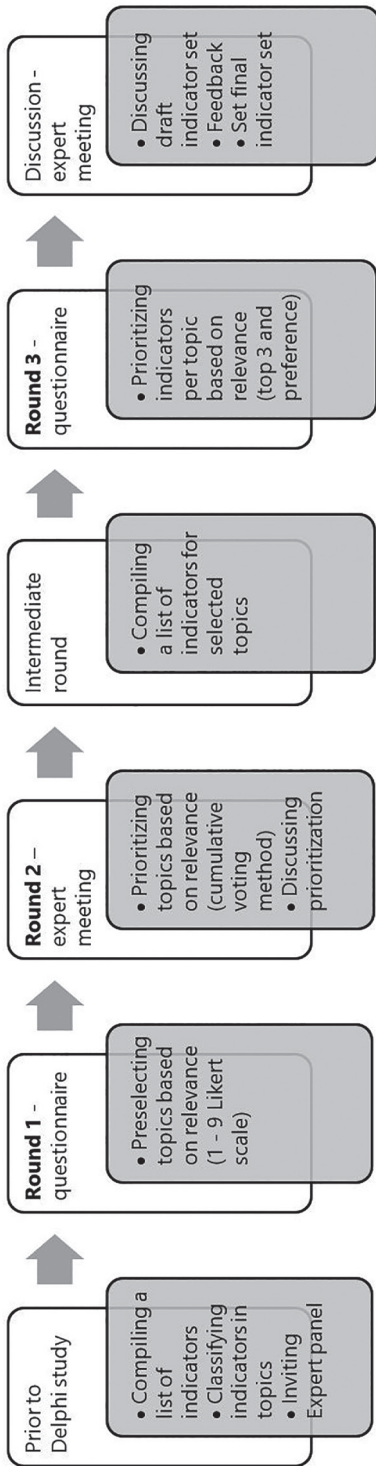
Figure 1. Schematic representation of the development and establishment of the indicator set

4

Prior to Delphi study

The study started with compiling a list of indicators originating from existing monitoring tools or documents from local coalitions, scientific and grey literature, and the indicator set used in the national Solid Start monitor (16, 22-26). The list of possible indicators was long (in a first endeavour >350) because the scope of the first thousand days is comprehensive. As this was expected to be a burden to the participants, we decided to first select topics instead of indicators directly. One researcher (JM, health scientist) categorized and named the topics in line with existing monitoring tools and documents, and another researcher (IB, former midwife and advisor integrated maternity care organizations) cross-checked this. We categorized and named the topics based on the shared characteristics and common themes in indicators (e.g. indicators relating to a low household income, debts, receiving social benefits and stress due to finances were categorized into the topic 'poverty'). Differences were discussed by three researchers (JM, IB and JS (expertise health economy)) until consensus was reached. We excluded topics that 1) did not have at least one operationalized indicator, or 2) exceeded the time period of the Solid Start program (i.e. beyond the first thousand days of life). Topics were classified in the three phases of Solid Start (preconception, pregnancy and after birth) with the reason to eventually get a sufficient number of indicators per phase. Some topics were relevant in more than one phase.

Expert panel

The expert panel consisted of a heterogeneous group of experts involved in Solid Start activities and experienced with monitoring, geographically distributed over the Netherlands (i.e. both rural and urban areas in the northern, eastern, western and southern parts of the country). We aimed for a balanced representation of experts in practice, policy and research (purposive sampling), including managers of local coalitions, policy makers, policy advisors, epidemiologists, researchers, educators, primary and secondary healthcare providers (e.g. midwife, nurse, gynaecologist, paediatrician) and social workers. We invited members of the monitoring support program (Appendix 1) and their network ('snowballing method'), and we recruited participants through social media, Solid Start-newsletters and webpages, and personal invitation. Those interested received more information about the aim, design and voluntary nature of the study. The views of participants all received equal weight during the study.

Delphi round 1: questionnaire

In an online questionnaire, the Delphi panel was instructed to rate 121 topics based on relevance to monitor Solid Start on a local level on a nine-point Likert-scale (1 = not relevant at all, 9 = highly relevant). We gave an example of a possible indicator for each topic for comprehensibility. In addition, experts were invited to comment on the topics or to suggest additional topics for each of the three phases in the open spaces of the questionnaire.

All ratings were analysed by calculating the median score and level of agreement between experts, following the RAND/UCLA Appropriateness Method user's manual (27). Based on the median scores, topics were classified as either inappropriate (median range 1 – 3), uncertain (median range 4 – 6) or appropriate (median range 7 – 9) (Appendix 1). Level of

agreement was assessed by the IPR-score (interpercentile range, difference between 30th and 70th percentile) and the IPRAS-score (interpercentile range adjusted for symmetry). If the IPRAS is larger than the IPR, there is agreement among experts and if the IPR is larger than the IPRAS, there is disagreement.

We planned to 1) accept topics with median score ≥ 7 with agreement, 2) reject topics with median score ≤ 3 with agreement, and 3) discuss all other topics (median score 4 – 6 or without agreement) in Delphi round 2. However, round 1 resulted in a large majority 'accepted' topics and well exceeded the number of intended indicators. We therefore decided to prioritize these 'accepted' topics in the second Delphi round and rejected all other topics.

The experts' suggestions for new topics were read and discussed by the researchers (JM, IB, JS) until consensus was reached on additional topics. New topics were combined or reformulated if necessary and added to Delphi round 2.

Delphi round 2: expert meeting
The second Delphi round consisted of expert meetings to prioritize the topics using the cumulative voting method. Meetings were held online due to Dutch COVID-19 policy restrictions and we organized three separate smaller meetings to encourage active participation during the online meetings. The meetings of +- 120 minutes were recorded. Experts were first informed about the results of Delphi round 1. Next, they were encouraged to prioritize topics by dividing 100 points at their own discretion. After the individual prioritization, experts entered their scores into an interactive program to aggregate scores of all participants in the meeting. We encouraged experts to reflect on these aggregated scores. After the discussion, experts were invited to reconsider their earlier individual scores again. This sequence was repeated for the three phases (preconception, pregnancy and after birth).

Subsequently, we aggregated all final scores and classified the topics from high to low sum scores. Within every phase (preconception, pregnancy and after birth) we searched for a sudden decline in sum scores as a natural cut-off point for prioritized topics. This led to a draft list of prioritized topics.

In addition, we transcribed the expert meetings verbatim and analysed the data using MaxQDA. One of the researchers (JM) coded the data for considerations in the prioritization and requirements for the indicator set. Coding was checked by a second researcher (IB).

The researchers (JM, IB, JS) consequently checked the draft list of prioritized topics against the experts' requirements for the indicator set. We checked whether the requirements were fulfilled or whether we should add lower prioritized topics to fulfil the requirements. At the end of the second Delphi round, we had a final list of prioritized topics.

Intermediate round
Based on the final list of prioritized topics, we made a list of possible indicators for each topic. Indicators were derived from our previous list of possible indicators (prior to Delphi study) as well as suggestions made by experts during Delphi round 1 and 2. Indicators were reformulated or merged in case they were not clearly defined or overlapped, based on consensus between two researchers and in line with the other indicators (JM, IB). In the rare case that there was no indicator available in the mentioned sources for one of the topics, the researchers (JM, IB) formulated potential indicators based on comparable indicators (e.g. indicators for the same topic in other phases). For each indicator, we described its numerator, denominator, data source, and data availability.

Delphi round 3: online questionnaire
The third Delphi round consisted of an online questionnaire to select and prioritize indicators. The experts received a list of possible indicators (including numerator and denominator) for each topic and were encouraged to 1) select a maximum of three indicators they considered suitable to monitor Solid Start on a local level, and 2) indicate their number one preference. In case only one possible indicator was presented, experts were asked whether or not they considered that indicator suitable. The experts were also invited to add comments.

For each indicator, we calculated the percentage of experts that selected the indicator within their top three or as their preference. The scores and comments were discussed by the researchers (JM, IB, JS) in order to select at least one indicator per topic. In this process, the following conditions were considered: 1) Is there a clear preference towards one indicator? 2) Is data available for this indicator in nationwide data sources for every municipality? 3) Is the indicator sufficiently operationalized? If all conditions were met, the preferred indicator was added to the draft indicator set. We additionally prepared a 'development agenda' for topics and indicators that were clearly preferred, but lacked data in nationwide data sources or a clear operationalization. In this case, a lower ranked indicator for this topic with data-availability and sufficient operationalization was added to the draft indicator set.

Discussion: expert meeting
In a final two-hour online expert meeting we presented the draft indicator set (including the 'development agenda') and asked experts for feedback. Specifically, we checked whether the set covers the various elements to appropriately monitor Solid Start on a local level. Experts were encouraged to share their thoughts in the meetings' chatbox or by e-mail afterwards. Pressing issues were discussed directly. Based on the meeting minutes and written feedback, we finalized the indicator set.

Ethical considerations
Following the Dutch Medical Research Involving Human Subjects Act (WMO), ethical approval was not necessary for this study (http://www.ccmo.nl), as we did not conduct medical-scientific research and participants were not exposed to treatment or required to

follow a certain behavioural strategy. All participants gave written informed consent. In an information letter and at the start of each round or meeting, we stressed that participation was voluntary and confidential, and that data were processed anonymously.

RESULTS

Participants
The expert panel consisted of 39 experts (Table 1). The full questionnaire to select topics (round 1) was completed by 39 experts and 28 experts joined the online expert meeting to prioritize topics (round 2). A total of 28 experts participated in the questionnaire to select indicators (round 3) and 21 experts were present during the final expert meeting. 18 experts joined during the full study.

Table 1. Characteristics of participants

	Total **	Round 1 - questionnaire	Round 2 - expert meeting	Round 3 - questionnaire	Discussion - expert meeting
Total number of participants	39	39	28	28	21
Field of expertise					
Policy*	22	22	16	16	14
Practice*	12	12	7	9	7
Social sector	4	4	2	3	1
Medical sector	3	3	1	1	1
Both	5	5	4	5	5
Research*	9	9	7	6	4
Other (e.g. providing support for collaboration and the formation of Solid Start coalitions in general)	3	3	3	3	2

*More than one field of expertise is possible
**The same pool of 39 experts was approached in each round (e.g. the discussion was attended by 21 of these 39 experts).

Figure 2 shows a flowchart of the selection of topics and indicators during the study.
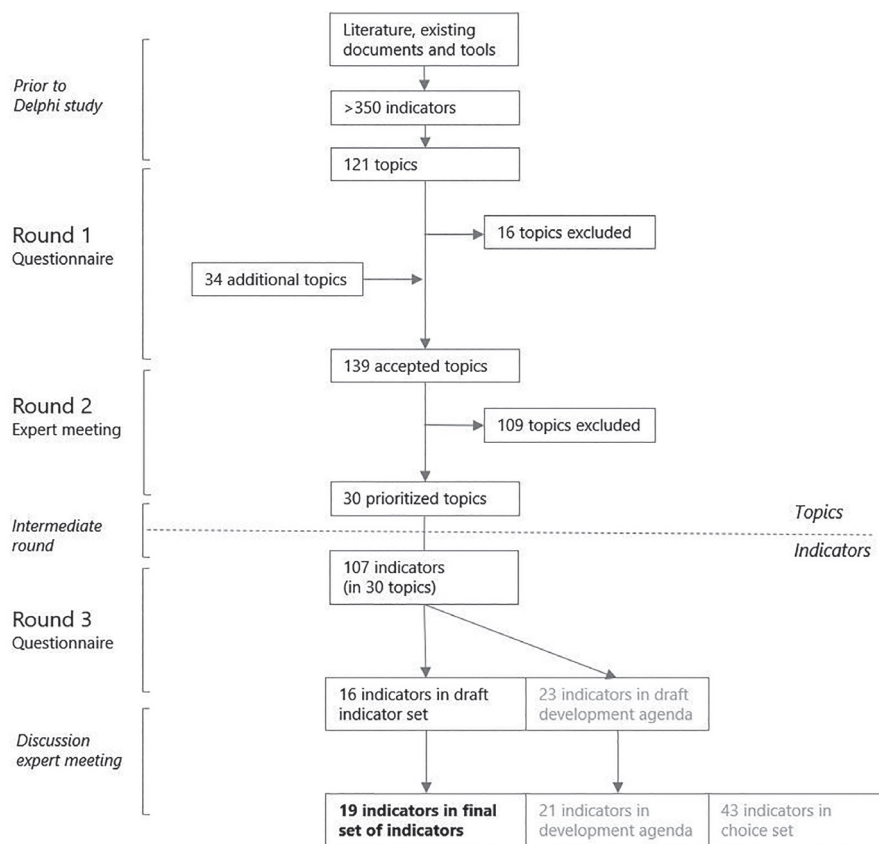
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Figure 2. Flowchart of the selection of topics and indicators to monitor Solid Start on a local level

Round 1 - questionnaire
The experts received 121 possible topics to rate. Out of these, 105 topics were selected (median score ≥7) and 16 topics were excluded (median score <7) (Appendix 2). These excluded topics mainly concerned complications or medical risks during pregnancy or after birth (e.g. gestational diabetes and caesarean-section). Based on the experts' suggestions, 34 topics were added. Some topics were completely new, but most were already mentioned in another one of the three phases (preconception, pregnancy, after birth). In total, 139 topics were selected for round 2.

Round 2 – expert meeting
Experts prioritized topics within each of the three phases (Appendix 2). For the preconception phase, the topic 'poverty' received the highest sum score. The topic 'cumulation of risk factors' received the highest sum scores for the phases of pregnancy and after birth. A decline in sum scores was clear in the pregnancy-phase after 10 topics (from 112 points to 96 points), but less clear for the other phases. We selected the prioritized 10 topics within each phase (a total of 30 topics, Table 2). Most topics belonged to two or three phases.

Table 2. Overview of the prioritized topics (n = 30)*

	Preconception	Pregnancy	After birth (up to two years)
Topics in all three phases*	Poverty	Poverty	Poverty
	Early detection by healthcare provider	Early detection by healthcare provider	Early detection by healthcare provider
	Health: psychological/ psychiatric problems	Health: psychological/ psychiatric problems	Health: psychological/psychiatric problems parents
	Health: stress	Health: stress	Health: stress
Topics in two phases*	Domestic violence (including screening)	Domestic violence (including screening)	
	Substance use: smoking	Substance use: smoking	
		Social network	Social network
		Cumulation of risk factors	Cumulation of risk factors
Topics in one phase	Preconception care	Care: multidisciplinary collaboration	Health outcomes child: premature birth
	Interventions (process indicators)	Unintended and/or unwanted pregnancy	Relation parent – child
	Low literacy		Health: intellectual disability parent
	Client characteristics: socioeconomic status		Child abuse and neglect

*The topics that occur in multiple phases are presented on the same row.

Experts mentioned multiple requirements for the final indicator set (see Appendix 3 for a description of all requirements and corresponding quotes). The indicator set should include indicators regarding both processes and outcomes, and both parents and children. Experts moreover wanted to include indicators that have the potential to be influenced (to identify early effects of policy) as well as indicators that show prevalence rates (to be used in making policy). The total indicator set should be balanced in terms of risk- and protective factors and in general it should provide a full picture of all relevant aspects. The indicator set should provide a starting point of the conversation within a cross-sector collaboration. Lastly, it was considered important that data are available for the indicators. No additional topics were added to the final indicator set based on these requirements, since the prioritized topics largely seemed to match these requirements.

Intermediate round

For the 30 prioritized topics, 107 unique indicators were found by the research team in the different sources. The number of potential indicators per topic varied from 1 to 7.

Round 3 - questionnaire

Based on the experts' selection and prioritization, the preferred indicator was clear for 20 topics (Appendix 2). 11 of these indicators lacked data and were added to the development agenda. As the 'second best' option, 5 lower prioritized indicators for the corresponding topics were added to the draft indicator set. The draft indicator set consisted of 16 indicators, the draft development agenda of 23 indicators.

Discussion – expert meeting

In general, experts appreciated the draft indicator set. They mentioned a number of extra non-prioritized indicators, which were added to an additional 'choice set' in case data

was available (Appendix 2). This set complements the basic indicator set and allows local coalitions to use additional indicators (e.g. regarding educational level, single parent family, long-term low income) if they want to.

In reflecting on the indicator set, experts mentioned some conceptual considerations (e.g. indicators are often formulated as risks, while the reverse can be a protective factor). They also mentioned methodological considerations (e.g. indicators regarding children's health at age two are currently missing and should be added when more youth healthcare data is available). Experts gave their consent to the indicator set provided that the set will be piloted in practice. Based on the experts' feedback, the indicator set and development agenda were finalized.

Final indicator set
Finally, 19 indicators could be selected to monitor Solid Start on a local level (Table 3): 7 in the preconception phase, 5 during pregnancy and 7 after birth (up to two years). Some examples are debts, psychological or psychiatric problems, late antenatal care, smoking during pregnancy, vulnerability during pregnancy and after birth, not receiving postpartum care, and preterm birth and/or low birth weight for gestational age (SGA). Appendix 2 describes the selected indicators in more detail. Data is available in nationwide data sources for all these operationalized indicators and can be presented at local (municipality) level.

The development agenda consists of 21 indicators (Appendix 2). These (preferred) indicators lacked data or a clear operationalization. Some examples are smoking before pregnancy, stress due to finances, unwanted or unplanned pregnancy, stress during pregnancy, loneliness among parents, secure bonding, abuse or neglect of children, and stress with parenting.

Table 3. Selected indicators to monitor Solid Start on a local level (n = 19)

Preconception
Percentage of women and men in the reproductive age with debts
Percentage of women and men in the reproductive age with psychological or psychiatric problems
Percentage of women and men in the reproductive age with stress
Percentage of women in the reproductive age who smoke
Percentage of families reached with a preconception consultation (preconception care)
Percentage of low literacy among young people (<30 years) without partner and children
Percentage of women and men in the reproductive age living in a neighbourhood with a low liveability score
Pregnancy
Percentage of pregnant women with debts *
Percentage of pregnant women who have their first antenatal care visit after the 10th week of pregnancy *
Percentage of pregnant women with psychological or psychiatric problems
Percentage of women who smoke at some point during pregnancy
Percentage of pregnant women in a potentially vulnerable situation (3 or more risk factors to vulnerability)
After birth (up to two years)
Percentage of children born in a family with debts
Percentage of families not receiving postpartum care (at home) after birth *
Percentage of children aged 0 to 2 years of whom one or both parents have psychological or psychiatric problems
Percentage of children born in a family in a potentially vulnerable situation (3 or more risk factors to vulnerability) *
Percentage of children with a preterm birth or with a low birth weight for gestational age (SGA) *
Percentage of children born in a family of which one or both parents have a mild intellectual disability
Number of out-of-home placements for children before the age of 2 (per 1,000) *

* These indicators are also included in the indicator set to monitor the national Solid Start program.

DISCUSSION

In this paper, we present an indicator set to monitor the Solid Start program in Dutch local coalitions, and we describe how we used a modified Delphi technique to reach consensus. The final indicator set consists of 19 indicators, covering the three phases of the Solid Start program: preconception (n = 7), pregnancy (n = 5) and after birth (up to two years) (n = 7). These indicators are available in nationwide data sources and can be presented on local (municipality) level. The indicator set meets the requirements as mentioned by the experts; it contains indicators that cover both processes and outcomes, both parents and children, and both risk- and protective factors. Additionally, the indicator set reflects both medical and social factors. A development agenda was established with topics and indicators that were prioritized, but lacked data in nationwide data sources or a clear operationalization.

The indicator set covers the following topics: poverty, psychological/psychiatric problems, stress, smoking, cumulation of risk factors, preconception care, low literacy, socioeconomic status, premature birth, intellectual disability, and child abuse and neglect. The first four topics are presented in the indicator set for all three phases (preconception, pregnancy and after birth). In general, the social determinants of health (7, 28) are represented in the indicator set (e.g. debts, low literacy and living in a neighbourhood with a low liveability score). Specific clinical aspects that belong to one group of care providers (e.g. caesarean section, a child's hearing) are less present. Nonetheless, the indicator set reflects both medical and social care, which aligns with the aims of the Solid Start program. In comparison to the indicators used in the current national Solid Start monitor (Appendix 4), there is some overlap (e.g. debts during pregnancy, preterm birth and low birth weight for gestational age) but also differences. For instance, the national monitor also includes indicators such as 'the percentage of municipalities that implemented the program 'Not Pregnant Now''. These differences are arguably caused by the different purposes of both indicator sets. The indicators in the national monitor can be used to monitor and evaluate the nationwide implementation of the program, and to monitor health outcomes of parents and children on a national level. As the implementation and health outcomes vary between municipalities, the indicator set of the local monitor aims to enhance the conversation between policymakers, managers, professionals and other stakeholders about the local situation and developments in order to prioritize interventions and policies at a local level.

A development agenda was made with indicators and topics that lacked data in nationwide data sources or a clear operationalization. Among others, the topics and indicators on the development agenda were related to stress, unwanted or unintended pregnancy, (quitting) smoking before pregnancy, loneliness, early detection, secure bonding, and child abuse or neglect. Multiple indicators related to stress were prioritized: stress due to finances, stress during pregnancy and stress with parenting. There is growing scientific evidence that stress during pregnancy or parenting has long- and short-term consequences for children's health and development (14, 29, 30). The multidimensional concept of stress (31) may require different indicators. It seems, therefore, valuable to explore which topics of the development agenda should be prioritized to be incorporated in routine registries for the purpose of local monitoring.

There are, to the best of our knowledge, no other studies that used a Delphi technique to identify indicators for local monitoring of the full first thousand days (approach). There are, however, several previous studies that sought to describe indicators for aspects of the first thousand days, including antenatal care (32), obstetrical care (33), children's health (34), birth centre care (35), and maternal and newborn health (36) or care (25) during pregnancy, childbirth and the postpartum period. Next to that, we found several programs in other countries that were focused to the first thousand days, but the aims, scope and key-design elements of the programs and their evaluation differ (37-41). These programs were often not directly comparable to the Dutch Solid Start program and not (yet) focused on supporting monitoring on a local level. Consequently, a comparison between our indicator set and indicators in the aforementioned studies is hampered, with the exception of a

study from Sweden (26). In this Swedish study, the researchers developed indicators, sub-indices and a summary index in order to support municipalities with monitoring children's health. In comparison to our study, they also mentioned both risk- and protective factors and also selected indicators related to poverty, smoking and low birth weight.

Strengths and limitations

A strength of this study is that the indicator set is developed based on the expertise of a heterogenic and balanced group of experts in policy, practice and research related to the first thousand days, who have an interest in using the set in daily practice (20). The focus of the indicator set to the first thousand days, involving both the social and medical sector, is necessary for programs aimed at reducing health inequities as health outcomes are directly and indirectly influenced by both social and medical factors (6, 9, 42). The experts exchanged information and expressed their views during two expert-meetings, as done in previous Delphi-studies (20). We organized a meeting to discuss and prioritize topics (Delphi round 2) and a final expert meeting. We considered this final moment of reflection on the (draft) indicator set very important to increase the support and future uptake of the indicator set in practice.

