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Reducing the administrative burdens and regulatory pressure for patients, parents, and healthcare professionals in the Duchenne Muscular Dystrophy (DMD) care pathway: an observational case study

Master Thesis
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Preface

With this research project and master thesis, my time as a student is coming to an end. I studied for just over five years with great pleasure at the Radboud University in Nijmegen. During the first 3 years, I obtained my bachelor's degree in medical biology after which I started the two-year master's program in Science, Management, and Innovation (SMI). In the beginning, I needed time to get used to the SMI track since it is still completely different from the bachelor in Medical Biology. Because of the many collaborative projects and social fellow students, I felt comfortable in this specialization. I would like to thank dr. Ian Cameron for his support as coordinator of the health track of this master's program. I could contact him with all my questions, and he also helped me in finding a suitable organization for my thesis research.

I would like to thank Erik Wackers for organizing the weekly intern meeting at IQ Healthcare. During these pleasant meetings, we discussed each other's research progress and gave each other tips on approaching certain issues. In addition, I would like to thank prof. dr. Patrick Jeurissen for his feedback and tips throughout the internship. I would also like to thank drs. Saskia Houwen, Gera Peters, and Maarten Stessel from the rehabilitation department at the center of expertise in Nijmegen for their help and feedback during the research and the opportunity to observe a day at their department. Then, I want to give a great appreciation to dr. Niek Stadhouders who gave me the possibility to work on this interesting and important research. I appreciated the way we worked together, I could always