However, this study also has several limitations. First, we selected indicators based on consensus without considering the scientific evidence for these indicators. This does not necessarily mean that indicators that were not prioritized are not valid and vice versa. For most indicators to monitor maternal and neonatal health, their level of evidence is not well described (25). In general, the rare availability of evidence is one of the reasons to (partly) select indicators based on experts' opinions in a Delphi study (20). Another limitation was that not all indicators in the final set were the preferred option by experts as a consequence of limitations in data availability. Hence we included some 'second best' indicators and added the preferred indicators to the development agenda. Other limitations relate to the inclusion of experts. This depended on the availability and willingness of experts to participate within the study's time period, and on the decisions of the researchers in how and who to invite. Moreover, we invited experts from practice, policy and research in both the social and medical sector. Making a clear distinction between and within those categories is not always possible, as multiple experts work at the intersection of the various fields of expertise (practice, policy and research) or in multiple sectors (medical and social). For example, managers of local coalitions can be categorized as working in both practice and policy, as well as within the medical and social sector. The inability to distinguish between the field of expertise and sector is however in line with the aims of the program (i.e. integrating service delivery across the medical and social sector). Therefore, we do not expect that this may have influenced the results. This is also reflected in our results, as the experts from different fields of expertise and sectors did not prioritize different topics and indicators. Additionally, some experts dropped out during the study period, but the three groups of experts from practice, policy and research were all well represented during the various rounds. In addition, we missed the perspective of parents themselves. Finally, due to the COVID-19 pandemic, we were unable to organize physical meetings. Our decision to organize three smaller online meetings hindered the exchange of information and

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considerations between all experts. However, since the results of each of the meetings were highly comparable, we expect little influence on the results.

Future research and practice
Recently, the first indicators were quantified and presented to all municipalities in the Netherlands at www.regiobeeld.nl/kansrijkestart. In the future, we will further refine the website with additional indicators and new functionalities (among which maps with geospatial variation). In quantifying the indicators, we use nationwide observational data sources with routinely collected data, which are linked on individual level. In the last decade, the opportunities of linking observational data sources has increased at an enormous pace, which enhances the usefulness and applicability of the developed indicator set (43).

The indicator set has yet to be used and evaluated in practice, as we can only determine the feasibility through empirical testing. A previous systematic review concluded that not many published indicators for maternal and neonatal health are empirically tested for validity and feasibility (25). Starting in 2022, we will evaluate and refine the indicator set in close collaboration with the participants of the monitoring support program (Appendix 1) in order to stimulate the uptake and adoption in daily practice. During this process, we expect to also discover which indicators are most often used and how, also for indicators that are similar across two or three phases (e.g. debts before pregnancy, during pregnancy and after birth). Using the indicator set should not be a one-time action, because the strength of using indicators for monitoring in municipalities is the comparison with previous comparable figures (26). In the future, the indicator set will be refined because of new developments, changing demographics, new evidence and increased data-availability. In reflecting on the use of the indicator set, it is also important that we pay attention to questions about obtaining and presenting the data.

In the coming years, the topics on the development agenda will be prioritized and addressed in collaboration with national parties and local professionals. Central in this process is the formulation and operationalization of indicators and the expected increase of data-availability. Next to the indicator set and development agenda, the choice set with extra, non-prioritized indicators is also publicly shared (including where to find the data) for local coalitions to use.

Relevancy
We consider our study scientifically relevant as it increases our understanding of relevant indicators for Solid Start and of using a systematic approach in developing indicators for monitoring a cross-sectoral program. In addition, it is relevant for society, as we can directly benefit from the study results by using the indicator set in practice. In the Netherlands, the indicator set can be used by local coalitions in collaboration with local stakeholders to describe their population, to identify gaps in current processes, to make or adapt policies, to prioritize interventions, to monitor developments and to stress the importance of investing in the first thousand days. In this monitoring process, combining quantitative data with

qualitative data about experiences, facilitators and barriers (in a mixed-methods approach) can help to interpret the quantitative data, gain more insight into processes and explore opportunities for improvement (44). Using the indicator set in combination with qualitative data in a continuous learning cycle with local stakeholders can support an integrated approach that is adapted to the local context in Dutch municipalities. On an international level, the topics and indicators can potentially be a starting point for monitoring similar cross-sectoral programs into the first thousand days in other Western countries (37-41). Additionally, countries that aim to develop a supported and comprehensive indicator set to monitor a cross-sectoral program can learn from our systematic methodology of collaborating with experts with varying backgrounds. Using a co-creative process can increase the support, relevancy and therewith impact of the research project (45, 46).

CONCLUSION

In this study we present an indicator set for monitoring the Dutch Solid Start program on a local level, which will be used and evaluated from 2022 onwards. The indicator set consists of 19 indicators that reflect both social and medical factors. The indicator set can be used by local coalitions to enhance the conversation between stakeholders about the local situation and developments in order to prioritize interventions and policies. Using the indicator set for monitoring is a continuous process that supports the optimalisation and promotion of integrated service delivery across the medical and social sector at a local level. Ultimately, the indicator set contributes to the reduction of health inequities within the preconception period, during pregnancy and after birth in order to give each child a solid start.

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APPENDIX 1.
RIVM monitoring support program – 'Learning Local Monitor Solid Start'

In 2021, the National Institute for Public Health and the Environment (Dutch abbreviation: RIVM) started a support program focused on monitoring Solid Start on a local level. Key elements of the support program include learning from and with other stakeholders (both within and between local coalitions) and sharing best practices. The program stimulates local coalitions to use monitoring as a tool to further develop and improve their local approach.

There are eleven Solid Start coalitions that participate in regular learning sessions. These coalitions already started to monitor their local Solid Start program at an early stage; before or soon after the start of the national program. During these regular learning sessions (four in 2021), the specific needs for support are identified. These needs for support are discussed during several theme sessions (five in 2021) that are accessible to a wider audience. Everyone involved or interested in (monitoring) Solid Start can participate: professionals in the medical and social domain (e.g. midwives, social teams), researchers, managers, representatives of local organizations, etcetera.

The development of an indicator set to monitor Solid Start on a local level was considered by the eleven coalitions as the essential first step to stimulate monitoring on a local level. Other themes that were covered during the support program in 2021 were: 1) gaining insight into vulnerability, 2) monitoring the collaboration between medical and social domain, and 3) using monitoring and evaluation to learn, for example by involving experts-by-experience (parents or future parents) in local monitoring.

The relation between the national Solid Start program, national monitor and local monitor is illustrated in Figure 1.
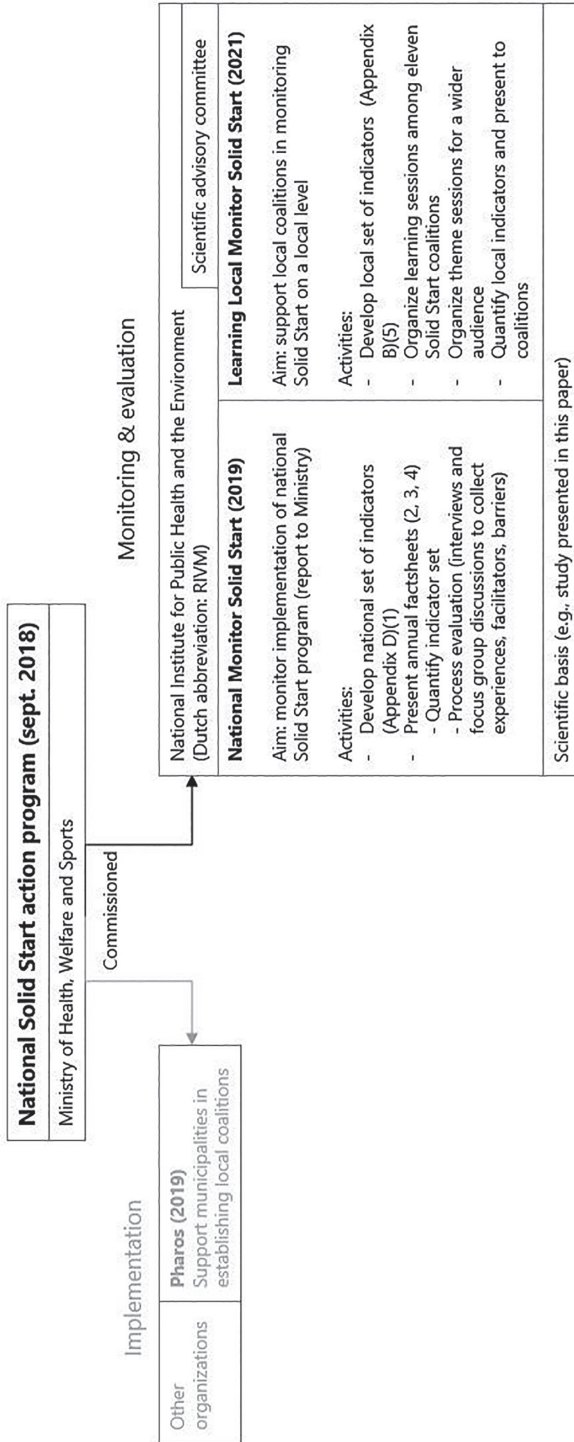
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Figure 1. Diagram illustrating the relation between the national Solid Start program, national monitor and local monitor

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APPENDIX 2.
Results of Delphi round 1, 2, and 3, final local indicator set, choice set and development agenda

Appendix 2 (Excel document) is available for download at https://doi.org/10.5334/ijic.6508.s2

Table of content

Name of worksheet	Content
Round 1	• Results of Delphi round 1: online questionnaire to select topics. • Experts rated topics based on relevance to monitor Solid Start on a local level on a nine-point Likert-scale. This worksheet contains an overview of the median scores and level of agreement between experts for each topic.
Round 2	• Results of Delphi round 2: expert-meetings to prioritize topics. • Experts individually divided 100 points over the topics during three meetings. This worksheet contains the aggregated sum scores for all topics for each seperate meeting and for all meetings together.
Round 3	• Results of Delphi round 3: online questionnaire to prioritize indicators. • Experts selected a maximum of three suitable indicators and one preference for each topic. This worksheet contains an overview of the percentage of experts that selected an indicator in their top 3 and as their preference.
Final indicator set	• An overview of the final indicator set for local monitoring, including each indicator's denumerator, data source and additional information regarding data availability.
Choice set	• An overview of the choice set: extra, non-prioritized topics and indicators.
Development agenda	• An overview of the development agenda: topics and indicators that were preferred, but lacked data or a clear operationalization.

APPENDIX 3.
Considerations in the prioritization and requirements for the final indicator set

Indicators regarding both parents and children	• Indicators concerning parents' health and well-being are important to develop policies that can improve the environment in which children grow up • Children are key within the first thousand days program and child outcomes can reveal whether policy eventually has the desired effect *"I think you need a good mix in that and not only include the characteristics of the parents and the family where the child grows up."*
Indicators regarding both processes and outcomes	• Process indicators indicate how care and support is currently provided. This information can be jointly discussed to learn from • Outcome indicators can help to identify the status quo and to check whether measures have effects. This information can be used to adapt policies and to account for expenditures to the city council. It stresses the importance to invest in children's health/ the first thousand days *"Initially, the process is of course most interesting, because that is where most will happen. But in 5 years I find poverty considerably more interesting because then I expect that what I have done in the process will have an effect on poverty."*
Indicators have the potential to be influenced (e.g. through policy)	• The indicators' potential to be influenced (through policy or other measures) is key to show short term successes *"I also see it as a good outcome measure: if you give extra help and support, this is often noticeable in the percentage of mothers who will breastfeed."*
Indicators show prevalence rates to use in making policy; both overarching (red flag) and specific	• Indicators that cannot easily be changed are also important to include in the indicator set if it concerns prevalence rates necessary to determine policy • Indicators showing a 'red flag' are important for monitoring since they provide a general picture and necessity to take measures • Specific prevalence rates on risk- or protective factors indicate which measures to tackle or which challenges to tackle *"Indeed, you cannot really change education level, but […] if you know that there are many low-educated people, you will take different measures than if you know that your population mainly consists of higher-educated people."*
Indicator set should be a balance between risk and protective factors	• Protective factors to vulnerability are often overlooked while they are very important *"It is of course very much about risk factors and I think there is an opportunity to look more at protective factors."*
Indicator set should provide a full picture of all relevant aspects	• The indicator set should provide a full picture of all relevant aspects *"It is important in the prioritization to have a total view across the board - so that the prioritized topics/indicators in the various phases say something about physical / mental / social / financial-work / environment-living / relationship-parenting / background / support / interventions (and for example not a lot of indicators on physical and none or little on mental [health])."*
Indicator set should provide a starting point of the conversation in a cross-sectoral collaboration	• Indicators that require the exchange of information in the local setting are required; collaborative partners can learn and work together based on this information • Preferably, indicators should not belong to individual care providers only, but cross domains. *"In any case, these are things you especially want to learn together."*
Indicators with data availability	• Data should be (easily) available on a local level *"For multiple topics, it's about whether they are available locally."*

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APPENDIX 4.
National indicator set

An overview of the indicator set that is used for monitoring the Solid Start action program on a national level, including each indicators' numerator, denominator and data source. This national indicator set was developed in 2019 (2). A Dutch version of the set of indicators (and its development), as well as the annual factsheets with a quantification of the indicators can be found online: www.rivm.nl/zorg/organisatie-van-zorg/kansrijke-start. Some indicators are formulated slightly different over the years, due to data-availability. The overview on this worksheet is based on the monitor in 2021.

Preconception			
Indicator	**Numerator**	**Denominator**	**Data source**
Percentage of municipalities with a local or regional coalition around the first thousand days of life	Number of municipalities with a local or regional coalition around the first thousand days of life	Number of municipalities (that receive additional subsidies for Solid Start)	Questionnaire among municipalities
Percentage of municipalities with a joint Solid Start-action plan	Number of municipalities with a joint Solid Start-action plan (completed or under development).	Number of municipalities (that receive additional subsidies for Solid Start)	Questionnaire among municipalities
Percentage of (central) municipalities that started the program 'Nu Niet Zwanger' (Not Pregnant Now)	Number of (central) municipalities that started the program 'Nu Niet Zwanger' (Not Pregnant Now)	Number of (central) municipalities	GGD GHOR Nederland (association for public health and safety in the Netherlands)

Pregnancy			
Indicator	**Numerator**	**Denominator**	**Data source**
Percentage of municipalities in which youth healthcare offers prenatal home visits	Number of municipalities in which youth healthcare offers prenatal home visits	Number of municipalities (that have answered the question)	Questionnaire among municipalities
Percentage of municipalities in which the program 'VoorZorg' (Nurse Family Partnership) is offered	Number of municipalities in which 'VoorZorg' (Nurse Family Partnership) is offered	Number of municipalities	Nederlands Centrum Jeugdgezondheid (Dutch centre of youth healthcare)
Percentage of midwifery practices trained in the program 'CenteringZwangerschap' (CenteringPregnancy)	Number of midwifery practices of which at least one midwife has followed a 'CenteringZwangerschap' (CenteringPregnancy) training.	Number of midwifery practices	Stichting Centering Nederland (foundation for Centering in the Netherlands)
Percentage of pregnant women who have their first antenatal care visit after the 10th week of pregnancy	Number of pregnancies from 24 weeks of gestation whereby the first antenatal care visit took place after the 10th week of pregnancy	Number of pregnancies from 24 weeks of gestation	Perined through DIAPER*

| Percentage of pregnant women with debts	Number of pregnant women with a registration of debt restructuring and/or delayed health insurance payments for more than six months in year of childbirth	Number of pregnant women in year of childbirth	CBS-microdata through DIAPER*
After birth			
Indicator	**Numerator**	**Denominator**	**Data source**
Percentage of youth healthcare organizations that offer the program 'CenteringOuderschap' (CenteringParenting)	Number of youth healthcare organizations that offer the program 'CenteringOuderschap' (CenteringParenting)	Number of youth healthcare organizations that offer care and support for parents during the first thousand days	Stichting Centering Nederland en TNO
Percentage of families not receiving postpartum care (at home) after birth	Number of live births of whom the mother had no declaration for postpartum care after birth	Number of live births	Vektis and CBS-microdata through DIAPER*
Percentage of children born in a family in a potentially vulnerable situation (three or more risk factors to vulnerability)	Number of live births born in a family with three or more of the following risk factors to vulnerability: low household income (<10th percentile), mental healthcare services use, use of medication related to psychological or psychiatric problems, having debts, detention, high healthcare expenditure, death of partner, divorce	Number of live births	CBS-microdata through DIAPER*
Percentage of children with a preterm birth or with a low birth weight for gestational age (SGA)	Number of children born after 22 weeks of gestation with a birth weight below the 10th percentile (according to Hoftiezer et al. (1)) and/or with a gestational age of less than 37 weeks	Number of children born after 22 weeks of gestation	Perined through DIAPER*
Percentage of children with a negative score on speech-language development around the age of two	Number of children with a negative score for the developmental characteristics 'says sentences of two words' and 'points out six body parts on a doll', during the contact moment with youth healthcare around the age of two	Number of children with available data on speech-language development	Inquiry among all youth healthcare organizations

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Percentage of children with overweight (including obesity) around the age of two	Number of children with a BMI score in the categories 'overweight' or 'obesity' during the contact moment with youth healthcare around the age of two	Number of children with available data on BMI	Inquiry among all youth healthcare organizations
Number of out-of-home placements for children before the age of 2 (per 1.000)	Number of children till the age of two who at any time received a youth protection measure for at least one day, overlapping with youth care with residence	Number of children till the age of 2	CBS-microdata through DIAPER*

* DIAPER (Data-InfrAstructure for ParEnts and childRen) is a nationwide population-based data infrastructure that integrates routinely collected data from three Dutch nationwide data sources (Perined, Vektis, Statistics Netherlands) at individual level. More information in Dutch can be found at www.rivm nl/diaper.

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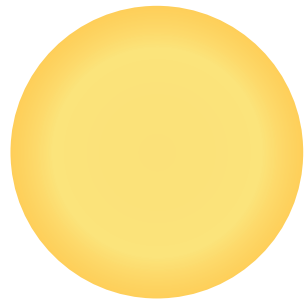
The Dutch Solid Start program: describing the implementation and experiences of the program's first thousand days

Joyce M. Molenaar
Inge C. Boesveld
Jeroen N. Struijs
Jessica C. Kiefte-de Jong
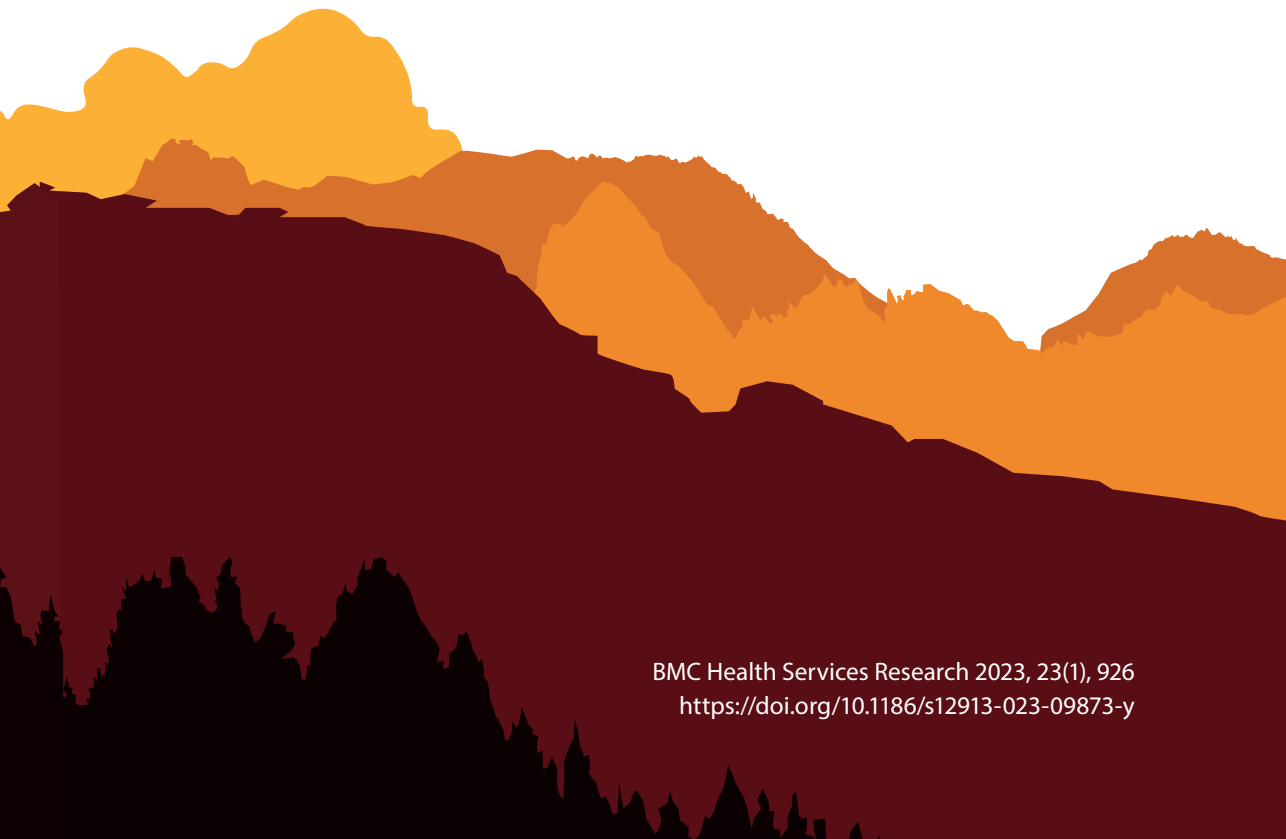
ABSTRACT

Background

In 2018, the Dutch government initiated the Solid Start program to invest in the first thousand days of life. A central element of the program is improving collaboration between the medical and social sector by creating Solid Start coalitions. This mixed-methods study aimed to describe the implementation of the Dutch Solid Start program, in order to learn for future practice and policy. Specifically, this paper describes to what extent Solid Start is implemented within municipalities and outlines stakeholders' experiences with the implementation of Solid Start and the associated cross-sectoral collaboration.

Methods

Quantitative and qualitative data were collected from 2019 until 2021. Questionnaires were sent to all 352 Dutch municipalities. Qualitative data were obtained through focus group discussions (n = 6) and semi-structured interviews (n = 19) with representatives of care and support organizations, knowledge institutes and professional associations, Solid Start project leaders, advisors, municipal officials, researchers, clients and experts-by-experience. Qualitative data were analysed using the Rainbow Model of Integrated Care.

Results

Findings indicated progress in the development of Solid Start coalitions (n = 40 in 2019, n = 140 in 2021), and an increase in cross-sectoral collaboration. According to the stakeholders, initiating Solid Start increased the sense of urgency concerning the importance of the first thousand days and stimulated professionals from various backgrounds to get to know each other, resulting in more collaborative agreements on cross-sectoral care provision. Important elements mentioned for effective collaboration within coalitions were an active coordinator as driving force, and a shared societal goal. However, stakeholders experienced that Solid Start is not yet fully integrated into all professionals' everyday practice. Most common barriers for collaboration related to systemic integration at macro-level, including limited resources and collaboration-inhibiting regulations. Stakeholders emphasized the importance of ensuring Solid Start and mentioned various needs, including sustainable funding, supportive regulations, responsiveness to stakeholders' needs, ongoing knowledge development, and client involvement.

Conclusion

Solid Start, as a national program with strong local focus, has led to various incremental changes that supported cross-sectoral collaboration to improve care during the first thousand days, without major transformations of systemic structures. However, to ensure the program's sustainability, needs such as sustainable funding should be addressed.

BACKGROUND

Preconception, pregnancy and the first two years of life (the first thousand days) are crucial for children's development and health, and a decisive period in the emergence of health inequities (1, 2). These avoidable differences in health outcomes (3) that start in early life pose an important challenge (2). Years of research that aimed to understand the nature and scope of health inequities showed both social and medical-related drivers, hence they should be addressed together in reducing health inequities (2, 4-6). Factors such as poverty, housing difficulties, stress and unemployment also highly influence health and wellbeing and cannot be addressed in the medical sector alone. Therefore, as stressed in several recent studies and reports, cross-sectoral collaboration between actors from the medical, social and public health sectors is considered essential to provide every child the best start in life (2, 7-10).

Internationally, multiple countries have implemented programs and policy reforms to reduce health inequities by integrating medical and social services in early life (11-14). In the Netherlands, the nationwide action-program 'Solid Start' (in Dutch: Kansrijke Start) was launched by the Dutch Ministry of Health, Welfare and Sport (Dutch abbreviation: VWS) in 2018 (15). The program aims to provide each child the best start in life by stimulating cross-sectoral collaboration, with a specific focus towards (future) parents and young children in vulnerable situations. The program strategy is based on the foundations of previous programs that aimed to integrate medical and social services, including the local 'Ready for a baby' program in Rotterdam (2008-2012) (16) and the subsequent 'Healthy Pregnancy 4-All' programs in several municipalities (since 2011) (7, 17, 18). Solid Start has a comprehensive population-based and upstream strategy, which means that its preventive and supportive measures aim to address the underlying factors that influence health and wellbeing at an early stage, in order to prevent or mitigate problems in later life. Policy measures were implemented for three periods: prior to pregnancy, during pregnancy and after birth, in order to prevent inequity and improve later health and well-being. The measures are aimed at preventing unintended pregnancies, preparing parents better for pregnancy, identifying medical and non-medical problems sooner, and supporting (future) parents in vulnerable situations better. The Dutch government financially supported municipalities to build a cross-sectoral approach for the first thousand days by forming or strengthening integrated 'Solid Start coalitions'. These coalitions consist of representatives of local organizations and providers working in the medical, social and public health domain, including midwives, obstetricians, maternity care assistants, youth healthcare providers, neighbourhood/social teams, social workers, debt counsellors, and municipal officials. The approach is supposed to be based on local data, challenges and existing networks. Hence, each municipality formulates its own objectives, agreements, actions and strategy to tackle the local problems.

Previous studies on collaboration during the first thousand days often focused on either the medical or social sector, or a specific temporal window such as pregnancy or after birth only. For example, several studies within the medical sector in the Netherlands (19-23) and

in other countries (24-26) reported on facilitators and challenges with interprofessional and interorganizational collaboration during pregnancy and childbirth. Some of the reported challenges were competition, suboptimal communication, power imbalances and fragmented structures, while facilitators included trust, feeling valued, formalized procedures and insight into each other's knowledge and competences (19-22, 24-26). Other studies that reported on integrated youth (health)care (27-29) found similar facilitators and challenges and also mentioned the need for further collaboration. Collaboration in maternity care is often described as complex and not self-evident, as healthcare providers historically have worked relatively autonomous with separated organizational structures, education programs, protocols, cultures and practices (8, 22, 30). More integrated care requires changes at different interrelated levels (micro, meso and macro), as outlined by Valentijn and colleagues (31).

Although these previous studies have furthered our understanding on collaboration, to date, there is limited knowledge into the development of cross-sectoral collaboration between the medical and social sector during the complete trajectory of the first thousand days as only few studies have devoted attention to this topic as a whole (7, 8). This knowledge is particularly relevant as we do not know if collaboration between sectors presents different challenges compared to collaboration within a sector, due to for example the larger differences in cultures and structures. Moreover, limited qualitative research has been conducted to comprehensively examine client experiences within the Dutch context (32), despite enhanced client experiences being one of the ultimate goals of cross-sectoral collaboration and integration. Existing studies primarily include either the perspectives of healthcare professionals and policymakers, or adopt a more quantitative approach (33, 34). The overall exploration of the implementation of Solid Start can be enriched if the viewpoints of those who provide, organize, examine ánd receive care are considered. Additionally, monitoring and reflecting on the development towards cross-sectoral collaboration during the implementation of a national policy program is important to support learning for future practice and policy developments in this direction.

Therefore, in this study, we aimed to describe the implementation of the Dutch Solid Start program during 2019, 2020 and 2021. We formulated the following two research questions: 1) To what extent is the Solid Start program implemented within municipalities? 2) What are the experiences of stakeholders with the implementation of the Solid Start program and cross-sectoral collaboration?

METHODS

Research design
The first research question was answered by using quantitative data from questionnaires among municipalities. The second research question was answered with qualitative data from interviews and focus group discussions (FGDs). We had several rounds of data collection in subsequent years after the implementation of the nationwide Solid Start program in September 2018 (Figure 1).

Study setting
The national Solid Start program was launched by the Dutch government in September 2018. The Ministry of Health, Welfare and Sport facilitated various (supportive) measures, including the possibility for municipalities to request Solid Start funds at three time points (March 2019, October 2019 and April 2020) to start building or strengthening their local Solid Start coalition. The funds could be utilized at municipality level to start a local coalition, but municipalities could also choose to pool their resources and collectively work towards a (sub-)regional approach or coalition with other municipalities. Municipalities were in the lead to create coalitions of partners from the medical and social sector who jointly made agreements about care and support during the first thousand days and to families in vulnerable situations. Some basic elements of these coalitions were set (35). Municipalities received support from Pharos (the Dutch Centre of Expertise on Health Disparities) to build their coalition. Additionally, municipalities were able to use an analysis tool to map the current and envisioned situation, an inventory of effective interventions, key local, regional and national data, and inspiration from successful examples across the country.

Appendix 1 provides a description of the Dutch care and support system during the first thousand days. This study was part of the national monitor of the Solid Start program that is conducted by the National Institute for Public Health and the Environment (Dutch abbreviation: RIVM) by commission of the Ministry of Health, Welfare and Sport. The RIVM monitors the implementation of the Solid Start program by collecting both quantitative data on process- and outcome indicators as well as qualitative data on experiences and developments. Since 2021, the RIVM also provides support to municipalities in monitoring their local approach. Appendix 2 provides an overview of the national and local monitor.
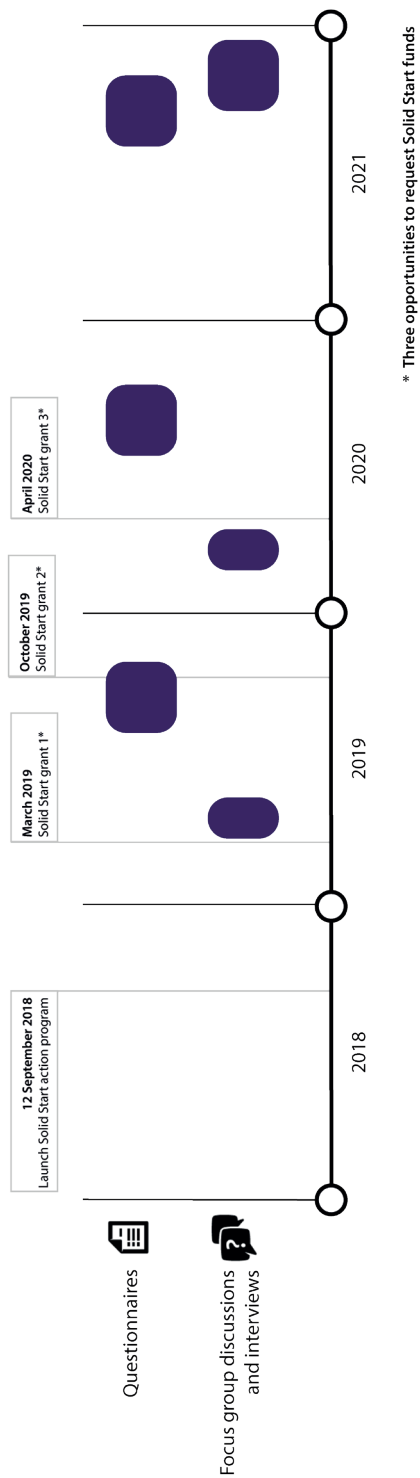
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Figure 1. Timeline data collection

Quantitative data collection

Participants
The questionnaire was distributed among all municipalities that requested funds in 2019 (N = 147) and among all municipalities -including those without funds- in 2020 and 2021 (N = 355 and 352 respectively). The Ministry of Health, Welfare and Sport invited the municipalities that requested funds to participate in the questionnaire, the Association of Dutch Municipalities (Dutch abbreviation: VNG) invited the other municipalities to participate in the questionnaire.

Data collection
The online questionnaire focused on the local implementation of Solid Start and consisted of questions about municipalities' development towards Solid Start coalitions. The questions mainly had closed answer categories and were slightly different each year depending on national developments and new insights. The questions that were relevant to this article and comparable over the years included the following topics: Solid Start funds, local coalition, action plan, goals and ambitions, partners, activities, monitoring, support and COVID-19. Examples of questions included: 'Has your municipality formed a Solid Start coalition?' and 'What is the status of monitoring Solid Start in your municipality?' An overview of the questions can be found in Table 2 (results section).

Data analysis
Quantitative data were analysed using descriptive statistics. We used Excel and R to calculate frequencies and percentages.

Qualitative data collection

Participants
For the interviews and FGDs, we used purposive sampling to ensure heterogeneity (36). We invited representatives of care and support organizations (managers and care providers), Solid Start project leaders or advisors, other municipal officials, representatives of national knowledge institutes and professional associations, and researchers to join a FGD at a predefined time. In 2021, we organized individual interviews with those not available if their perspective was otherwise missing. Additionally in 2021, we invited clients and experts-by-experience for individual interviews at their preferred time and place, because we wanted to create the conditions in which they felt comfortable to share their personal stories in more detail than possible during a FGD. Clients received care and support during the first thousand days at the time of the interview. The experts-by-experience had collective experiential knowledge or were trained in using personal and collective experiences to support families in vulnerable situations. Most participants received an invitation to participate directly through an e-mail by the research team. One of the experts-by-experience supported the recruitment of clients by providing them information and discussing a feasible date and place.

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Table 1. Topics in FGDs and interviews

General topics
• General experiences with Solid Start within the organization/ municipality/ region • Involved parties • Collaboration between medical and social sector (in the formation of coalitions and in daily practice) • Facilitators: what went well, factors that facilitated development • Barriers: what went wrong, factors that impeded development • Needs for the future and priorities

Year-specific topics		
2019 (shortly after the start of the program in sept. 2018) • Transition: before and after implementation of Solid Start • Relation between previous/ current initiatives and Solid Start	*2020* • Funding and financing • Objectives and monitoring • Knowledge exchange	*2021 (shortly before the end of the initial program)* • Effects/ added value of Solid Start • Continuity of the program • Involvement of experts-by-experience • Early detection (screening) • Support for professionals • Solid Start as example for other sectors?

Data collection

The qualitative data were collected online (2020 and 2021, as a consequence of COVID-19 regulations) or live (2019 and several interviews in 2021). The interview guide focused on the experiences with the implementation of the Dutch Solid Start program and included a series of fixed open questions that were similar in each interview or FGD, and flexible questions adapted to the type of respondents or year of data collection to reflect the progress of the Solid Start program. Table 1 provides an overview of the main topics. FGDs lasted between 70 to 110 min. Interviews lasted on average 35 min, ranging from 11 to 52 min. All individual interviews were held one-on-one, with some exceptions. The expert by experience who assisted with client recruitment was also present during these interviews with clients to provide reassurance to clients and ask supplementary questions to gain more meaningful insights. Additionally, 4 project leaders and advisors within the same coalition were interviewed together.

Data analysis

All interviews and FGDs were audio-recorded, transcribed verbatim and analysed in MaxQDA. We conducted a thematic analysis based on deductive coding, while remaining open to add relevant elements emerging from the data. A coding frame was set based on the Rainbow Model of Integrated Care (RMIC) by Valentijn et al. (2013). The RMIC was developed as a framework to describe integrated care in 6 interrelated dimensions (Figure 2). Integrated care, in our paper, refers to the collaborative efforts of multiple professionals and organizations across the medical and social care system to provide comprehensive, accessible, and coordinated care for the benefit of (future) parents and their children (37, 38). The RMIC outlines contact between client and care provider at microlevel (clinical integration), collaboration between professionals and organizations at mesolevel (professional- and organizational integration) and the wider policies and rules within the health system that influence collaboration at macrolevel (system integration). These levels are linked and enabled through supportive structural functions such as resources- and
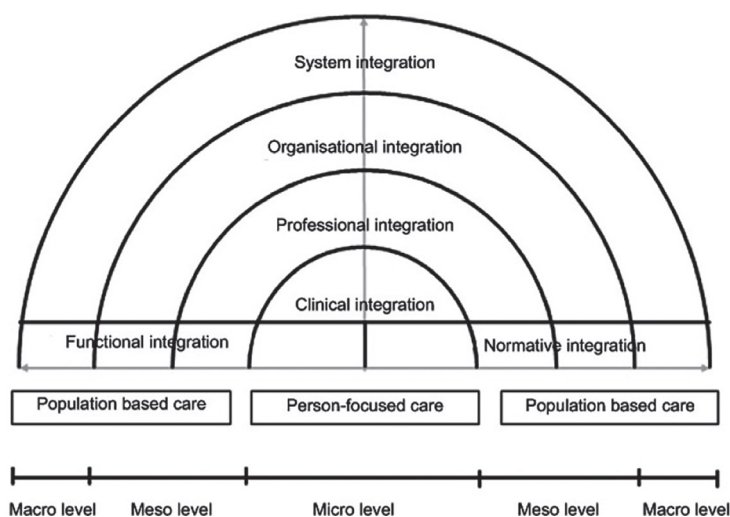
Figure 2. Rainbow model of integrated care (RMIC) by Valentijn et al. (2013)

information management (functional integration) and softer aspects including shared vision, culture and informal collaboration (normative integration). The six dimensions are outlined in a taxonomy of 59 key features (38). We used these 59 key-features for coding and described the results according to the 6 dimensions. Two authors (JM and IB) coded the first 2 transcripts independently and compared coding to refine the coding frame. Next, JM coded all transcripts and IB cross-checked coding for three transcripts. The codes were analysed and discussed in several meetings with the research team. Doing so, we sought for links between levels of integration within the RMIC and for patterns over the years.

RESULTS

The results are presented in two sections according to the research questions. In part one, we explain to what extent the Solid Start program is implemented within municipalities. In part two, we outline the experiences of stakeholders with the implementation of the Solid Start program and cross-sectoral collaboration.

Development towards Solid Start coalitions
There were 355 municipalities in the Netherlands in 2019 and 2020, whereas there were 352 in 2021 due to merging. Municipalities had the opportunity to request the Solid Start funds from the Dutch government at three time points: March 2019, October 2019 and April 2020. The first two rounds were only open to a specific group of 150 municipalities that joined the national Health In The City program (in Dutch: Gezond In De Stad), focused on tackling health inequalities at local level. The number of municipalities that requested funds increased from 98 in March 2019 to 275 in April 2020 (Figure 3).
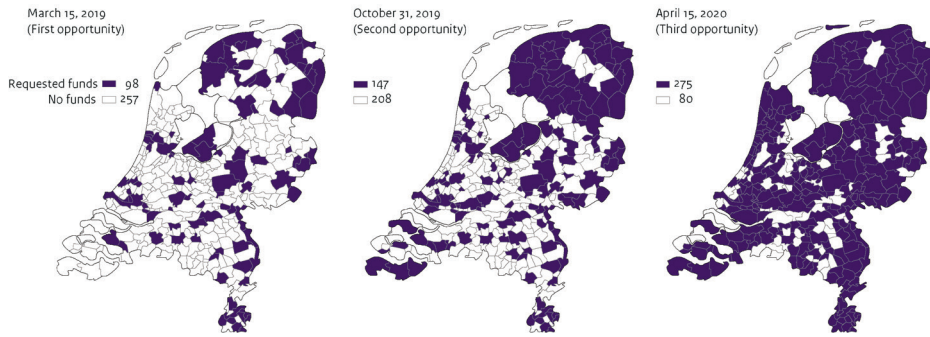
Figure 3. Municipalities that requested the Solid Start funds

Solid Start coalitions

Municipalities completed an online questionnaire in 2019 (n = 140), 2020 (n = 251) and 2021 (n = 217) (Table 2). Figure 4 shows the number of municipalities reporting to have formed a local coalition across the country. The numbers increased since 2019 (n = 40), especially from 2020 (n = 59) to 2021 (n = 140). Around half of the municipalities that had a coalition in development in 2020, reported to have formed their coalition a year later. In 2021, 65% (n = 140) of the responding municipalities that received funding formed a coalition. More than half of them collaborated with other municipalities (Table 2).
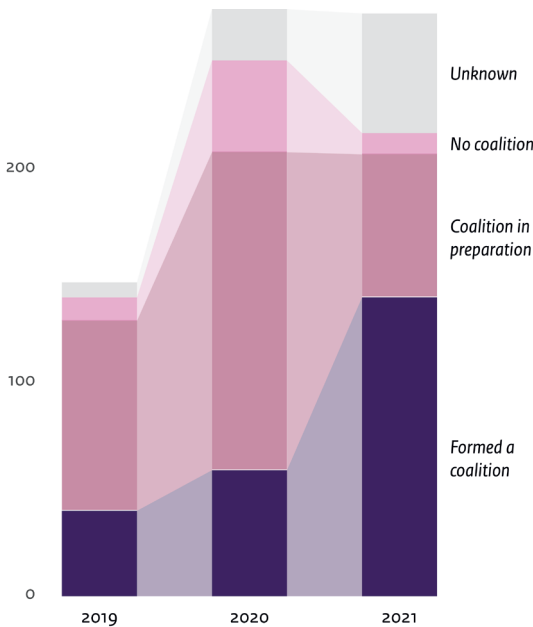

Figure 4. Development of coalitions during 2019 – 2021.
The figure shows municipalities' answers to the question "Did you form a local Solid Start coalition?"

Table 2. Overview of quantitative findings

	2019	2020	2021
Number of municipalities in the Netherlands	355	355	352[d]
Number of municipalities that requested the Solid Start funds	147	275	273[d]
Number of municipalities that have been invited to participate in the questionnaire	150[b]	355	352[d]
Number of municipalities that responded to the questionnaire	140	289	229
Response among municipalities with the Solid Start funds	140 (95%)	251 (91%)	217 (79%)
Response among municipalities without the Solid Start funds	Not applicable	38 (48%)	12 (15%)

Topics	Questions + answer categories	n (% among all municipalities with Solid Start funds, % among all municipalities [c])		
		2019	**2020**	**2021**
Coalition	**Has your municipality formed a Solid Start coalition?**			
	Yes	40 (27%, 11%)	59 (21%, 17%)	140 (51%, 40%)
	No, not yet. We are in talks/ preparation for forming a coalition	89 (61%, 25%)	149 (54%, 42%)	67 (25%, 19%)
	No	11 (7%, 3%)	43 (16%, 12%)	10 (4%, 3%)
	Is your municipality collaborating with other municipalities to form or strengthen a (sub-)regional coalition?			
	Yes	No data	157 (57%, 44%)	138 (51%, 39%)
	No	No data	94 (34%, 26%)	79 (29%, 22%)

Topics	Questions + answer categories	n (% among all municipalities responding to the question, % among all municipalities [c])		
		2019	**2020**	**2021**
Plan of action	**Has your municipality developed an action plan or roadmap together with collaborative partners for Solid Start?**			
	We have an action plan	26 (19%, 7%)	39 (14%, 11%)	73 (32%, 21%)
	We have started an action plan	48 (34%, 14%)	100 (35%, 28%)	83 (36%, 23%)
	No	23 (16%, 6%)	148 (52%, 42%)	72 (32%, 20%)
	Other	43 (31%, 12%)	-	-

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Table 2. Continued.

Objectives		2019	2020	2021
Has any objectives been set for Solid Start within your coalition/municipality?[a]				
	Yes	No data	44 (15%, 12%)	100 (44%, 28%)
	In development	No data	155 (54%, 44%)	92 (41%, 26%)
	No	No data	87 (30%, 25%)	29 (13%, 8%)
	Other	No data	17 (6%, 5%)	5 (2%, 1%)
If yes / in development/ other, what time period(s) are the objectives focused on?[a]				
	Before pregnancy	No data	171 (81%, 48%)	156 (79%, 44%)
	During pregnancy	No data	200 (95%, 56%)	185 (94%, 53%)
	After birth till the age of 2	No data	195 (92%, 55%)	184 (93%, 52%)
	After birth till the age of 4 or 18	No data	77 (36%, 22%)	60 (30%, 17%)
Have joint ambitions around Solid Start been formulated within your municipality/coalition?				
	Yes	68 (49%, 19%)	122 (43%, 34%)	166 (72%, 66%)
	No	72 (51%, 20%)	165 (57%, 46%)	63 (28%, 25%)
Is Solid Start part of a wider policy framework?				
	Yes	No data	250 (87%, 70%)	196 (86%, 56%)
	No	No data	36 (13%, 10%)	32 (14%, 9%)
Have collaborative agreements been made on an approach to the first thousand days and families in vulnerable situations?[a]				
	Yes, at the implementation level	54 (39%, 15%)	95 (33%, 27%)	137 (60%, 39%)
	Yes, at the managerial/policy level	19 (14%, 5%)	44 (15%, 12%)	60 (26%, 17%)
	No	85 (61%, 24%)	171 (60%, 48%)	82 (36%, 23%)
Are there any activities within your municipality on the topic of Solid Start?				
	Yes	No data	198 (70%, 56%)	182 (80%, 51%)
	No	No data	86 (30%, 24%)	46 (20%, 13%)
If yes, when did these activities begin?				
	After the implementation of the Solid Start program (September 2018 – present)	No data	101 (53%, 28%)	122 (67%, 35%)
	Before the implementation of the Solid Start program (before September 2018)	No data	89 (47%)	60 (33%, 17%)

Table 2. Continued.

		2019	2020	2021
Monitoring	**What is the status of monitoring Solid Start in your municipality? [a]**			
	We monitor the progress (process) of our program (e.g. implementation of interventions)	No data	59 (21%, 17%)	71 (31%, 20%)
	We monitor the outcomes of our program (e.g. pregnancy outcomes)	No data	33 (12%, 9%)	22 (10%, 6%)
	We have plans to monitor progress	No data	105 (37%, 30%)	102 (45%, 29%)
	We have plans to monitor outcomes	No data	81 (29%, 23%)	75 (33%, 21%)
	None of the above	No data	112 (39%, 32%)	43 (19%, 12%)
	Do you have insight in the statistics and facts concerning the first thousand days in your municipality? (2019)/ Did you conduct a baseline assessment to gain insight in the statistics and facts concerning the first thousand days in your municipality? (2020 and 2021)			
	Yes	102 (73%, 29%)	118 (42%, 33%)	154 (68%, 44%)
	No	38 (27%, 11%)	166 (58%, 47%)	71 (32%, 20%)
COVID-19	**Has the COVID-19 pandemic affected activities and progress of Solid Start in your municipality/coalition? [a]**			
	Yes, it has caused a delay	No data	197 (70%, 55%)	167 (75%, 47%)
	Yes, other (e.g. changes in the approach/ strengthening of collaboration/ fewer financial resources for Solid Start)	No data	8 (3%, 2%)	16 (7%, 5%)
	No, it has not had any major consequences so far	No data	68 (24%, 19%)	52 (23%, 15%)
Partners	**Which parties are part of the local coalition or with which parties do you collaborate? [a] (see Figure 5 for an overview of specific partners in 2021)**			
	At least 1 partner in the medical sector	110 (79%, 31%)	225 (78%, 63%)	217 (95%, 86%)
	At least 1 partner in the social sector	120 (86%, 34%)	262 (91%, 74%)	224 (98%, 89%)
	At least 1 partner within the municipal organization (other departments)	96 (69%, 27%)	255 (89%, 72%)	220 (96%, 87%)
	At least 1 partner within the community	No data	No data	83 (36%, 33%)
	Which parties that are not currently involved would you like to consult with? (top 3) [a]			
	General practitioners	No data	No data	77 (34%, 22%)
	Experts-by-experience	No data	No data	55 (20%, 16%)
	Health insurers	No data	No data	44 (16%, 13%)

[a] Multiple answers allowed
[b] In 2019, only Health In The City municipalities (in Dutch: Gezond In De Stad (GIDS) gemeenten) were requested to participate (n = 150). At that time, they were the only municipalities that could request the Solid Start funds
[c] The percentage in relation to the total number of municipalities is often an underestimation, given the large number of missing values, particularly for municipalities that did not apply for the Solid Start funds. All percentages can be considered conservative, representing a minimum lower limit; the actual percentage is likely to be higher.
[d] The number of municipalities changed during the years due to the merging of municipalities

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Development within municipalities
Over the years, the number of municipalities with a plan of action, objectives, ambitions and activities increased (Table 2). By 2021, almost all responding municipalities (85%) were engaged in setting objectives. More than one in four municipalities set objectives aimed at a longer period (children until 4 or 18 years) than the original Solid Start program (up to 2 years), and Solid Start was almost always part of a wider policy framework. In 2021, 64% of the responding municipalities made collaborative agreements about the Solid Start approach at implementation level, managerial/policy-level, or both. Moreover, 80% of the responding municipalities reported having activities on the topic of Solid Start, and two-thirds of them started these activities in the timeframe after receiving the Solid Start funds. The quantitative data also showed that several municipalities started with monitoring Solid Start, and many reported having plans to monitor. Municipalities reported that they more often monitored processes than outcomes. Additionally, 68% of the responding municipalities in 2021 conducted a baseline assessment to gain insight into the statistics and facts concerning the first thousand days in their municipality. Three-quarters of the municipalities indicated that COVID-19 influenced Solid Start activities and progress in 2020 and 2021; it mostly caused a delay.

Involved stakeholders
There was a wide variety of stakeholders involved in Solid Start. Figure 5 shows which parties municipalities mentioned when they were asked who is part of the local coalition or with whom they collaborate. Most often mentioned were midwives, maternity care assistants, youth healthcare, Public Health Services, neighbourhood/social teams and policy makers within other municipal departments on the topics of youth healthcare and public health. In 2021, around one-third of the municipalities collaborated with experts-by-experience or other community-partners (Figure 5). General practitioners (GPs), health insurers and experts-by-experience were most often regarded as missing parties (Table 2).

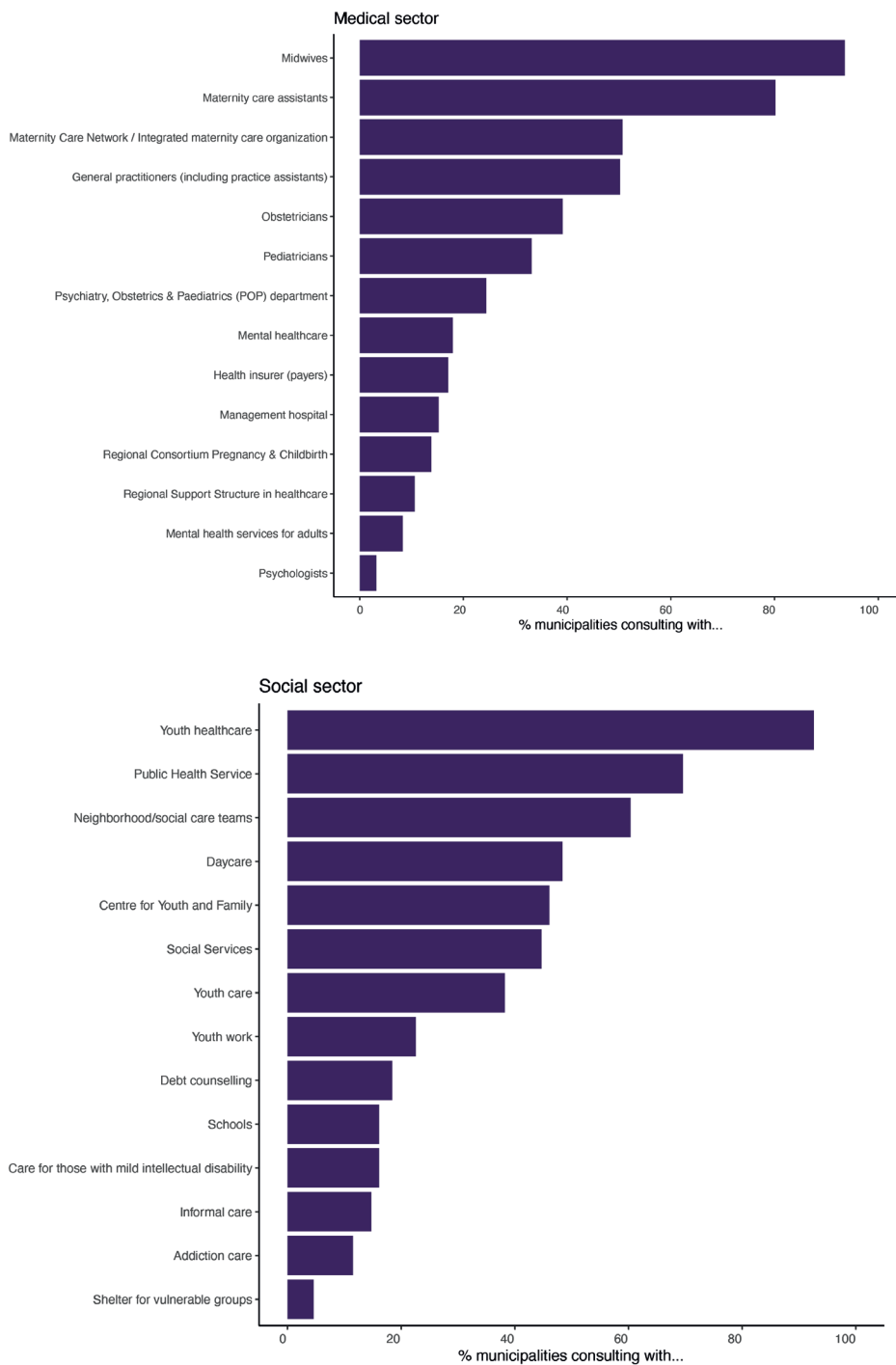## Medical sector

Social sector

Figure 5a. Parties that are part of the local coalition or with whom municipalities collaborate
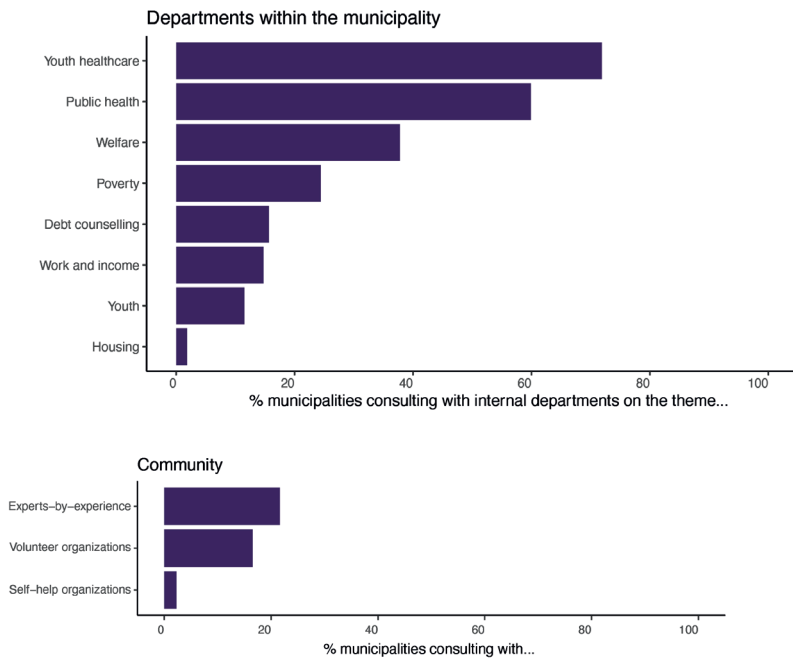
Figure 5b. Parties that are part of the local coalition or with whom municipalities collaborate

Experiences with the implementation of the Solid Start program and cross-sectoral collaboration

A total of 6 FGDs and 19 interviews were conducted, as detailed in Table 3. The findings were outlined in the six dimensions of the RMIC and summarized in Table 4. Table 4 presents an overview of both positive experiences and developments in the implementation of Solid Start and cross-sectoral collaboration, as well as the challenges that remain and the needs for improvement.

In the next sections, we explain the key results, provide examples and highlight the interconnections between different levels of the RMIC. The order of the dimensions was determined by the stakeholder's narratives. As normative integration seemed to be a fundamental step towards more integration in relation to Solid Start, this dimension was positioned at the top of the table and discussed first.

Table 3. Number of participants in FGDs and interviews

	Total (unique)[a]	2019	2020	2021
Representatives of care and support organizations (both managers and healthcare providers)	14	7	4	4 (incl. 1 individual interview)
Social sector	7	5	2	0
Medical sector	7	2	2	4
Solid Start project leaders or advisors	18	6	4	12 (8 individual interviews and 2 interviews with 2 respondents from the same coalition)
Other municipal officials	4	2	2	NA
Researchers and representatives of national knowledge institutes and professional associations	18	6	8	10 (incl. 1 individual interview)
Social sector	9	4	3	4
Medical sector	9	2	5	6
Clients and experts-by-experience	7	0	0	7 (all individual interviews)
Data collection	6 FGDs; 19 interviews	2 FGDs	2 FGDs	2 FGDs; 19 interviews (with 1 or 2 respondents)

[a] Some stakeholders participated in 2 or 3 rounds.

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Table 4. Overview of qualitative findings

Dimensions	Positive experiences and recent developments	Challenges ahead and needs for improvement
Normative integration	• Increased sense of urgency of importance first thousand days • Increased mutual acquaintanceship (knowing each other) • Visionary leaders facilitated Solid Start (e.g. national advocates and local 'coalition of the willing')	• Further transcending domain perceptions and overcoming cultural differences and fragmented structures
Professional integration	• Agreements on interdisciplinary collaboration have increased • Multidisciplinary guidelines, protocols, interventions and policies for Solid Start have increased • Shared goal to provide every child a solid start stimulated collaboration • Experiencing value creation ('what's in it for me as a professional') stimulated collaboration	• Successful implementation of agreements, guidelines, protocols, interventions and policies in practice • Integration of Solid Start into all professionals' daily practice
Organizational integration	• Centering the needs of the target population as binding agent stimulated collaboration • Dedicated initiators or project leaders were a driving force • Increased learning and knowledge sharing • Increased monitoring and evaluation • Learning community to support the setup of local monitoring • Experiencing value creation ('what's in it for the organization') stimulated collaboration • Support from (executive) board members and aldermen	• Continuation of driving forces at institutional level • Challenges related to organizational features • Complexity in one sector hinders cross-sector collaboration • Challenges in monitoring like data-availability, selecting relevant indicators, correct interpretation • Continuing learning between and within Solid Start coalitions • Learning from sectors other than Solid Start (e.g. elderly care) and disseminating knowledge based on Solid Start experiences to other sectors
Clinical integration	• Increased attention for continuity of care, case management and client-centered care • Improved interpersonal interaction between clients and professionals • Increased client involvement in the organization of care • Increased client involvement in daily practice (shared decision-making) • Learning programs to support client involvement	• Further shifting from supply-oriented care and support to prioritizing clients' needs • Improving interpersonal interaction • Standardizing client involvement • More involvement of partner/spouse and wider informal network • Better focus to clients' experiences and satisfaction for improvements • Improving the completeness, diversity and communication of client-information to enhance alignment and identification.
Functional integration	• Support for coalition building and implementation of Solid Start activities at local level	• Integrated information system to share information between professionals

Table 4. Continued.

Dimensions	Positive experiences and recent developments	Challenges ahead and needs for improvement
System integration	• Solid Start funds facilitated implementation on local level • Increased involvement stakeholders from social and medical sector • The Solid Start program's structure was appreciated for its governmental stewardship and strong local focus • Solid Start creates social value at system level • Previous local cross-sectoral projects targeted at health and disparities (during pregnancy) were used as starting point	• Ensuring structural and sustainable funds for long-term planning • Involving more GPs, health insurers, clients-by-experience • Adapting the scope of laws and regulations to stimulate Solid Start, with regard to cross-sectoral collaboration and task-division • Aligning integration with coalitions' and professionals' needs for support • Facilitating knowledge development and dissemination • Acknowledging Solid Start as ultimate form of prevention • More insight into impact, cost-effectiveness and success factors to maintain its prioritization and political support at local level • Solid Start is regarded as a transition rather than an innovation; transitions are complex and time-consuming • Balancing workload, limited time and capacity (workforce shortage) with adequate care and support is challenging • COVID-19 pandemic decreased funds, manpower and priorities for Solid Start

Normative integration

The experiences of stakeholders seemed to reveal that normative integration was the starting point for more collaboration and integration in relation to Solid Start. During almost all conversations, stakeholders stressed how Solid Start created more sense of urgency regarding the importance of the first thousand days and prevention, and feelings of collective responsibility to coordinate care and support for parents and children. This increased sense of urgency had implications at different levels (micro, meso and macro) and was a starting point to initiate or intensify activities. However, especially in 2019, stakeholders also described difficulties in deciding where and how to begin with the local implementation of Solid Start. Most municipalities started building their coalition by organizing a kick-off meeting with relevant parties to discuss current workflows, challenges and strengths. These and other meetings contributed to mutual acquaintanceship between individuals from different organizations, as they got to know each other and gained insight in each other's expertise. This led to quick gains such as the exchange of contact details and casuistry, and long-term benefits such as increased trust, understanding, learning and contact for future clients. This quote of a participant in a FGD illustrates how getting to know each other can improve the collaboration:

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> *"And I think, the moment we know about each other, what each other's expertise is, what you're good at, how you can support the other, that's already very helpful to be able to start forming a local coalition and to start organizing care together around vulnerable pregnant women."* - FGD, 2020

Stakeholders described how the historical separation and fragmentation between the medical and social sector was persistent and challenging to overcome. Involved organizations often had different cultures, languages, ways of working, legislations, structures, focus areas, networks and missions, which were frequently mentioned as barriers to collaboration. Practical examples included differences in working hours that impeded finding a time to meet. Other examples included a difference between working supply-oriented or demand-oriented, curative versus preventive, focused on children versus parents, and no common understanding of vulnerability. Stakeholders expressed a need for providers to move beyond their own professional perspectives, to further transcend domain perceptions, and overcome cultural differences and fragmented structures. Besides getting to know each other, elements such as developing a shared vision and objectives, and joint multidisciplinary education were considered as helpful.

Professional integration
At the professional level, stakeholders reported more agreements on interdisciplinary collaboration. Over the past years, there has been an increase in multidisciplinary guidelines, protocols, interventions and policies for the first thousand days. For example, multiple stakeholders reported the initiation or expansion of multidisciplinary meetings and joint intakes. Moreover, there has been an increase in the use of structured risk screening tools that focused on both medical and social factors. Additionally, tailored multidisciplinary care pathways for vulnerable pregnancies have been developed or refined to ensure timely and appropriate referral. However, the agreements made did not ensure successful implementation in practice, due to several reasons. For example, the high number of professionals made it difficult to get everyone together, and sometimes there was sufficient funding to develop guidelines but not enough to implement them, despite a stakeholder's view that *"implementation remains most important, regardless of all the documents"* (FGD, 2021). Implementation in practice was considered an ongoing challenge and stakeholders wished for greater alignment in processes in the coming years.

Additionally, notwithstanding numerous developments at the professional level, the Solid Start program and the need for collaboration are not yet fully incorporated into all professionals' everyday practice. Stakeholders have emphasized the need for everyone to acknowledge its importance and take responsibility. As one stakeholder stated: *"There is a need for change, there is potential for change, if we do it together."* (FGD, 2021) Several stakeholders agreed that this can be stimulated by including Solid Start in curricula and professional profiles. Moreover, professionals must receive practical tools, adequate support, and training to enhance their competences. These competences include, but are not limited to, effective communication and interacting with clients in a cultural and stress sensitive way.

Organizational integration

The Solid Start program enabled organizational integration by centering the needs and preferences of the target population as a binding agent at the core of all activities. One of the stakeholders explained this by noting: *"What the added value is, is the focus on the child. And not just on disciplines or sectors, domains and acquired practices."* (FGD, 2020). Moreover, a dedicated local initiator, project leader or coordinator as driving force was considered essential for coalitions' progress. Someone who brings parties together, facilitates and takes an ambassadorial role. Despite differences, this position was often filled by someone from Public Health Services, a regional support structure, the municipality, or another neutral party. Stakeholders provided examples where the development halted when that person left. Therefore, they suggested that these driving forces should be institutionalized and supported financially and practically in the future.

Several challenges that arose at the organizational level were related to different organizational features. For example, municipalities and care and support organizations had different structures and their physical working areas often did not completely overlap. The social sector was described as fragmented, in contrast to birth care in which organizations often united in obstetric partnerships. Additionally, several organizations, including youth healthcare (preventive public health services to promote health and development for children from birth till the age of eighteen), work in multiple municipalities. The differences and fragmentation made it harder to reach agreements between organizations. Stakeholders also mentioned how the perceived difficulties arising from developments within one organization or sector (e.g. integrated birth care and transitions in youth care) could complicate cross-sectoral collaboration for Solid Start as well.

Learning and sharing knowledge were frequently mentioned as essential to improve integration. Stakeholders highlighted how the existence and design of Solid Start fostered learning opportunities. Municipalities sought to learn from best practices in other municipalities in order to avoid unnecessary duplication of efforts. They did so for topics ranging from 'developing a local approach with a comprehensive set of interventions' in 2019 to 'monitoring and ensuring/embedding the approach' in later years. One of the stakeholders mentioned: *"[...] we also keep a close watch on what other regions are doing, what can we learn from them?"* (FGD, 2020). As such, stakeholders emphasized the importance of learning and knowledge sharing in the future, both between and within coalitions.

The qualitative data showed that municipalities started with monitoring and evaluation. Examples were provided about discussing data and indicators on processes and outcomes during the first thousand days with professionals at municipal or neighbourhood-level, in order to understand local developments and prioritize future actions. However, many municipalities had questions regarding monitoring, such as: which indicators to include, how to start monitoring, where to find data and how to interpret the data? Support from RIVM's learning community to aid the setup of local monitoring was appreciated.

Clinical integration

Primarily clients, experts-by-experience and professionals have reported on the concepts of continuity of care, case management and client-centered care. These concepts, which prioritize the central role of clients' needs, have gained increasing attention in recent years and have come to influence the provision of care and support. For example, several local coalitions engaged in discussions on how (future) parents navigate care and support provided during the first thousand days to uncover areas for improvement. Despite progress, stakeholders mentioned that care and support were still too much driven by policy and professionals (supply-oriented) and prioritizing clients' needs was not yet routine practice. Stakeholders expressed the need to better address the multiple challenges faced by (future) parents in vulnerable situations (e.g. related to housing, work, education and parenting). This requires restructuring and improved communication among the professionals involved. In some areas, case managers were appointed or central telephone numbers for referrals have been implemented. One of the clients described her experiences with having one case manager:

> *"I had one person I could share everything with, so that was very nice. [...] [she had] conversations with me about how I feel, but also about finances."* - Client, 2021

Stakeholders also reported that although improvements have been made in the interpersonal interaction between professionals and clients, there remains a need for further development. Clients and experts-by-experience shared both positive and negative experiences. Positive experiences were associated with the keywords empathy, understanding, respect, transparency, safety, trust, and being heard and understood. Negative experiences, however, were marked by incidents of prejudice, judgement and underestimation. To enhance interpersonal interaction, stakeholders have emphasized the need for training in sensitivity and communication. Everyone is different and '[…] *to me this means that you really look at the person and the situation.'* (Expert-by-experience, 2021).

Lastly, stakeholders have noted increased client involvement, both in the organization of care and in daily practice. For example, several organizations established parent or client councils, and the Ministry of Health, Welfare and Sport invited a group of experts-by-experience to reflect on national policy measures since mid-2020. However, stakeholders also mentioned the need to expand and standardize client involvement for quality improvement. They mentioned challenges including how to start and involve the right people, and emphasized that it is important to consider financial reimbursements. Mainly since 2021, client involvement became a more central topic for coalitions and Pharos started to organize learning programs to support this effort. In daily practice, shared decision-making and positive health principles supported client involvement, allowing for putting parents' needs and preferences first in decisions concerning their own health and well-being.

Functional integration

Pharos has supported municipalities since 2019 in building their coalition, which was highly valued. Municipalities had varying needs for support, depending on the coalitions' developmental stage and other factors. The need for one-on-one support seemed to have shifted towards a need for mutual knowledge-exchange over time. As previously explained, stakeholders requested additional support for professionals to incorporate Solid Start into everyday practice. A participant in a FGD said:

> *"Ultimately, you do it for the children and their parents, but you need to give the professionals tools to be able to continue to do this."* - FGD, 2021

The FGDs revealed difficulties in sharing information between professionals, particularly in the context of referrals. This was complicated by General Data Protection Regulations according to the stakeholders. Some stakeholders called for an integrated information system and more transparency. Although digital data exchange in birth care has been in development for a few years, it was not yet standard practice.

System integration

We have found several systemic determinants that influenced collaboration at meso- and microlevel. Overall, most challenges that arose in the interviews and FGDs seemed to concern systemic integration. Hence, stakeholders highlighted a range of needs that should be addressed in order to embed Solid Start and ensure the program's sustainability. One of the stakeholders explained her view, which was supported by many others:

> *"It is really a transition from the system as it was, you know, quite a fragmented system. […] Even four years is very short for that, right? So you're really setting a movement in motion, and I think that program is really setting that in motion. But it is really a long-term issue, simply because you are changing a lot of things. [...] When you really want to get it into the system, and therefore want to see improvements in collaboration everywhere, then these annoying prerequisites come up again, right? Then you have to make sure that financing follows as well, that it supports care instead of getting in the way, for example. Those kind of things."* - FGD, 2021

In relation to available resources, stakeholders mentioned that the Solid Start funds helped to start activities at local level. However, the funds were frequently described as limited, temporary and project-oriented, thereby impeding long-term planning. Municipalities reported difficulties to obtain funds for interventions, and to bring partners together without reimbursements. Stakeholders noted that funds were often invested in innovation and curation rather than implementation and prevention. Moreover, they generally mentioned unclarity regarding prevention. For various preventive activities related to Solid Start, it was unclear to the stakeholders whether the municipality or health insurers should bear the financial responsibility, resulting in occasions where funds were unavailable. There are five different Dutch laws that include prevention, which complicated the financing and funding thereof. Another difficulty was that investing in preventive measures can be

uncertain and may not always benefit the investor (wrong pocket issue). Over the years, but peaking in 2021, stakeholders have called for structural and sustainable funding to ensure Solid Start's sustainability.

Next, stakeholders noted increased involvement of organizations and professionals from the medical and social sector. The composition of coalitions varied based on factors such as the municipalities' focus, challenges and historical context. General practitioners, health insurers and experts-by-experience were mentioned as major missing parties. Stakeholders anticipated that GPs, who are potentially vital in preconception care, were often unavailable due to their heavy workload and because they did not view Solid Start as a core activity. Health insurers were seen as a potential source of funding for preventive activities, although discussions about this were experienced as difficult due to the health insurers' focus on individuals (indicated prevention) rather than on groups (universal or selective prevention).

Moreover, stakeholders mentioned several laws and regulations that hindered cross-sectoral collaboration. One example concerned the legal task of youth healthcare to enhance children's health and development (0 – 18 years), which lacks a focus on pregnancy and (future) parents. At the time of data collection, a law was being prepared that gave municipalities the responsibility to implement prenatal home visits by youth healthcare. This expands the scope of youth healthcare and was well-received. Another example was the ambiguity of midwives' role in promoting preconception health, as they usually meet expectant mothers during pregnancy. Several stakeholders called for better preconception care arrangements. Lastly, when other crises were perceived as more immediate (e.g. COVID-19 pandemic for Public Health Services), organizations tend to focus on their core activities written in laws and regulations, which may not always include Solid Start. Hence, stakeholders expressed a need to adapt the scope of laws and regulations to facilitate Solid Start. Additionally, they mentioned that well-defined procedures, roles and responsibilities could help to eliminate a lack of commitment. They suggested for example that an organizational entity should be allocated with the responsibility to serve as the driving force to continue with Solid Start, even if funding by the Ministry of Health, Welfare and Sport would stop.

Stakeholders appreciated the national Solid Start program's design and structure, which features national governmental stewardship and a strong local focus and infrastructure. They acknowledged that the program's emphasis on local considerations was appropriate, given the unique contextual and societal challenges faced by different municipalities. The program provided sufficient autonomy to implement locally without following a rigid, prescriptive checklist. However, stakeholders also sought to ensure the institutionalization and long-term integration of Solid Start and its interventions. Municipalities reported difficulties in moving out of the innovation- and pilot-phase. Stakeholders emphasized, especially in 2021, that they considered Solid Start a 'transition' or 'movement' rather than a short-term project. Although progress was being made, stakeholders recognized

that the program's shift from managerial, policy and executive board levels to individual professionals in daily practice takes time and effort:

> *"And we really still need to take the step towards the individual care provider who should work with it, because they are actually in direct contact with that family. [...] I think that's maybe the most difficult thing, that it doesn't just stay on those governance tables, but that it's now transported to where it really needs to be." - FGD, 2021*

In this process, stakeholders suggested to focus on coalitions and professionals' needs for guidance and support, and to further facilitate knowledge development and dissemination. One of the stakeholders proposed an increase in interactions between national and regional/local level to facilitate these objectives.

Lastly, stakeholders commented that Solid Start should be considered in a wider societal perspective as the ultimate form of prevention to address health disparities and tackle poverty issues. This means that Solid Start should maintain its prioritization. Currently, the system is not entirely in alignment with the overarching mission. The underlying reasons for initiating Solid Start are deeply rooted, complicated and not easily resolved, which was why the stakeholders emphasized that a continuous focus is necessary:

> *"I am incredibly happy with a program like Solid Start, because you can just work with many more people and many more municipalities, and extract the effective elements. [..] But if the Solid Start program only lasts four or five years, we haven't tackled the problem, we've just become more aware, and hopefully we've been able to find each other better and hopefully there are people in many municipalities who want to continue being a driving force, but we haven't solved the problem. And we have to get rid of that illusion [that we solve it in four of five years] somehow." - FGD, 2020*

DISCUSSION

This study aimed to describe the implementation of the Dutch Solid Start program during 2019 - 2021. Questionnaires, interviews and FGDs revealed progress in cross-sectoral collaboration over the years, with a growing number of municipalities forming Solid Start coalitions involving diverse stakeholders. Coalition development varied due to municipalities' unique challenges, focus and historical contexts. According to the stakeholders, initiating the Solid Start program increased the sense of urgency for the first thousand days and stimulated professionals from various backgrounds to get to know each other, resulting in more collaborative agreements on care provision. Stakeholders appreciated the program's local focus and opportunities for learning. However, they experienced that Solid Start is not yet fully incorporated into all professionals' everyday practice. Most common barriers related to systemic integration at macro-level, including limited resources and collaboration-impeding regulations. Stakeholders emphasized the importance of ensuring Solid Start's sustainability.

Our findings suggest that the Solid Start program contributes to the shift from traditional, fragmented care towards a more integrated, population health-based care system as described in literature (39). This approach involves an increased focus on prevention, recognition of the social determinants of health and improving equity in health and wellbeing (39). In line with literature about complex persistent problems, societal transitions, system changes and transformations (40-43), stakeholders mentioned that these developments take time and effort. Historically grown specializations and demarcations that once facilitated progress in healthcare now pose significant integration barriers due to separated cultures, structures and legislations. Nevertheless, it seems that Solid Start has created a nationwide movement to integrate medical and social services for early life within a relative short time (mid-2018 till 2021), with modest funding (€41 million allocated throughout the program's duration) (15, 44) and without a system reform or refiguration. According to Barsties et al.'s transition research in Dutch obstetric care (8), social obstetrics is a new way of thinking, doing and organizing that challenges the incumbent regime that may provide a sustainable addition to the current system, instead of a disruptive transformation. The authors note that social obstetrics can be a starting point for further transformations in obstetrics and other societal systems. Several experts also suggest that systemic structures (e.g. financial structures, laws and regulations) must ultimately transform to achieve greater sustainability and long-term impact than possible through improvements within the current system (43, 45). The trajectory of such transformational processes is often unpredictable and nonlinear (46). Our findings reveal various practical and systemic barriers that impede stakeholder efforts to effect change, calling for systemic transformations as well. The path towards improvements in early life will be influenced by political decisions made in the Netherlands.

In any case, stakeholders emphasized the importance to institutionalize Solid Start and ensure the program's sustainability, to guarantee that the incremental changes result in lasting improvements. Drawing on stakeholders' perspectives and previous literature, several factors can accelerate this transition. The first factor is structural and sustainable funding. Short-term grants should be considered a bridge towards stable financial arrangements for long-term integration and value-creation (45, 47). Meanwhile, sustainable arrangements with municipalities, healthcare insurers, and others should be considered to fund prevention and health promotion, which may require local experiments and legal enforcements. The second factor is adapting the scope of laws and regulations to facilitate Solid Start and cross-sectoral collaboration. The recent changes to the Public Health Act since July 1, 2022, for example, require municipalities to provide prenatal home visits by youth healthcare to parents-to-be in vulnerable situations. Stakeholders have requested additional changes, such as legally outlining preconception care and early detection of vulnerability. If such activities are regarded as core tasks due to laws and regulations, organizations and professionals may be less likely to drop Solid Start activities during crises such as COVID-19 and (expected) labour shortages. The need fits the wider call in the Netherlands to embed public health benchmarks in legislation to increase accountability, similar to environmental legislation (48).

Stakeholders have expressed other needs, which concern responsiveness to stakeholders' needs, ongoing knowledge development, and client-centered care. Firstly, an increased focus to coalitions and professionals' needs is required, as policy recommendations often fail to be implemented in practice without adequate support (49). Further developed partnerships require different types of support compared to those in early stages (45, 47). Additionally, professionals must be supported in adapting to their changing roles and responsibilities in daily practice, as behavioural change is difficult and influenced by multiple factors, including knowledge and skills development (50-52). Secondly, ongoing knowledge development and dissemination are vital to overcome collaborative challenges and stimulate learning. Many systemic barriers cannot be resolved by individual parties at local level and require changes at higher levels. More interaction between local, regional and national levels through intermediary partners, platforms or boundary spanners may help to create learning opportunities at all levels and to adequately collect and respond to different needs (21). An example is the RIVM's local monitoring support program: various coalitions regularly discuss local challenges and successes for mutual learning, and pressing issues are shared with the Ministry of Health, Welfare and Sport to inform the policy agenda. Thirdly, stakeholders emphasized the importance of putting clients' experiences and needs central in daily care and its organization. Although there has been progress, stakeholders felt that this required improvement. Client-centered care and participation (in decision-making) can improve the professional-client relationship, increase satisfaction and promote sustainable innovations by considering clients' preferences, needs, strengths and weaknesses (53, 54).

Our findings are in line with the needs and learning points described in both national and international papers on integrated care and cross-sectoral collaboration in other fields (41, 45, 55-57). For example, these papers also reported on the importance of interpersonal contact and mutual recognition of each other's roles and expertise, engaging all stakeholders (including clients), ensuring sustainable finances, fostering learning cycles, adapting to new roles and skills, and having good governance and leadership throughout all levels of the system. Additionally, we identified comparable obstacles to collaborative efforts as documented within the medical maternity care sector such as fragmented structures and cultures, limited resources and impeding regulations (19-26, 30). Nevertheless, collaborating between sectors seemed to pose additional challenges, given the greater disparities in relational and organizational aspects. For example, the differences between municipal structures and the healthcare system required more investment to foster mutual understanding and familiarity with each other's work environments and interests. Moreover, the financial system was more compartmentalized and governed by distinct regulatory frameworks, presenting challenges in financing preventive measures that are at the intersection of different laws. Lastly, we found that the perceived difficulties from developments within one sector (e.g. integrated birth care, youth care transitions) can complicate cross-sectoral collaboration.

In 2022, the Ministry of Health, Welfare and Sport published the follow-up approach Solid Start 2022-2025 *Strong parents, healthy children!,* which aspires to create a structural

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Solid Start approach in every municipality (58, 59). The approach aligns with the needs expressed in our study. There is a continuous focus on cross-sectoral collaboration at local and regional level, and extra focus to client involvement, facilitating professionals and strengthening informal networks. The approach outlines a commitment to sustainable funding, supportive regulations, governance agreements, a learning infrastructure, monitoring and retain a sense of urgency. Some specific actions have been defined to attain these intentions, while others will be developed. The follow-up approach highlights embedding Solid Start in wider prevention policies and linking it with other policy themes (e.g. poverty) to ensure its sustainability. Given that changes can take decades or span generations (40), during which leadership and contextual circumstances will inevitably change, we need long-term plans beyond the time horizons of a few years to reduce inequities and improve health and well-being (45, 60, 61).

This study offers relevant insights to future policy developments and collaborative practices, and contributes to the knowledge base on cross-sectoral collaboration. Multiple other countries started programs to reduce health inequities by stimulating cross-sectoral collaboration in early life. Examples are the First 1000 days-program in Massachusetts (US) (11), Sure Start in England (12), Strong Start and Healthy Start in the US (13, 62), Strong Start in Australia (14) and Germany's Early Childhood Intervention program (63). Future research should synthesize learning points from successes and failures across these programs and countries. Monitoring processes and outcomes on an ongoing basis can support learning for continuous improvements, consistent with the concepts of reflexivity and reflexive monitoring (49, 64, 65). The importance of monitoring applies to both national and local (municipality) level (66). Future research should also focus on the effects of Solid Start on health outcomes and utilization.

Strengths and limitations
Strengths of this study were the extensive data collection over multiple years and the inclusion of a wide mix of stakeholders, including clients and experts-by-experience. Our data collection seemed to have reached saturation. However, the perspectives of some important parties such as GPs, health insurers and councillors were missed and could have given additional insights. Also, municipalities that did not request Solid Start funds responded less to questionnaires, and we may have involved a selective group of more active and motivated stakeholders in interviews and FGDs. This may have led to more positive findings, although we noticed that our approach provided a good understanding of barriers to implementation at various levels as well. The approach in which we combined FGDs, interviews and questionnaires contributed to the credibility of our results (67). Quantitative data increased our understanding of Solid Start implementation nationwide, and qualitative data provided detailed, contextualized insights.

Using the RMIC as analytical framework for our qualitative data was considered useful to better understand collaboration across professionals, organizations, levels and sectors. The RMIC is one of the theoretical models and definitions on collaboration, integrated care and Population Health Management that sought to outline its important elements (e.g. 31, 56,

68, 69). The model is well able to capture cross-sectoral collaboration. However, as with any other model, the RMIC's reliance on predefined domains and elements may overlook the complexity and variability of integrated care initiatives in practice. Nonetheless, the multilevel and multidimensional RMIC has a strong theoretical and empirical foundation, as it is based on extensive literature review (31, 38) and widely used in research, also in Dutch maternity care (70). For this study, using the model has provided greater insight into the significance of normative integration as a primary step in cross-sectoral collaboration, the dynamics among different layers, and the potential for improvement even in the presence of systemic-level barriers that should be addressed over time. In future endeavours, it may be valuable to explore the underlying cognitive processes influencing the implementation of the Solid Start program, for example as outlined by the normalization process theory (71).

CONCLUSION

This study shows that the Dutch Solid Start program has created a movement towards a more integrated and population health-based care and support system. Solid Start, as a national program with strong local focus, has led to various incremental changes that supported cross-sectoral collaboration for early life, without major transformations of systemic structures. This study highlights several barriers and needs to address in order to ensure the program's sustainability. Those include sustainable funding, supportive regulations, responsiveness to professionals' and coalitions' needs, ongoing knowledge development, and client involvement. In the near future, it is essential to monitor whether the follow-up approach effectively addresses the barriers and needs.

Abbreviations
FGD: Focus Group Discussion
GP: General Practitioner
Ministry of VWS: Dutch Ministry of Health, Welfare and Sport
RIVM: Dutch National Institute for Public Health and the Environment
RMIC: Rainbow Model of Integrated Care

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Authors' contributions

The study was designed by JM, IB and JS, and reviewed by JKdJ. Data collection was done by JM, IB and JS. Quantitative data analysis was done by JM. Qualitative data analysis and interpretation was done by JM and IB, with JS and JKdJ providing feedback. JM authored the article, while the other members of the research team provided feedback and revised the manuscript over several iterative rounds. All authors have read and approved the final version of the manuscript.

Authors' information

The research-team consisted of a health scientist with expertise in collaboration, vulnerability and shared decision-making in maternity care (JM), a former midwife and researcher on integrated maternity care (IB), a researcher with expertise in maternity care and alternative payment models (JS) and a professor in Population Health with emphasis on life course health research (JKdJ).

Availability of data and materials

The datasets generated and analysed during the current study are not publicly available due to the restrictions claimed in the information to respondents and to ensure the protection of anonymity of the participants. Any templates used for data collection and analysis are available from the corresponding author on reasonable request.

Ethics approval and consent to participate

All methods were carried out in accordance with the Declaration of Helsinki. All study participants received an information letter and were informed that participation was voluntary. Additionally, participants were reassured that all information they shared was treated confidentially. The results were reported anonymously, without any personal identifying information that could be traced back to individuals or organizations. Prior to the start of each interview or FGD, all participants provided written or oral (recorded) informed consent. Municipalities received an invitation explaining that the results and publications would not be traceable to individual municipalities, and they provided informed consent by completing the online questionnaire. At the start of the national Solid Start monitor in 2019, we checked by the Medical Research Ethics Committees United whether medical-ethical approval was necessary according to Dutch legislation. They stated and confirmed that the monitor does not fall within the scope of the Medical Research Involving Human Subjects Act (in Dutch: Wet Medisch-Wetenschappelijk Onderzoek, WMO), hence it was exempted for further approval by the ethical research committee (reference number: W19.063). By that time, only FGDs were planned and included in the proposal. Next, we checked whether this exemption also holds for the additional interviews with stakeholders in 2021 at the Clinical Expertise Centre of the RIVM

as part of their legal task. The Clinical Expertise Centre confirmed that this specific study as described in this manuscript, which is part of the national monitor and includes both FGDs and interviews, was also exempted for further approval by the ethical research committee according to Dutch legislation as the study does not fall within the scope of the Medical Research Involving Human Subjects Act (reference number: VPZ-574).

Consent for publication
Not applicable

Competing interests
The authors declare that they have no competing interests.

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APPENDIX 1.
Description of the Dutch care and support system during the first thousand days

In the Netherlands, (future) parents and children generally receive care and support from different service providers, depending on the (expected) health risks and need for support. During pregnancy, women without medical risk factors are generally seen by primary care midwives and they can choose to give birth at home or in an outpatient clinic. In case of increased medical risks or complications, women are referred to general hospitals (secondary care), or, in case of severe morbidity, to academic hospitals (tertiary care) (1). Obstetricians, hospital-based midwives, obstetric nurses, and pediatricians provide care in the hospital. After birth, maternity care assistants provide postnatal care to mother and baby at home or in a maternity hotel. Children receive youth healthcare services by youth doctors, youth nurses and assistants till the age of 18. Youth healthcare services also provide prenatal home visits to pregnant women and families in a vulnerable situation, following a change in the Public Health Act in July 2022 as part of the Solid Start program (2, 3). Furthermore, depending on the (future) parents' circumstances and need for support, they can be referred to service providers or organizations in the social domain or youth care. For example, this could be a municipal housing official for help related to housing, a dept counsellor for support with financial issues, or a social worker or Safe Home (in Dutch: Veilig Thuis, the national report center for domestic violence and child abuse) to intervene in cases of domestic violence (4, 5). Within each municipality, the type of support that is available to parents can differ. Social support services are paid by municipalities under the Social Support Act (in Dutch: Wet Maatschappelijke Ondersteuning, Wmo) and medical services are reimbursed, mostly on a fee-for-service base, through health insurance companies under the Healthcare Insurance Act (in Dutch: Zorgverzekeringswet, Zvw) (4).

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APPENDIX 2.
Description of national and local Solid Start monitor by the National Institute for Public Health and the Environment

The Ministry of Health, Welfare and Sport (Dutch abbreviation: VWS) commissioned the National Institute for Public Health and the Environment (Dutch abbreviation: RIVM) to monitor the implementation of the Solid Start program, launched in September 2018. The RIVM began monitoring the program at the national level in 2019 and launched a support program for municipalities called the "Learning Local Monitor Solid Start" in 2021.

National Solid Start monitor
The national monitor includes both a quantitative and a qualitative component. In a Delphi study conducted in 2019, experts from policy, practice, and research developed a set of 15 quantitative indicators (1). The indicators reflect both processes (e.g. percentage of municipalities with a local Solid Start coalition) and outcomes (e.g. percentage of children born prematurely or with a low birth weight). Annual factsheets (2-4) report the figures for each indicator to monitor the program's progress and developments/trends in health outcomes. The RIVM uses several data sources to quantify the indicators, including:

1. Data from the nationwide population-based data-infrastructure DIAPER (acronym for Data-InfrAstructure for ParEnts and ChildRen). DIAPER integrates routinely collected observational data from three Dutch nationwide data sources (Perined, Vektis and Statistics Netherlands) at individual level. Perined is the national pregnancy, birth and neonatal data registry, based on data supplied by midwives, obstetricians and paediatricians. Vektis offers data on healthcare utilization and spending by collecting claims data under the Dutch Healthcare Insurance Act. Statistics Netherlands collects, disseminates and facilitates access to data on societal themes, including health, welfare, income, education and labour
2. Inquiries to national organizations involved in the implementation of interventions and youth healthcare organizations, as there is no national youth healthcare data registry in the Netherlands
3. Questionnaires among municipalities

The qualitative component involves interviews and focus group discussions with stakeholders that provide further insight into the factors that facilitate or hinder the implementation of the program (4).

The Ministry of Health uses the results of the monitor in combination with other data sources and expert opinions to determine whether goals are being achieved and to timely adjust policies. To underpin the key-messages within the factsheets and to provide a scientific base for our work for Solid Start, in-depth scientific research and analyses are conducted. This manuscript serves as an example.

Learning Local Monitor Solid Start

In 2021, the RIVM started providing support to municipalities in setting up local monitoring, as several of them expressed a need for such support. The support program aims to encourage local coalitions to utilize monitoring as a tool to enhance and refine their local approach. Key elements of the program include the establishment of a learning community that fosters mutual learning among stakeholders (both within and between local coalitions) and encourage the sharing of best practices.
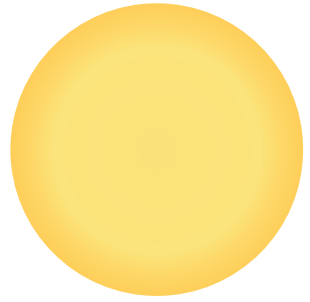
Eleven representatives from local coalitions participate in regular learning sessions. They were already engaged in monitoring Solid Start at the local level before or in the early stages of the national program. This group inspires each other by sharing their experiences and best practices. They also discuss challenges and needs for support in local monitoring. Examples include 'what is vulnerability?' and 'how to monitor the collaboration between the medical and social domain?' These themes are elaborated upon in thematic meetings that are accessible to a broader audience of other municipalities and professionals.

The representatives considered the development of a suitable indicator set the essential first step to stimulate monitoring on a local level. In a previous paper, we have described our approach in developing an indicator set to monitor the Solid Start program in Dutch local coalitions and we presented this indicator set (5). These local indicators are quantified and presented to all municipalities in the Netherlands at www.regiobeeld.nl/kansrijkestart. In the future, this website will be further refined with additional indicators and new functionalities.

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General discussion
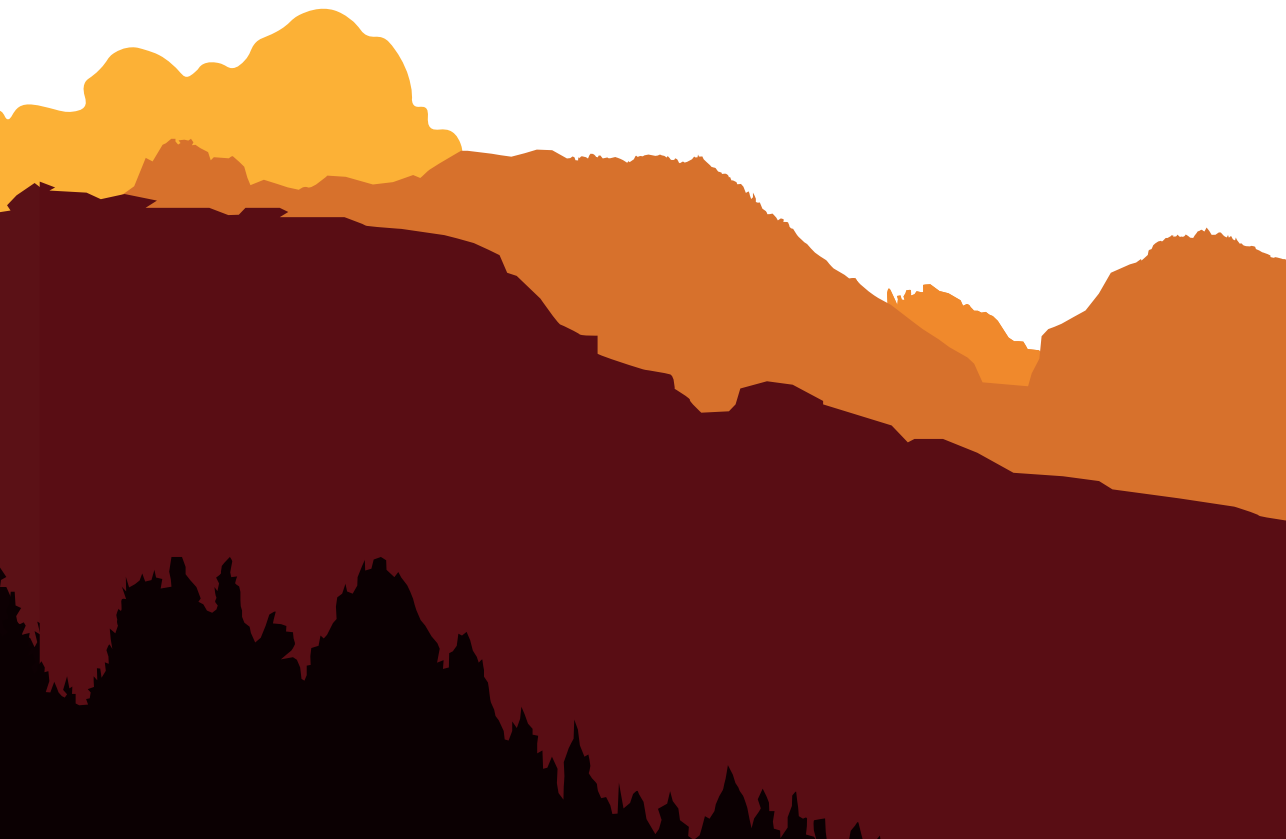
The first thousand days of our life, spanning from conception to our second birthday, lay the foundation for optimal future health and well-being (1-3). During those first thousand days, our development and opportunities are not only driven by medical factors, but strongly depend on the direct and indirect influences of social factors as well (4). Reducing health inequities and providing every child a good start in life therefore requires preventive and integrated initiatives across the social and medical sector. In 2018, the nationwide action program 'Solid Start' was launched by the Ministry of Health, Welfare and Sport (Dutch abbreviation: VWS), aiming for the best possible start for all children during the first thousand days of life (5). The action program Solid Start promotes cross-sectoral collaboration and focuses particularly on (future) parents and young children in vulnerable situations. Starting from 2019, the Ministry of Health, Welfare and Sport commissioned the National Institute for Public Health and the Environment (Dutch abbreviation: RIVM) to monitor the action program Solid Start. This thesis forms the scientific basis for the monitor.

The main objective of this thesis was to provide insight into the adoption of the action program Solid Start, thereby focusing on monitoring and cross-sectoral collaboration. Using a wide range of quantitative and qualitative research methods, the studies included in this thesis offer insights into what and how to monitor, as well as the developments and experiences with the action program Solid Start.

This final chapter begins with a summary of the main findings of the included studies in this thesis, followed by a reflection that outlines and contextualizes key lessons learned using the main findings and recent literature. Subsequently, this chapter highlights methodological considerations along with recommendations for research. This chapter closes with a future outlook, providing recommendations for policy, practice and education, and concluding remarks.

MAIN FINDINGS

What is vulnerability during pregnancy, and how to operationalize vulnerability for monitoring?
We studied the concept of vulnerability and its operationalization for monitoring purposes in Chapter 2 and 3. Both studies included pre-pregnancy data on a wide range of social risk and protective factors, as derived from nationwide routinely collected data sources within DIAPER (acronym for Data-InfrAstructure for ParEnts and childRen) and self-reported data on health, wellbeing and lifestyle from the Public Health Monitor 2016 (PHM-2016).

The study in Chapter 2 aimed to provide more insight into vulnerability by identifying classes (groups) of pregnant women with similar characteristics, and their relation with adverse outcomes to validate classes. A latent class analysis among pregnant women showed five different vulnerability-classes with varying combinations of risk and protective factors to vulnerability: multidimensional vulnerability, socioeconomic vulnerability, psychosocial vulnerability, high care utilization, and the healthy and socioeconomically

stable-class. Women in the multidimensional vulnerability-class shared multiple risk factors in various domains and a lack of protective factors. These women more often had adverse outcomes, including premature birth and caesarean section, as compared to the healthy and socioeconomically stable-class. The three classes with risk factors in one domain and protective factors in others did not. These results show the importance of considering the co-existence of multiple risk factors and protective factors that may act as positive exposures or buffering mechanisms promoting resilience. The results also suggest that early detection of vulnerability and strategies to improve parental health and well-being might benefit from focusing on different domains and combining medical and social care and support, with attention to the systemic causes of vulnerability.

The next study, described in Chapter 3, further explored which data to use to predict multidimensional vulnerability at population-level. Our previous study was conducted in a non-representative subset of pregnant women, meaning that the prevalence of multidimensional vulnerability among all pregnant women in the Netherlands was unknown. It was unclear whether the prevalence could be assessed using routinely collected nationwide data as readily available in DIAPER. Hence, we studied the feasibility of using solely routinely collected data for predictions, the relevance of adding self-reported data, and the most important predictors. The results showed that it is feasible to use solely routinely collected data to predict multidimensional vulnerability. This data is readily available for the entire population and can provide a robust foundation for longitudinal monitoring and policy formulation at population-level. Nevertheless, results also showed that self-reported data was of added value in the predictions. Moreover, self-reported health variables were found to be important predictors to multidimensional vulnerability, next to socioeconomic characteristics and healthcare utilization. Hence, the results offer the opportunity to explore how self-reported health can be systematically included (e.g. in screening and care registries) to enhance the provision of personalized care and support while further improving population-level predictions in the future.

Which indicators can be used to monitor the action program Solid Start on a local level?
Chapter 4 describes the development of an indicator set to monitor the action program Solid Start at local level, using a modified Delphi study with several rounds of questionnaires and online meetings. For local monitoring, experts desired an indicator set covering both processes and outcomes, both parents and children, and both risk and protective factors. The final indicator set comprised nineteen indicators within the three phases of the action program Solid Start: preconception, pregnancy and after birth. Topics included poverty, psychological/psychiatric problems, stress, smoking, vulnerability, preconception care, low literacy and premature birth. The prioritized indicators primarily related to social determinants of health rather than specific clinical aspects. Additionally, a development agenda was set with topics and indicators lacking nationwide data or clear operationalization (e.g. stress, unintended pregnancy, loneliness). We identified both similarities and differences in the selected indicators for monitoring the action program Solid Start at local level compared to national level. These variations can reasonably be attributed to differing purposes and informational needs: monitoring and evaluating

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nationwide implementation versus facilitating the local monitoring and approach. In the local monitoring of the action program Solid Start, the indicator set can enhance the conversation between policymakers, managers, professionals and other stakeholders about the local situation and developments to prioritize local interventions and policies.

What are the developments and experiences with the action program Solid Start and specifically cross-sectoral collaboration?
Chapter 5 describes the implementation of the action program Solid Start during the program's own first thousand days (i.e. 2019, 2020 and 2021), with a specific focus on cross-sectoral collaboration. Generally, the findings from questionnaires, focus group discussions and interviews revealed progress in collaboration at different levels over the years. First, the study reflected on the development of local coalitions Solid Start. We found a growing number of coalitions Solid Start involving diverse stakeholders, and municipalities increasingly reported plans of action, objectives, ambitions and activities for Solid Start. Coalition development varied due to municipalities' unique challenges, focus and historical contexts. Secondly, our results provided insight into the experiences with the action program Solid Start and cross-sectoral collaboration, including facilitators, barriers and needs. Initiating the action program Solid Start increased the sense of urgency for the importance of the first thousand days and stimulated professionals from various backgrounds to get to know each other, resulting in more collaborative agreements and protocols on cross-sectoral care provision. Some general facilitators for effective coalitions Solid Start were an active coordinator as a driving force and a shared societal goal. Moreover, stakeholders appreciated the program's strong local focus and opportunities for learning. However, the action program Solid Start appeared not yet fully incorporated into all professionals' everyday practice. Most common barriers related to systemic integration at macro-level, including limited resources and collaboration-inhibiting regulations. Stakeholders emphasized the importance of continuing with Solid Start and suggested various needs to ensure the program's sustainability. Those needs included sustainable funding, supportive regulations, ongoing knowledge development and learning, responsiveness to stakeholders' needs, and better and more client involvement.

REFLECTION

The action program Solid Start is the first national program to address the full period of the first thousand days while bridging the medical and social sector. The elements of the action program Solid Start were increasingly adopted over the past years, reflecting a shift from traditional, fragmented care towards a more integrated, population health-based care system (6). The approach emphasizes prevention and acknowledges the social determinants of health, which are favoured but still sporadic elements of integrated care models (7). In this thesis, integrated care denotes the collaborative efforts of professionals and organizations across the medical and social sector to provide comprehensive, accessible and coordinated care for the benefit of (future) parents and their children (8, 9).

Considering the above, the developments within the action program Solid Start at national level appear to align with the principles of Population Health Management (PHM). The conceptualization and definition of PHM are subject to ongoing refinement, yet PHM typically refers to efforts aimed at integrating services across the public health, social and medical sector. The overarching goal is to enhance health equity, patient experiences, provider well-being and population health, while reducing costs (known as the quintuple aim) (10-12). Regularly, studies appear that describe various elements of PHM initiatives, offering valuable insights into their design, implementation and evaluation (6, 13-16). These elements include population segmentation, risk-stratification, understanding populations' strengths and needs, and monitoring and evaluating population-tailored strategies (10, 16, 17). In the context of the action program Solid Start and specifically the local coalitions, the extensive literature on Learning Health Systems (LHS) is also relevant (18-21). LHS emphasizes a cycle of continuous learning to improve care, using elements such as data-linkage and sharing. Further exploration of both PHM and LHS literature can provide valuable insight into the interpretation of the findings presented in this thesis.

Drawing upon our research findings and in light of recent scientific literature, we provide multiple lessons learned. These insights may guide future endeavours related to the action program Solid Start, and may also be relevant in the adoption and monitoring of other cross-sectoral initiatives. The lessons learned are structured alongside the components of the main research objective, namely the adoption of the action program Solid Start, monitoring and cross-sectoral collaboration.

Lessons learned in *the adoption* of the action program Solid Start
The adoption of the action program Solid Start encountered both facilitators and barriers and seemed to be a continuous learning process, but it showed overall progress. Chapter 5 outlined numerous program-elements and developments that may have contributed to the adoption of the action program Solid Start. Based on these findings, we draw two lessons learned.

1. The adoption was facilitated by a unifying narrative and dedicated champions at all levels
Having a unifying narrative that not only sets a clear societal goal but also resonates with stakeholders at different levels and sectors was instrumental in facilitating the adoption of the action program Solid Start. This narrative, emphasizing the importance of the first thousand days and the imperative for cross-sectoral collaboration, extends beyond immediate issues to build a foundation for long-term improvements. By creating common ground and instilling a sense of urgency, the narrative prompts a shared commitment to the idea that 'we are in the same boat' for the future, thereby laying a solid base for collaboration. These interrelated factors of a shared vision, commitment and societal urgency align with previous literature on cross-sectoral collaboration (9, 22-26) and are influential in initiating and sustaining integrated programs and partnerships.

The narrative, widely spread, was turned into action through the efforts of dedicated champions at different levels. At the local level, proactive coordinators or facilitators acted as driving forces for coalitions' progress. At regional and national level, these individuals were described as ambassadors, advocates or visionary leaders who consistently keep Solid Start on the policy agenda, and who inspire others. In the adoption of innovations or transformations, such strong and committed 'champions' or 'change agents' are frequently described as key factors to initiate, promote and sustain a certain movement or collaborative initiative (19, 20, 25, 27-30). In addition to designated champions, it is essential that informal champions emerge from intrinsically motivated frontline professionals in order to stimulate the engagement of peers (29). Moreover, collaboration among multiple champions, especially those in different positions (e.g. care provider and project coordinator), is described as beneficial (27). This may contribute to boundary-spanning across levels and services, and possibly help to address the gaps that currently arise when a champion leaves. Continuously emphasizing the narrative and providing support to champions across all levels may contribute to the long-term integration of the action program Solid Start into everyday practice of all professionals.

2. National governmental stewardship with strong local focus is a promising combination
The combination of stable national governmental stewardship with a strong local focus increased the action program's adoption, as it provided a clear direction and support while it simultaneously ensured alignment with local contexts, practices and networks. Previous evidence also indicates that implementing and sustaining integrated care involves balancing two approaches: a top-down and bottom-up approach (7, 31-33). Currently, literature describes missed opportunities due to an over-reliance on top-down approaches in integrated care (7).

A more traditional top-down approach can create favourable conditions (regulations, finances, governance) and external motivation for change (7, 31). In our study, a consistent 'push' and structured program with supportive mechanisms from the national government (referred to as national governmental stewardship) prompted a sense of urgency, guided local policy agendas and steered local action. These supportive mechanisms extended beyond mere financial support, including practical support in setting up a coalition Solid Start and implementing interventions. Simultaneously, our study also identified various barriers to integration at systemic level (e.g. limited resources, collaboration-inhibiting laws and regulations), and highlighted the need for more responsiveness to local stakeholders' needs, meaning there is still potential for improvement.

Next, a bottom-up approach fosters engagement and support from professionals who are directly involved in changes, allowing for future-proof innovations that align with local needs (31). In this thesis, encouraging and facilitating municipalities to create their own approach increased flexibility to respond to unique local situations and developments. The synthesis of Wodchis and colleagues (33) on integrated elderly care explains how bottom-up initiatives require top-down support to sustain and spread, and Behrendt and Ramanuj (32) convey that learning processes are part of the synergistic interaction

between the two approaches. Thus, a combined approach, where the top-down approach stimulates the bottom-up approach and vice versa, in an emergent way, is beneficial to implement integrated care. In our study, this combined approach may have promoted the continuation or strengthening of initiatives that existed since before the nationwide action program Solid Start, rather than resulting in stagnation or cessation. Applying a local focus avoids the pitfall of a one-size-fits-all strategy and allows for variation, local leadership, and continuous learning within an enabling policy environment (32).

Lessons learned in *monitoring*
In this thesis, we share our findings on monitoring the action program Solid Start at both national and local level. Our research has provided more insights into the operationalization of vulnerability (Chapter 2 and 3) and increased our understanding of useful indicators and data to monitor the action program Solid Start (Chapter 4). We share two lessons learned in monitoring based on these findings.

3. Considering both risk and protective factors is important for a comprehensive perspective
The findings from several of our studies highlighted the significance of considering both risk and protective factors to vulnerability. In Chapter 2, we found that a unidimensional perspective to vulnerability, being focused on (single) risk factors in one domain, may be insufficient to correctly predict adverse outcomes during pregnancy and childbirth. The importance of considering protective factors as well was further supported in Chapter 4, in which we learned that a local indicator set to monitor the action program Solid Start should cover both risk and protective factors.

The interrelatedness between risk and protective factors and viewing health from a broader perspective has become more common. This trend is also evident in other fields like elderly care (34), GP-care and hospital-care, and accompanied by the emergence of more comprehensive concepts and methods (35) in the Netherlands and abroad, such as Positive Health (36) and Salutogenesis (37, 38). These concepts emphasize people's strengths, opportunities and positive experiences, rather than focusing (merely) on weaknesses or risks. Additionally, they adopt a more holistic perspective in health and well-being as they consider physical, social and emotional aspects. This broad perspective encourages cross-sector collaborations and preventive strategies by acknowledging the relevance of multiple sectors to gain insight and create solutions.

Academic literature in early life is also shifting its focus from adversity and risks towards emphasizing the importance of resilience and protective factors (39, 40). Traditionally, studies have primarily concentrated on risk factors in predicting adverse early life outcomes. There is still much to learn about the co-occurrence and interplay of risk and protective factors to improve the health and wellbeing of future generations. Those protective factors are not merely the absence of risks, but additional elements that increase well-being or guard against unfavourable outcomes (41). Hence, research into resilience and protective factors is emerging, with social support being most frequently studied and best supported in the social sciences (39, 40, 42, 43). Additionally, an increasing

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number of studies seem to consider the combination of various risks across domains, rather than focusing on single risk factors, as also exemplified in a recent latent class study of Helmikstøl et al. (44). From a preventive and solution-focused perspective, it is important to consider factors that are modifiable, rather than concentrating exclusively on immutable factors for both risk and protective factors. This was also deemed important for monitoring efforts, as stressed in Chapter 4.

4. Monitoring requires longitudinal cross-sectoral data and indicators

Chapter 2 and 3 showed that, in order to operationalize vulnerability among pregnant women, comprehensive data on a wide range of factors in different domains (e.g. socioeconomic, psychosocial and medical risk and protective factors) are necessary. Data within one sector alone cannot capture all relevant elements. Moreover, the indicators that were chosen to monitor the action program Solid Start (Chapter 4) reflected both social and medical aspects, and were not exclusively tied to a single profession or sector. Consequently, the findings in this thesis highlight the need for cross-sectoral data and indicators for longitudinal monitoring. This need aligns with previous literature (e.g. 18, 45). For example, several studies describe the potential of integrating data from various sectors to enhance a data-driven approach and internal-monitoring for population health and increased equity (10, 46), although the majority of PHM-initiatives rely on routine care data from one single sector or organization. In a wider perspective, incorporating data on the social determinants of health in daily care workflows is endorsed to support action (47, 48). Lastly, the linkage, storage and sharing of different data-sources (i.e. next to routine care data also data on the SDOH or patient-reported data) are frequently cited elements of LHS (18-20). In this thesis, we utilized DIAPER, which links individual level routinely collected data from the medical and social sector for parents and children on a national scale. Other examples of linked data infrastructures in the Netherlands include the regional Extramural LUMC Academic Network (ELAN), which supports the Healthy and Happy The Hague movement (46).

Lessons learned in *cross-sectoral collaboration*

Scientific literature emphasizes that cross-sectoral collaboration between the medical and social sector is needed to provide children the best possible start in life (4, 49-51). This statement finds support in the findings described in this thesis, as these point towards the cross-sectoral nature of a solid start and the need for cross-sectoral data and indicators for monitoring (Chapter 2, 3 and 4). Chapter 5 described the developments and experiences with the action program Solid Start and specifically cross-sectoral collaboration. Below, we present two lessons learned in cross-sectoral collaboration based on our findings.

5. Fostering normative integration is a fundamental first step to collaborate

We learned that normative integration was a fundamental step to increase cross-sectoral collaboration. Normative integration includes the 'softer' aspects of integration, such as creating a shared vision, culture, trust, and mutual acquaintanceship (9, 23, 52). Our findings showed for example that the increased sense of urgency coupled with knowing each other provided a solid basis to initiate or intensify activities within the coalitions Solid

Start. Studies in other fields make similar observations that interpersonal dynamics and creating a common frame of reference are essential (13, 22, 30, 34, 53-55). In the context of early life, a recent Danish study into cross-sectoral collaboration for pregnant women in vulnerable situations stressed that knowing each other's working context is helpful (56), and a study into a Canadian Child Health Network mentioned relationships as 'system triggers' that prompt change to professionals' everyday practice. The systematic review of Such et al. (30) used systems thinking to explore the dynamics between relational and structural governance components in successful collaboration across sectors. Their causal loop diagram showed positive feedback loops between relational aspects, and also high interrelatedness between structural and relational components. Both contribute to the credibility and legitimacy of collaborations.

The above implies that continuous efforts should be made to stimulate encounters between professionals from diverse backgrounds, who can then build a collaborative culture and make plans together. While it may be time-consuming and requires acknowledgement of contexts (e.g. historical, political and sociocultural conditions), this seems to be a necessary part of integration. This may be specifically true for collaboration between multiple sectors in contrast to collaboration within one sector, given the larger differences in relational and organizational aspects that require additional investments to foster mutual understanding. Moreover, our findings suggest that positive interpersonal dynamics may facilitate improvements even in the presence of systemic barriers. At the same time, these relational elements are essential to start learning together, which in turn can help to overcome collaborative challenges at different levels (including systemic barriers).

6. Processes of learning are indispensable in cross-sectoral collaboration
A final lesson learned from reviewing this thesis' findings and other scientific and grey literature is that 'learning' and its associated elements of reflection and knowledge sharing should be central in the adoption and monitoring of cross-sectoral approaches (18, 30, 53, 57, 58). Developing coalitions Solid Start, or initiating and sustaining cross-sectoral collaboration in a wider perspective, are novel and non-linear processes that unfold in a rapidly evolving field with changing contexts. In these processes, learning is relevant at different levels and scales, between different stakeholders and for different short and long-term purposes (59, 60). Examples in this thesis include learning from (and with) other professionals and experts-by-experience, within and between local coalitions Solid Start, across local and regional levels, and from other integrated care programs and sectors (Chapter 5). We reported that stakeholders use and want to use learning opportunities to improve, share knowledge, prevent duplication of efforts, and overcome collaborative challenges together. Moreover, our Delphi study (Chapter 4) began with stakeholders expressing an interest to learn from other coalitions about monitoring at local level, and their need for local indicators and data to facilitate discussions about local developments in order to learn for future practice and policy.

A recent mapping review into LHS of De Bruin and colleagues (18) described three processes of learning. The most often reported learning processes were information

sharing between clinical practice and research, and ongoing cyclical improvement processes (from performance to data, data to knowledge, knowledge to performance, and so forth) (18). This second, more 'rapid' process of learning requires (recent) data and a data infrastructure as key elements (18, 20, 61). PHM-literature refers to continuous testing, quality improvement processes and learning cycles, using data-driven insights (10, 16, 19, 62). Fewer papers discuss the third process of learning: recurrent interaction between stakeholders for collaborative learning (18). This facilitates the sharing of best-practices, evaluating processes, identifying opportunities for improvement, setting goals and discussing underlying values (18). Less is known about this form of learning in cross-sectoral collaboration, despite its crucial role in "constantly adapting strategies to changing circumstances and unanticipated situations"(57) (p. 1). Possibly, collaborative learning could also facilitate the other learning processes (i.e. research and cyclical improvement). It also often occurs together with one or two of the others (18). In this thesis, we did not dive into these learning processes in detail, but previous studies described that learning and reflection to support a transformation process was time-consuming and requires certain conditions and competencies (e.g. suitable data and indicators, openness, self-reflection, leadership- and teamwork skills, expertise, regular reflection moments and a supportive culture/climate) (20, 63).

We believe that all three types of learning are relevant in the context of Solid Start. Cross-sectoral collaboration requires an adaptive strategy in order to manage upcoming challenges and changing contexts, while simultaneously work towards the goals and aims that were set. Although learning has been used to some extent (e.g. based on DIAPER), there is untapped potential, for example regarding up-to-data data and opportunities for collective learning. Therefore, it is important to think about how to facilitate learning in a more structured way, together with all relevant stakeholders from policy, research and practice, including experts-by-experience.

METHODOLOGICAL CONSIDERATIONS AND RECOMMENDATIONS FOR FUTURE RESEARCH

Several methodological considerations should be kept in mind when interpreting the results from this thesis. Most were discussed in the separate chapters. The following three sections provide overarching methodological considerations, along with recommendations for future research.

A broadened scope in monitoring

The monitoring efforts described in this thesis illustrate a growing link between research, policy and practice. In the monitoring of the action program Solid Start conducted by the National Institute for Public Health and the Environment, we have broadened our traditional research role, building upon the foundations laid by previous efforts. Conventionally, the National Institute for Public Health and the Environment offers a cyclical annual update

on quantitative indicators and stakeholder experiences, enabling the Ministry to provide a rationale for her activities and shape future policy developments (e.g. as input for a follow-up approach). Our evolving role implies the adoption of more flexible and collaborative approaches oriented at learning for policy and practice, and involves using a wide variety of qualitative and quantitative methods, and engaging more experts-by-experience. This was most evident in the setup of the learning local monitor Solid Start, which comprised small scale-learning sessions, larger-scale theme sessions and the development of a local indicator set. Responding to participant's needs, we consequently developed a dashboard displaying municipality-level data for these local indicators, available at www.regiobeeld. nl/kansrijkestart, to stimulate local conversations and decision-making. This tool is still in development, with requests to expand its capabilities (e.g. adding neighbourhood- and regional data) and add indicators from the developmental agenda (e.g. stress, unintended pregnancy, loneliness). We believe that involving the experts that will use the indicators from the start of development was beneficial to increase acceptance and utilization, just like previous research described how involving care providers is crucial for transformational change (64). Additionally, our research on the operationalization of vulnerability was notably driven by local and national demands for a better understanding on the prevalence, geographical distributions and trends in vulnerability. As a result of the broadened monitoring scope, we have produced a diverse array of products relevant for policy (factsheets), research (scientific papers) and practice (indicator set, websites, meetings).

The shift away from conducting research in isolation, particularly in the context of complex and cross-sectoral programs, is increasingly apparent in other monitors and projects as well. Examples include the reflexive evaluation of the program 'Right Care at the Right Place' (65, 66) and the monitor of the 'Healthy and Active Living Agreement' (67). Moreover, there is growing adoption of participatory research designs, characterized by collaborations between researchers and local stakeholders to create and apply relevant knowledge for societal issues in practice, to evaluate and facilitate integration across the medical and social sector (e.g. 68). Engaging with knowledge users throughout all research phases can significantly enhance the uptake of research in practice or policy, facilitating a transition from scientist-driven to problem-based research (69-71). Beckett and colleagues (72) call to embrace the inherent complexity and uncertainty of 'research co-production' and not just focus on the end-goal of changing practice, emphasizing that more nuanced effects on knowledge sharing, relationships and research capacity building can be expected.

Hence, we recommend applying more participatory and co-productive forms of research to advance the Solid Start movement. By doing so, we can address questions that align with the needs and priorities of stakeholders in practice, enhance collaborative learning and better support the processes towards integration. In this regard, the National Institute for Public Health and the Environment can further evolve to a learning organization, actively engaging in iterative processes with stakeholders. Embracing participatory and co-productive research approaches also necessitates different competencies (20) and more flexibility. It implies that the authority for decision-making, including goal-setting and methodologies, is no longer exclusively entrusted to those in the academic world (as

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an ivory tower), but is instead increasingly driven by those who are directly affected by research, policy or practice.

Therefore, an important aspect in future research and monitoring efforts is to truly involve experts-by-experience and individuals with experiential knowledge in all research phases. While this thesis partially achieved this inclusion in certain aspects, we were limited in others. For instance, an expert-by-experience was involved in organizing and conducting interviews with clients to gain insights into experiences with the action program Solid Start (Chapter 5). However, their perspective was missing in the development of an indicator set (Chapter 4). Experts-by-experience and (future) parents could have suggested and prioritized alternative indicators. Genuine involvement of experts-by-experience and individuals with experiential knowledge could be characterized by elements such as sufficient time and resources, shared responsibility, active listening, respect and a motivation to meaningful engagement. As articulated by Goedhart and colleagues (73), engaging citizens in vulnerable positions in research can involve several strategies, tools and methods that should be context-based and require a supportive cultural shift. Their paper provides a welcome overview of ways to address common concerns to engage citizens in vulnerable positions (e.g. moving beyond the 'usual suspects' with time and budget constraints, navigating predefined research questions, managing power dynamics and addressing diverse priorities).

Experienced progress versus measurable effects
This thesis provided insights in the developments and experiences with the adoption of the action program Solid Start and cross-sectoral collaboration. This thesis did not focus on the effects of the action program Solid Start on aims related to health, wellbeing and equity for parents and children. Stakeholders aspire more insights into early effects to inform ongoing monitoring efforts and to maintain support for the action program Solid Start. Especially policy makers may be seeking measurable outcomes to assess the tangible impact of the program. To address this need, we propose further research into the program's early effects, employing methods such as Difference-in-Differences (DiD). DiD is a quasi-experimental study design in which the relation between policy changes and outcomes can be compared over time between participating and non-participating groups (in our case: municipalities) (74). DiD gained popularity with the increase of longitudinal data and has been successfully applied to assess policy interventions during the first thousand days in other countries (e.g. 75, 76). In a future DiD study, we can build upon the findings of this thesis by utilizing the operationalization of vulnerability to compare municipalities and study equity, and by including the selected indicators as outcome measures. DIAPER presents itself as a suitable data infrastructure for this purpose.

Blindly staring at the measurable effects of the collaborative efforts however, fails to fully grasp the complexity of cross-sectoral collaboration. Despite widespread enthusiasm for cross-sectoral collaboration, there is currently little empirical evidence to suggest that cross-sectoral collaboration in itself is sufficient to improve health outcomes and health equity (30, 77, 78). Several potential reasons for this lack of evidence have been proposed.

The reasons range from overestimating the effects of collaboration in the first place, to difficulties in measuring the effects, especially when the effects are diverse, long-term and influenced by other factors. In the case of the action program Solid Start, an absence of immediate improvements in health outcomes would not necessarily imply a lack of impact, and conversely, any potential improvement in outcomes cannot unequivocally be attributed to the program. Alderwick and colleagues (77) suggest that while collaboration may not directly improve health, it may facilitate other developments and contribute to improvements as part of broader strategies to improve health. Moreover, especially programs focusing on early life and intergenerational aspects may not yield immediate measurable effects on health outcomes or equity. Nonetheless, investing in preventive programs for a good start in life, with the potential for positive impact across generations, remains justifiable even without conclusive evidence of immediate effects.

Consequently, the scientific literature supports our understanding that it is valuable to monitor long-term through varied methods, and, rather than concentrating on outcomes, consider the context and processes in collaboration as well (30, 77). This entails a deeper exploration of the connection between these processes and outcomes within certain contexts, aiming to illuminate the causal pathways that contribute to successful collaboration. Starting from this objective, the previously mentioned systematic review of Such et al. (30) adopted a realist-informed perspective to outline the components and dynamics of collaboration in a causal loop diagram. Based on these insights and our finding that context matters in coalition development for Solid Start, we recommend applying a realist approach in monitoring the action program Solid Start for more insights into what works, for whom, in which context and for which outcomes (53, 79). Such an approach further stimulates learning and facilitates adaptations in daily practice and policy. Moreover, a realist approach also holds promise for studying specific interventions for (future) parents in greater depth (80, 81). It is well-documented that preventive interventions designed to improve overall health inadvertently can widen existing inequities in the population, as individuals in more vulnerable situations participate, respond and benefit less (82, 83). Recent Dutch studies also found that the implementation of early life interventions for parents in vulnerable populations are influenced by many factors (84, 85). Taking a realist approach in studying interventions may further help to identify potential improvements of interventions in a given context, particularly for the benefit of individuals in vulnerable situations. In this line of thought, it may also be beneficial to focus on coalitions that achieve better outcomes despite facing comparable challenges, and to seek understanding of what is working well and why, similar to a positive deviance approach (86).

For the longitudinal monitoring of the action program Solid Start, we also propose that stakeholders from policy, practice, research and experts-by-experience together deepen their understanding on the objectives and theories of change of the program. This can be achieved by addressing questions such as: what does 'a solid start for every child' entail? What matters to whom? How is success defined for the action program Solid Start? What short-term developments and proxy measures are anticipated? Which indicators

and monitoring strategies are suitable for subsequent stages in the action program's implementation, with a focus on ensuring sustainability?

Challenges and opportunities with routinely collected data

Several papers in this thesis used DIAPER, a unique data infrastructure that links routinely collected data from several Dutch data sources. Covering the life course from preconception to adulthood, DIAPER provides insights to policy makers, payers and providers in several early life projects (87). Routinely collected data provide the opportunity to study real world situations, leading to results that have strong external validity without additional costs and time spent in collecting data. However, it is essential to consider potential challenges or risks as detailed in Scheefhals'(87) and Ardesch'(46) paper, which related to the quality of the data and its linkages, privacy concerns, missing data and administrative delay. Regarding data quality, it is important to acknowledge that, since data is primarily collected for care purposes and only subsequently used for research, some data may be incomplete or inaccurate. In the linkage of data, there is a risk of errors introducing bias that may disproportionately affect disadvantaged groups that are underrepresented in the data. Privacy concerns that may arise because of increased (technical) possibilities require constant attention and advanced methods for privacy protection. Concerning missing data, the absence of nationwide youth healthcare data (88) is of notable concern. It poses challenges to study children's health, development and underlying determinants, as well as the program's ultimate impact. Additionally, self-reported (experience) measures of parents are lacking. Administrative delays further impede research efforts, as data often becomes available only after undergoing several integration steps and quality checks, limiting its utility in rapid processes of learning and decision-making. Despite these challenges, DIAPER appeared a valuable source to gain insight into vulnerability and to present the data to indicators at both national and local levels.

In order to optimize the utilization of DIAPER for (flexible) practice- and policy-oriented research related to the action program Solid Start and other early life initiatives, three recommendations are proposed. Firstly, the inclusion of youth healthcare data and self-reported (experience) measures, and openness to the possibility of adding additional data depending on the research topic. This could be data from GP practices, schools or specific population-based birth cohorts. Secondly, a proactive approach to identify knowledge gaps relevant to everyday practice and policy, and a flexible allocation of our research time to answer those more ad-hoc questions. Thirdly, an exploration into the feasibility of developing a DIAPER 2.0 version that presents real-time data (e.g. on a monthly basis) from various sectors to support short-term decision-making in daily practice and policy. Throughout these endeavours, it is essential to address stakeholders' potential concerns and communicate the shared benefits to facilitate a supported approach. This also entails that we increase our efforts towards 'open science' and adherence to the FAIR guiding principles ensuring the Findability, Accessibility, Interoperability and Reuse of data (89). For example, we can improve by pre-registering our research methodologies and analysis plan, sharing our scripts (via platforms like GitHub), and uploading preprints of scientific papers.

FUTURE OUTLOOK: RECOMMENDATIONS FOR PRACTICE, POLICY AND EDUCATION

There was a continuous focus on Solid Start in Dutch policy and practice during the time that this thesis was written, marked by new policy developments that further encouraged prevention and integration. In 2022, an Integrated Care Agreement (Dutch abbreviation: IZA) was signed by a wide range of representatives from the health and social care sector. Solid Start was mentioned as one of the approaches to ensure the future quality, affordability and accessibility of healthcare. Moreover, Solid Start was prominently featured in the healthy and active living agreement (Dutch abbreviation: GALA), published at the start of 2023, to contribute to a healthy generation in 2040 (90). These agreements voiced the ambition to integrate a Solid Start approach in every Dutch municipality. This has been translated into structural funding for Solid Start, enabling municipalities to request specific allocation (Dutch abbreviation: SPUK) funds to 1) initiate, strengthen and secure their coalition, 2) enhance the implementation of Solid Start interventions, and 3) establish regional collaborative agreements. In 2022, the Ministry of Health, Welfare and Sport published the follow-up approach *Solid Start 2022–2025 Strong parents, healthy children!* that outlines the mission, vision, strategy and actions to provide all children the best start in life. Additionally, there has been an increase in policy developments and research agendas that focus on prevention and integration through cross-sectoral collaboration for other populations. Essentially, the action program Solid Start must be seen as part of this wider movement.

In light of these developments, this thesis provides several points of discussion that can inform the optimization of the action program Solid Start and potentially similar initiatives. Multiple implications are addressed in the separate chapters. The paragraphs below describe our overall recommendations for practice, policy and education, based on our lessons learned and methodological considerations.

Create a long-term perspective and maintain sense of urgency by positioning Solid Start as the ultimate form of prevention
Integrating the structures, cultures and practices of all involved in the first thousand days is no easy task, but the movement that has been initiated is important to sustain over the long-term to make lasting improvements. Considering that fragmentation has accumulated over time, it is reasonable and well-described that integration or transformation extends over a period of multiple years or decades as well (91). Therefore, we recommend positioning Solid Start even more prominently as an ultimate form of prevention and means to improve health, well-being and equity across the lifespan. In this regard, we believe that the IZA, GALA and follow-up approach help to spread the 'first thousand days-narrative' and maintain a sense of urgency for the action program Solid Start and cross-sectoral collaboration in early life. Next, the IZA, GALA and follow-up approach can provide the necessary sustained governmental stewardship (including structural funding) to build capacity and sustainability in local actions. It still remains important to allow local

and regional coalitions Solid Start the flexibility to develop approaches tailored to their specific context, and to remain responsive to their evolving needs.

Additionally, we suggest closer involvement of other ministries and a more prominent inclusion of the perspective of future generations. Improving health, wellbeing and equity starting in early life entails responsibilities that extend beyond the Ministry of Health, Welfare and Sport to other ministries (i.e. Interior and Kingdom Relations, Social Affairs and Employment, Economic Affairs and Climate Policy, and Education, Culture and Science) (92). These ministries have a role to prevent and solve the larger societal causes of vulnerability, and their policies can influence protective factors and resilience. Enhanced collaboration between different policy sectors can strengthen their separate efforts and contribute to aims both within and outside the health sector (from Health *in* All Policies to Health *for* All Policies) (93, 94). Similarly, the Dutch Council of Public Health & Society (Dutch abbreviation: RVS) stresses the need to safeguard children's rights and interests across all policies and legislations. Hence, aligning with the principles of 'it takes a village to raise a child', we advise to include the perspective of future generations in all policy making.

Integrate the action program Solid Start into everyday practice: facilitate champions and foster 'normative integration'
Following the aforementioned points, we recommend prioritizing the integration of the action program Solid Start into the daily practice of all professionals. More specifically, we suggest providing both financial and practical support to champions within local coalitions Solid Start, regional structures and individuals who serve as advocates at the national level. Additionally, we propose to foster normative integration (i.e. knowing each other, developing a shared vision and culture) through interactions among professionals with diverse backgrounds. This can be achieved through regular in-person coalition meetings or multidisciplinary team gatherings. Additionally, activities such as shadowing peers in their daily practice or organizing work visits can offer valuable insights and promote mutual understanding by providing a behind-the-scenes view. These practices are already done in various locations. Lastly, we recommend identifying coalitions and professionals' needs to integrate the action program Solid Start into everyday practice on an ongoing basis, potentially through a learning infrastructure as elaborated in the next paragraph.

Stimulate and integrate learning processes at different levels
Drawing from the lesson that learning is pivotal, we would recommend to stimulate and accelerate learning processes in different ways and at different levels. Professionals in daily care and support should receive sufficient support and time (working hours) to prioritize collaborative learning activities and cyclical improvement processes. Additionally, a learning infrastructure across local and regional coalitions Solid Start is recommended and could be facilitated by national levels. This includes the provision of resources, practical tools and guidance in how to use learning for reflection and improvement within and across coalitions Solid Start. Next, it is advised to expand a learning infrastructure between local, regional and national levels, and across practice, policy, research and experts-by-experience. These boundary spanning learning processes may help to adequately respond

to the different needs and barriers which cannot be solved separately. In the design and facilitation of such learning infrastructures, we can draw insights from both international as well as national examples, such as the learning network designed within the ZonMw program for unintended pregnancy and vulnerable parenthood (Dutch abbreviation: KOOZ) (95). This national network brings together multiple research projects and collaborative (learning) networks across the country.

Related to learning are education and training. Ideally, young professionals are educated in the importance of cross-sectoral collaboration and the first thousand days right from the start of their career. In this context, interprofessional training is optimal to gain insight into each other's value and start practicing the necessary competencies for collaboration at an early stage.

Broaden the scope beyond the first thousand days
In efforts to broaden the scope of the action program Solid Start, certain municipalities are working towards a 'first 100 + 1000 days-approach', or a 'first 2000 days' approach. The former more explicitly includes the preconception period. A recent study into preconception care showed that there is potential to increase the awareness and uptake of preconception care (96), and several experts call for normalization of the question 'do you want to become pregnant in the coming year?'. It is recommended to place greater emphasis on preconception care and the promotion of a healthy pregnancy, and to make it a more explicit component of the approach. The latter 'first 2000 days' approach continues to age five, and thus extends till the school period. In scientific literature, these 'next thousand days' are described as a critical period to reinform and establish healthy development, including executive and cognitive functions, social-emotional interactions, language and literacy, and self-regulation (97). This period marks a transition from a predominantly home-based environment to increased exposure to the outside environment, as children prepare for and enter formal schooling. Recognizing that development trajectories extend beyond the age of two and acknowledging the importance of coherent resource allocation and alignment, it may be relevant to extend the program's focus to the first 2000 days to ensure continuity between early life and school.

6

CONCLUDING REMARKS

This thesis provided insight into the adoption of the Dutch nationwide action program Solid Start, with a specific focus to monitoring and cross-sectoral collaboration. Throughout our studies, we learned that monitoring vulnerability in early life and monitoring the action program Solid Start at local level requires a consideration of both risk and protective factors spanning across multiple sectors. This underscores the relevancy for preventive programs that connect the medical and social sector, and it implies that monitoring requires cross-sectoral longitudinal data and indicators.

Throughout the years, we found various incremental changes that supported collaboration across the medical and social sector to improve care and support during the first thousand days. Especially getting to know each other and processes of learning seemed to be indispensable in these processes towards cross-sectoral collaboration. In the wider adoption of the action program Solid Start, a unifying narrative, dedicated champions and a strong local focus appeared important facilitators. However, several challenges remain, and it is vital to learn from those to protect the health and well-being of current and future generations. Therefore, for future practice and policy, we advise to create a long-term perspective by positioning Solid Start as the ultimate form of prevention, and integrate the action program Solid Start into everyday practice, navigating in tandem with the IZA and GALA. Additionally, we recommend to stimulate learning processes within and across local, regional and national levels. These recommendations could help to further optimize the action program Solid Start, and contribute to the wider movement towards prevention and integration. Overall, this thesis implies a solid start for the Dutch first thousand days-approach.

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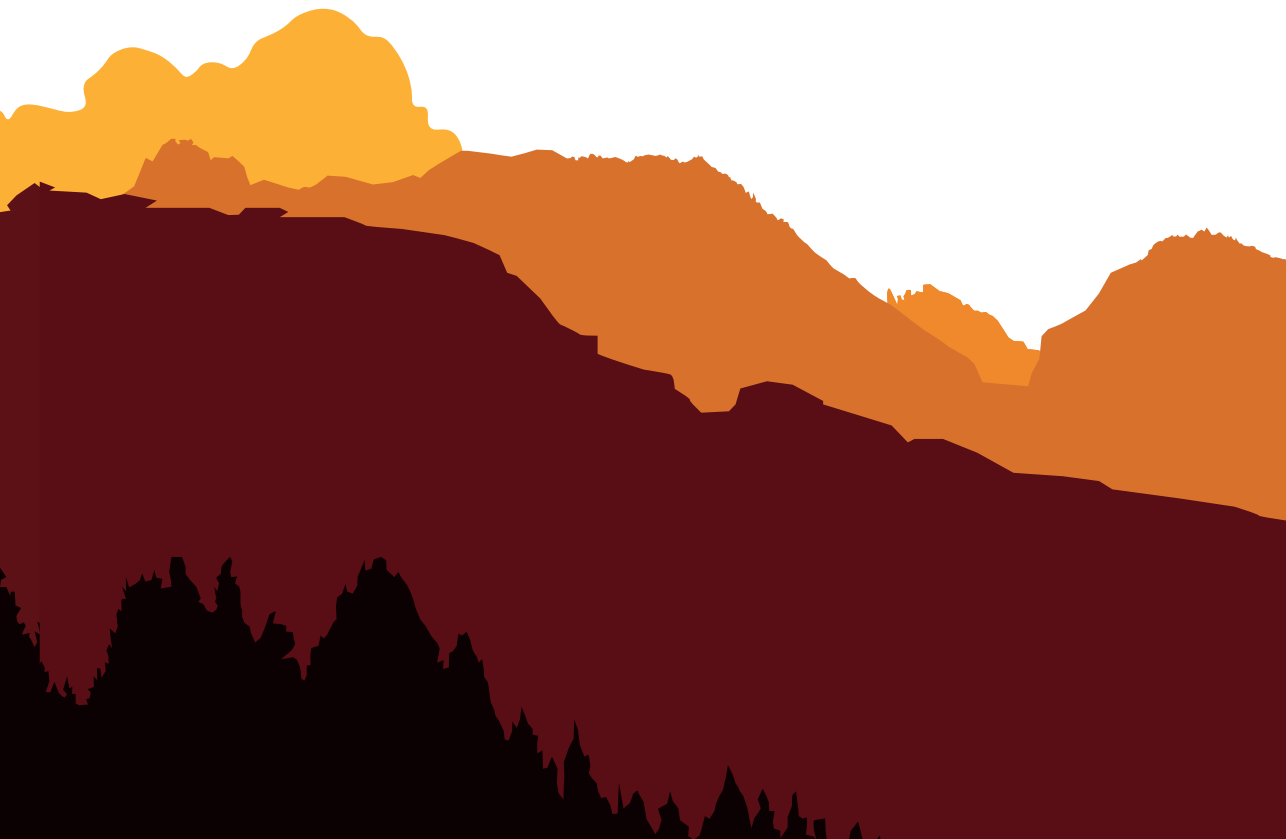
SUMMARY

In 2018, the nationwide action program Solid Start was launched by the Dutch Ministry of Health, Welfare and Sport, aiming to ensure the best possible start for all children during the first thousand days of life. The 'first thousand days' refer to the period from conception to a child's second birthday, which lays the foundation for health and wellbeing in later life and across generations. Children's development and opportunities during this period are influenced by medical factors, but also strongly depend on social risk and protective factors. Addressing health inequities and vulnerability in early life therefore requires integrated and preventive approaches which prioritize collaboration across the medical and social sector. The action program Solid Start promotes cross-sectoral collaboration and focuses particularly on (future) parents and children in vulnerable situations. Since 2019, the National Institute for Public Health and the Environment has been commissioned to monitor the action program Solid Start. This thesis forms the scientific basis of the monitor.

In **Chapter 1**, the rationale, context and aims of this thesis are described in more detail. The main objective of this thesis was to provide insight into the adoption of the Dutch nationwide action program Solid Start, thereby focusing on monitoring and cross-sectoral collaboration. Three research questions guided our research: 1) What is vulnerability during pregnancy, and how to operationalize vulnerability for monitoring? 2) Which indicators can be used to monitor Solid Start on a local level? 3) What are the developments and experiences with Solid Start, specifically regarding cross-sectoral collaboration?

Chapter 2 and 3 focused on monitoring vulnerability during pregnancy. **Chapter 2** provided more insight into vulnerability by identifying classes of pregnant women with similar risk and protective factors, and studying the relation with adverse outcomes. Data were derived from routinely collected data sources in DIAPER (acronym for Data-InfrAstructure for ParEnts and childRen) and self-reported data of the Public Health Monitor. Results showed five classes: multidimensional vulnerability, socioeconomic vulnerability, psychosocial vulnerability, high care utilization, and the healthy and socioeconomically stable-class. Women in the multidimensional vulnerability-class shared multiple risk factors in various domains (psychosocial, medical and socioeconomic risk factors) and lacked protective factors. These women in the multidimensional vulnerability-class more often had adverse outcomes, as compared to the healthy and socioeconomically stable-class. The three classes with risk factors in one domain and protective factors in others did not experience worse outcomes. These results point to the importance of considering the co-existence of multiple risk factors and protective factors that may act as positive exposures or buffering mechanisms promoting resilience.

In **Chapter 3**, we explored the possibility to predict multidimensional vulnerability at population-level using solely nationwide routinely collected data (without self-reported data). Additionally, we reviewed the relevance of adding self-reported data, and identified the most important predictors. Results showed the feasibility of using readily available routinely collected data to predict vulnerability, providing a robust foundation for

longitudinal monitoring and policy making. Nevertheless, results also showed that self-reported data was of added value in the predictions, and self-reported health variables were found to be important predictors to multidimensional vulnerability.

In **Chapter 4**, we described the development of an indicator set to monitor the action program Solid Start on a local level. Experts preferred an indicator set that covers both processes and outcomes, both parents and children, and both risk and protective factors. The final indicator set comprised nineteen indicators within the three phases of the Solid Start program: preconception, pregnancy and after birth. Topics included poverty, psychological/psychiatric problems, stress, smoking, vulnerability, preconception care, low literacy and premature birth. The indicators focused on social determinants of health rather than specific clinical aspects. Additionally, a development agenda was set with topics and indicators that lacked nationwide data or clear operationalization (e.g. stress, unintended pregnancy, loneliness). In the local monitoring of the action program Solid Start, the indicator set can enhance the conversation between policymakers, managers, professionals and other stakeholders about the local situation and developments to prioritize local interventions and policies.

Chapter 5 described the developments and experiences with the action program Solid Start, with a specific focus on cross-sectoral collaboration. This study took place during the program's own first thousand days (i.e. 2019, 2020 and 2021). Quantitative results showed an increasing number of local coalitions Solid Start that involved diverse stakeholders from the medical and social sector, and a growing number of municipalities with plans of action, objectives, ambitions and activities. Qualitative results showed various positive experiences, but also challenges and needs for improvement. Initiating the action program Solid Start increased the sense of urgency for the importance of the first thousand days and stimulated professionals to get to know each other, resulting in more collaborative agreements. Coalition-development varied due to municipalities' unique challenges, focus and historical contexts. Some facilitators for local coalitions Solid Start were an active coordinator as driving force and a shared societal goal. Moreover, stakeholders appreciated the program's strong local focus and opportunities for learning together. However, the action program Solid Start appeared not yet fully incorporated into all professionals' everyday practice. Most common barriers related to systemic integration at macro-level, including limited resources and collaboration-inhibiting regulations. Stakeholders suggested various needs to ensure the program's sustainability, including sustainable funding, supportive regulations, responsiveness to stakeholders' needs, ongoing knowledge development and learning, and better and more client involvement.

Chapter 6 outlines a general discussion with main findings, a reflection on these findings, methodological considerations and recommendations for research, policy, practice and education. We highlight six lessons learned into the adoption of the action program Solid Start (lesson 1 & 2), monitoring (3 & 4) and cross-sectoral collaboration (5 & 6):

1. The adoption was facilitated by a unifying narrative and dedicated champions at all levels
2. National governmental stewardship with strong local focus is a promising combination
3. Considering both risk and protective factors is important for a comprehensive perspective
4. Monitoring requires longitudinal cross-sectoral data and indicators
5. Fostering normative integration is a fundamental first step to collaborate
6. Processes of learning are indispensable in cross-sectoral collaboration

This thesis' monitoring efforts showed a growing link between research, policy and practice. Future research should prioritize participatory methods, realist approaches and engaging experts-by-experience. We also recommend optimizing the use of routinely collected data and studying early effects of the action program Solid Start. For future practice and policy, we advise to create a long-term perspective, integrate the action program Solid Start into everyday practice and stimulate learning processes at different levels. Despite opportunities for improvement, this thesis implies a solid start for the Dutch first thousand days-approach.

NEDERLANDSE SAMENVATTING

In 2018 heeft het ministerie van Volksgezondheid, Welzijn en Sport het landelijk actieprogramma Kansrijke Start gelanceerd, met als doel om ieder kind de best mogelijke start te bieden tijdens de eerste duizend dagen van het leven. De 'eerste duizend dagen' verwijst naar de periode van conceptie tot de tweede verjaardag van een kind, die de basis legt voor gezondheid en welzijn later in het leven en over generaties heen. De ontwikkeling en kansen van kinderen tijdens deze periode worden beïnvloed door medische factoren, maar zijn ook sterk afhankelijk van sociale risico- en beschermende factoren. Het aanpakken van gezondheidsongelijkheid en kwetsbaarheid in het vroege leven vraagt daarom om een geïntegreerde en preventieve aanpak waarin de samenwerking tussen het medische en sociale domein centraal staat. Het actieprogramma Kansrijke Start stimuleert domeinoverstijgende samenwerking en richt zich met name op (toekomstige) ouders en kinderen in een kwetsbare situatie. Sinds 2019 heeft het Rijksinstituut voor Volksgezondheid en Milieu (RIVM) de opdracht om het actieprogramma Kansrijke Start te monitoren. Dit proefschrift vormt de wetenschappelijke basis van de monitor.

In **Hoofdstuk 1** worden de achtergrond, context en doelstellingen van dit proefschrift uitgebreider beschreven. Het overkoepelende doel van dit proefschrift was om inzicht te geven in de invoering van het landelijk actieprogramma Kansrijke Start, met een focus op monitoring en domeinoverstijgende samenwerking. Drie onderzoeksvragen stonden centraal in dit proefschrift: 1) Wat is kwetsbaarheid tijdens de zwangerschap, en hoe kan kwetsbaarheid worden geoperationaliseerd voor monitoring? 2) Welke indicatoren kunnen worden gebruikt om Kansrijke Start op lokaal niveau te monitoren? 3) Wat zijn de ontwikkelingen en ervaringen met Kansrijke Start, specifiek rondom domeinoverstijgende samenwerking?

Hoofdstuk 2 en 3 richtten zich op het monitoren van kwetsbaarheid tijdens de zwangerschap. **Hoofdstuk 2** gaf meer inzicht in kwetsbaarheid door groepen zwangere vrouwen met vergelijkbare risico- en beschermende factoren te identificeren, en door de relatie met nadelige uitkomsten te onderzoeken. Het onderzoek gebruikte routinematig verzamelde data zoals beschikbaar in DIAPER (acroniem voor *Data-InfrAstructure for ParEnts and childRen*) en zelfgerapporteerde data vanuit de Gezondheidsmonitor. Er werden vijf groepen gevonden: meervoudige kwetsbaarheid, sociaaleconomische kwetsbaarheid, psychosociale kwetsbaarheid, hoog zorggebruik en een gezonde en sociaaleconomisch stabiele groep. Vrouwen in de meervoudig kwetsbare-groep deelden diverse risicofactoren voor kwetsbaarheid op verschillende domeinen tegelijk (zowel psychosociaal, medisch als sociaaleconomisch), en hadden vaak een gebrek aan beschermende factoren. Deze vrouwen in de meervoudig kwetsbare-groep hadden vaker nadelige uitkomsten in vergelijking met de gezonde en sociaaleconomisch stabiele-groep. De drie groepen vrouwen met risicofactoren op één domein en beschermende factoren in andere domeinen hadden geen slechtere uitkomsten. Deze resultaten wijzen erop dat het belangrijk is om rekening te houden met zowel de combinatie van verschillende

risicofactoren, als met de aanwezigheid van beschermende factoren die een positieve invloed kunnen hebben of kunnen dienen als buffer om veerkracht te bevorderen.

In **Hoofdstuk 3** onderzochten we of het mogelijk is om op populatieniveau meervoudige kwetsbaarheid in kaart te brengen met alleen routinematig verzamelde data (zonder zelfgerapporteerde data). Daarnaast bekeken we de toegevoegde waarde van het toevoegen van zelfgerapporteerde data in de voorspellingsmodellen, en onderzochten we de belangrijkste voorspellende factoren. De resultaten lieten zien dat het mogelijk is om kwetsbaarheid te voorspellen met direct beschikbare routinematig verzamelde data. Dit biedt een stevige basis voor het langdurig monitoren en ontwikkelen van beleid. De resultaten lieten echter ook zien dat zelfgerapporteerde data de voorspellingsmodellen kunnen verbeteren, en zelfgerapporteerde gezondheidsfactoren bleken belangrijke voorspellers van meervoudige kwetsbaarheid.

In **Hoofdstuk 4** beschreven we de ontwikkeling van een indicatorenset om het actieprogramma Kansrijke Start op lokaal niveau te monitoren. Experts gaven de voorkeur aan een set met indicatoren over zowel processen als uitkomsten, zowel ouders als kinderen, en zowel risico- als beschermende factoren. De vastgestelde indicatorenset bestond uit negentien indicatoren binnen de drie actielijnen van Kansrijke Start: voor de zwangerschap, tijdens de zwangerschap en na de geboorte. Onderwerpen waren onder andere armoede, psychologische/ psychiatrische problemen, stress, roken, kwetsbaarheid, preconceptiezorg, laaggeletterdheid en vroeggeboorte. De nadruk bij de indicatoren lag meer op sociale determinanten van gezondheid dan op specifieke klinische aspecten. Daarnaast werd een zogenoemde 'ontwikkelagenda' opgesteld met geselecteerde indicatoren waarvan gegevens ontbreken of die niet goed uitgewerkt zijn (bijvoorbeeld stress, onbedoelde zwangerschap, eenzaamheid). In het lokaal monitoren van het actieprogramma Kansrijke Start kan de indicatorenset het gesprek bevorderen tussen beleidsmakers, managers, professionals en andere betrokkenen over de lokale situatie en ontwikkelingen, wat kan bijdragen aan het stellen van prioriteiten voor lokale interventies en beleid.

Hoofdstuk 5 beschreef de ontwikkelingen en ervaringen met het actieprogramma Kansrijke Start, specifiek gericht op domeinoverstijgende samenwerking. Dit onderzoek vond plaats tijdens de eerste duizend dagen van het actieprogramma (in 2019, 2020, 2021). De kwantitatieve resultaten toonden een toenemend aantal lokale coalities Kansrijke Start met diverse betrokkenen vanuit sociaal en medisch domein, en een groeiend aantal gemeenten met actieplannen, doelstellingen, ambities en activiteiten. Uit de kwalitatieve resultaten kwamen verschillende positieve ervaringen naar voren, maar ook uitdagingen en behoeften aan verbetering. Het actieprogramma Kansrijke Start versterkte het gevoel van urgentie voor het belang van de eerste duizend dagen en stimuleerde professionals om elkaar te leren kennen, wat leidde tot meer samenwerkingsafspraken. De ontwikkeling van coalities varieerde per gemeente vanwege de unieke uitdagingen, focus en historische context van iedere gemeente. Belangrijke bevorderende factoren voor lokale coalities Kansrijke Start waren een actieve ambassadeur/ kartrekker en het hebben van een

gezamenlijk doel. Bovendien waardeerden betrokkenen de sterke lokale focus van het programma en mogelijkheden om met elkaar te leren. Het actieprogramma Kansrijke Start bleek echter nog geen onderdeel van de dagelijkse praktijk voor veel zorgverleners. Veelgenoemde barrières hadden betrekking op systemische integratie op macroniveau, waaronder beperkte financiële middelen en wet- en regelgeving die samenwerking bemoeilijkt. Betrokkenen noemden verschillende behoeften om het programma te borgen, waaronder structurele financiering, passende wet- en regelgeving, aandacht voor behoeften van betrokkenen, voortdurende kennisontwikkeling en -leren, en het vaker en beter betrekken van ervaringsdeskundigen.

Hoofdstuk 6 bevat een algemene discussie met hierin de belangrijkste bevindingen, een reflectie op deze bevindingen, methodologische overwegingen en aanbevelingen voor onderzoek, beleid, praktijk en onderwijs. We benadrukken zes geleerde lessen over de invoering van het actieprogramma Kansrijke Start (les 1 & 2), monitoren (3 & 4) en domeinoverstijgende samenwerking (5 & 6):

1. De invoering werd gefaciliteerd door een verbindend narratief en toegewijde ambassadeurs op meerdere niveaus
2. Ondersteuning vanuit de landelijke overheid gecombineerd met een sterke lokale focus is een veelbelovende combinatie
3. Het meenemen van zowel risico- als beschermende factoren is belangrijk voor een compleet beeld
4. Monitoring vereist longitudinale domeinoverstijgende data en indicatoren
5. Het bevorderen van normatieve integratie is een fundamentele eerste stap voor samenwerking
6. Leerprocessen zijn onmisbaar in domeinoverstijgende samenwerking

De monitoringsactiviteiten zoals beschreven in dit proefschrift tonen een toenemende verbinding tussen onderzoek, beleid en praktijk. Voor toekomstig onderzoek is het belangrijk om de focus te leggen op participatieve methoden, *realist* benaderingen en het betrekken van ervaringsdeskundigen. Wij raden ook aan om het gebruik van routinematig verzamelde data te optimaliseren en om de eerste effecten van het actieprogramma Kansrijke Start te onderzoeken. Voor toekomstig beleid en praktijk adviseren we om een langetermijnperspectief te creëren, het actieprogramma Kansrijke Start te integreren in de dagelijkse praktijk, en leerprocessen op verschillende niveaus te bevorderen. Ondanks ruimte voor verbetering, impliceert dit proefschrift een kansrijke start voor de Nederlandse eerste duizend dagen-aanpak.

LIST OF PUBLICATIONS

International publications

Molenaar, J. M., Leung, K. Y., van der Meer, L., Klein, P. P. F., Struijs, J.N., Kiefte-de Jong, J. C. Predicting population-level vulnerability among pregnant women using routinely collected data and the added relevance of self-reported data. Submitted.

Uzan, L. M., Brust, M., **Molenaar**, J. M., Leistra, E., Boor, K., & Kiefte-de Jong, J. C. (2024). A cross-sectional analysis of factors associated with the teachable moment concept and health behaviors during pregnancy. *BMC Pregnancy and Childbirth, 24*(1), 147.

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Molenaar, J. M., van der Meer, L., Bertens, L. C. M., de Vries, E. F., Waelput, A. J. M., Knight, M., Steegers, E. A. P., Kiefte-de Jong, J. C. & Struijs, J. N. (2023). Defining vulnerability subgroups among pregnant women using pre-pregnancy information: a latent class analysis. *European Journal of Public Health, 33*(1), 25-34.

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Lodenstein, E., **Molenaar**, J. M., Ingemann, C., Botha, K., Mkandawire, J. J., Liem, L., Broerse, J. E. W. & Dieleman, M. (2019). "We come as friends": approaches to social accountability by health committees in Northern Malawi. *BMC Health Services Research, 19*, 1-14.

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Bibliography available online at ORCID: https://orcid.org/0000-0002-8025-040X

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A.W. van Meijeren-van Lunteren, J.M. **Molenaar**, I.C. Boesveld, B.J. Hoffman, A.J. Brouwer-Prusak, R.J.P. Hendrikx, P.P.F. Klein, J.N. Struijs (2024). Monitor Kansrijke Start 2023. Bilthoven: Rijksinstituut voor Volksgezondheid en Milieu.

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Joyce Molenaar, Zoë Scheefhals, Eline de Vries, Jeroen Struijs (2021). Monitor Kansrijke Start 2020. Bilthoven: Rijksinstituut voor Volksgezondheid en Milieu.

J.N. Struijs, E.F. de Vries, Z.T.M. Scheefhals**,** J.M. **Molenaar**, C.A. Baan (2020). Integrale bekostiging van de geboortezorg: ervaringen na drie jaar en de eerste zichtbare effecten.

Jeroen Struijs, Eline de Vries, Anita Suijkerbuijk, **Joyce Molenaar**, Zoë Scheefhals, Caroline Baan (2020). Monitor Kansrijke Start 2019. Bilthoven: Rijksinstituut voor Volksgezondheid en Milieu.

Hendrix M, **Molenaar** J., Korstjens I, Nieuwenhuijze M. Gezamenlijke besluitvorming in de praktijk (2018). *Tijdschrift voor verloskundigen, 4.*

PORTFOLIO

Training, courses and congress visits

2023
- Visiting scholar at Child Health Evaluation and Research Center (CHEAR) at the University of Michigan, and the department of General Pediatrics of Boston Children's Hospital
- International Conference on Integrated Care. IFIC. Antwerp, Belgium
- Congres Sociale verloskunde. Erasmus MC. Rotterdam
- Loopbaancoachingstraject
- Bijeenkomst De plek van risico in de geboortezorg van morgen. Amsterdam UMC. Amsterdam
- Meeting (big) data for a Solid Start - needs and opportunities for early years professionals. Bernard van Leer Foundation. Utrecht

2022
- Normal Labour and Birth Conference 2022. Aarhus University. Aarhus, Denmark
- Bijeenkomst Gezondheidsverschillen verminderen? Pak stress aan. Kenniscentrum Psychologie en Economische Gedrag. Leiden
- Conferentie Samen voor een Kansrijke Start. College Perinatale Zorg. Driebergen-Zeist
- Congres Kansrijke Start Dichtbij. Ministerie van VWS. Utrecht
- MOOC coursera - Population Health: predictive analytics. Leiden University
- Difference-in-Differences training - Pedro Sant'Anna. Causal solutions. Online
- Lecture Inequalities in maternal and child health – where do the problems really lie? By Marian Knight at Leiden University

2021
- Congres Kansrijke Start heeft de toekomst. Ministerie van VWS. Online
- Course Academic writing for PhD's. Leiden University. Online
- Basistraining projectmatig creëren

2020
- Course Introduction to Latent Class Analysis. UCL GOS ICH Centre for Applied Statistics courses. Online
- Congres Samen verder met Kansrijke Start. Ministerie van VWS. Amersfoort
- Summer school Population Health Management. LUMC. Online
- MOOC coursera - Population Health: fundamentals of Population Health. Leiden University
- Regie op je energie-traject (coaching & workshops)
- Lectures Q-methodology and Introduction in Random Forest
- Webinars Bewoners in een achterstandssituatie betrekken bij gezondheid en Inequality. CBS. Online
- Startbijeenkomst Gezonde Kansrijke Start Leiden. Gemeente Leiden. Leiden

2019
- Jaarcongres Perined. Perined. Doorn
- Conferentie 'Uitgerekend Nu?!'. Ministerie van VWS. Utrecht
- Werkconferentie 'Van preconceptie tot peuter: de eerste 1000 dagen'. LUMC-campus Den Haag. Den Haag
- PhD introductory meeting. Leiden University. Leiden
- BROK course. NFU
- Course Basic methods and reasoning in biostatistics. LUMC. Leiden

2019 -	• Meelopen met verschillende zorgprofessionals die zorg en ondersteuning
2023	bieden tijdens de eerste duizend dagen
	• Attendance weekly and monthly research meetings at RIVM and Health Campus The Hague
	• Referee for multiple journals (including International Journal of Integrated Care)

Grants

2020	Meeschrijven onderzoeksvoorstel ZonMw subsidie 'Verbetering lokale praktijk: Verbinding medisch en sociaal domein omtrent ondersteuning onbedoelde zwangerschap en kwetsbaar ouderschap in Den Haag' (gehonoreerd)

Knowledge dissemination

2023	• Oral presentation at Child Health Evaluation and Research Center-seminar. Ann Arbor, U.S.
	• Oral presentation at BIDMC/Harvard Newborn Health Services and Epidemiology Seminar. Boston, U.S.
	• Presentaties bij Pharos en het College Perinatale Zorg. Utrecht
	• Co-guidance of thesis M. Peters-Koning, student NSPOH Arts Maatschappij en Gezondheid. Title: *Risicofactoren voor een langzame spraaktaalontwikkeling bij Haagse 2-jarigen*
2022	• Oral presentation Normal Labour and Birth Conference 2022. Aarhus, Denmark
	• Informatiemarkt congres Kansrijke Start Dichtbij. Utrecht
	• Begeleiden brainstorm ontwikkelen: plan van aanpak Haagse Kansrijke Start per 2023
2021	• Presentatie (video) congres Kansrijke Start heeft de toekomst. Online
	• Guidance thesis D. Kooijman, bachelor student LUMC Bachelor geneeskunde. Title: *Screening op kwetsbaarheid in gezinnen: het nu en later*
	• Co-guidance of thesis L. van Opstal, master student VU University Health Sciences. Title: *Pregnancy, a teachable moment for changing health behaviour*
	• Presentation symposium minor 'Taking care of tomorrow' 2020
2020	• Workshop congres Samen verder met Kansrijke Start. Ministerie van VWS. Amersfoort
	• Oral presentation International Conference on Integrated Care. Online
	• Presentation symposium minor 'Taking care of tomorrow' 2020
	• Deelname expert meeting Causal Loop Diagram - complexiteit van een ongezonde leefstijl tijdens de eerste twee levensjaren
2019 -	• Multiple presentations during Coffee & Research, science lunch, PhD-meetings
2023	• Informatiemarkt tijdens jaarlijkse werkconferenties Health Campus Den Haag
	• Assessing multiple internship reports
	• Participation Career Carousel for master Population Health Management

CURRICULUM VITAE

Joyce Molenaar was born 31 March 1993 in Schagen, the Netherlands. She completed a bachelor in Health Sciences at the Vrije Universiteit (VU) Amsterdam in 2014, and obtained a Global Health research master's degree at the same university in 2016. During several research internships, she became acquainted with research related to pregnancy and childbirth. She studied contraceptive decision-making at Bureau Studentenartsen in Amsterdam and conducted research on client-centered collaboration in maternity care networks in North Holland. Additionally, she traveled to Malawi for a study into social accountability in maternal health service delivery.

From 2016 to 2018, Joyce worked at the Research Centre for Midwifery Science, Academie Verloskunde Maastricht, Zuyd Hogeschool. Her research project was focused on shared decision-making in maternity care. In co-creation with healthcare providers and clients, she and her colleagues developed an intervention to promote shared decision-making during pregnancy, childbirth and postnatal care: a tailored e-learning for healthcare providers and a concise film for clients. During several periods from 2016 to 2018, Joyce also worked as a researcher and teacher at Athena Institute, VU Amsterdam. In this position, she contributed to a research project that reflexively monitored the system's innovation towards integrated maternity care. In 2017, Joyce also started volunteering at BuurtBuik, an initiative that tackles food waste and fosters social contact by preparing meals for and with local residents using surplus foods. She was a founding member and the chairperson of the first BuurtBuik-location in Utrecht.

In 2019, realizing that she greatly enjoyed doing research related to pregnancy and childbirth, Joyce started her PhD-project focusing on the action program Solid Start, which resulted in this dissertation. She was affiliated with the National Institute for Public Health and the Environment (RIVM) and the Health Campus The Hague, department of Public Health and Primary Care, Leiden University Medical Centre. During her PhD, Joyce also contributed to the national and local Solid Start-monitoring projects at RIVM, and was involved in various research and teaching activities in The Hague. From August to October 2023, she was a visiting scholar at the Child Health Evaluation and Research Center (CHEAR) at the University of Michigan, and the department of General Pediatrics of Boston Children's Hospital.

Joyce continues her work at RIVM on various projects related to the first thousand days and cross-sectoral collaboration. Next to that, she works as a postdoctoral researcher in a participatory action research project at Health Campus The Hague, focused on improving local practice regarding unintended pregnancies and vulnerability during parenthood by strengthening collaboration.

DANKWOORD

In de zomer van 2019, na een prachtige reis rond de wereld, startte ik mijn promotietraject. Ook dit avontuur had ik niet willen missen. Ik kijk terug op een bijzondere tijd vol kansen, inspiratie, leermomenten en fijne samenwerkingen. Hierbij wil ik graag mijn waardering uitspreken voor iedereen die eraan heeft bijgedragen dat ik dit proefschrift nu met een grote glimlach kan delen. Zonder jullie was het niet gelukt, of veel minder leuk geweest. Duizendmaal dank voor jullie bijdrage en aanmoediging.

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De combinatie van het schrijven van een proefschrift met projectwerk bij het RIVM was soms best even puzzelen, en toch had ik mijn promotietraject niet anders willen vormgeven. De afwisseling in werkzaamheden gaf plezier en voldoening. Het zorgde ook dat ik kon samenwerken met vele collega's bij het RIVM, de Health Campus Den Haag en verschillende organisaties daarbuiten. Mijn co-auteurs -Inge Boesveld, Eline de Vries, Peter Paul Klein, Ka Yin Leung, Lindsey van der Meer, Adja Waelput, Eric Steegers, Loes Bertens en Marian Knight- wil ik specifiek bedanken voor alle hulp en waardevolle suggesties voor de artikelen in dit proefschrift. Ook ben ik blij met ieder ander die heeft meegedacht, (statistische) ondersteuning bood en mijn enthousiasme verder aanwakkerde. Dat geldt zeker voor het DIAPER-team en alle promovendi in Bilthoven en Den Haag met wie ik samen kon leren en vieren. Ik heb veel plezier gehad met jullie en andere collega's tijdens lunchwandelingen, borrels, etentjes, congressen, sportieve activiteiten en schrijfweken/dagen. Dank jullie wel allemaal.

Alle deelnemers wil ik bedanken voor het delen van hun ervaringen en ideeën. Judith en Jenny, bedankt dat we van jullie als ervaringsdeskundigen mogen leren in onze projecten. Daarnaast waardeer ik dat ik met meerdere zorg- en hulpverleners mocht meelopen om inzicht te krijgen in de dagelijkse praktijk. Ik keek hier altijd naar uit en vertrok vol inspiratie. Dank daarvoor. Ik hoop van harte dat de inzichten uit dit proefschrift waardevol zijn voor beleid en praktijk, en dat we vanuit verschillende bronnen van kennis (ervaring, praktijk, wetenschap) blijven samenwerken aan een kansrijke start.

Gary Freed and Christopher Landrigan, thank you for warmly welcoming me to Ann Arbor and Boston. I highly enjoyed writing the final parts of my PhD-thesis in such vibrant and inspiring environments.

Inge en Rosanne, het voelt goed dat jullie als enthousiaste paranimfen naast me staan tijdens mijn verdediging. Inge, jouw wijze en bemoedigende woorden over het belang van

geduld, discipline en structuur hielpen me richting de eindstreep. Jij biedt een luisterend oor en gezelligheid, en ik waardeer onze samenwerking enorm. Roos, ik ben heel blij met jou als betrokken vriendin. Je inspireert mij en vele anderen met je inzet voor een eerlijke en duurzame wereld. Met jou, Jort en onze andere vrienden hoop ik nog lang belangrijke momenten te delen.

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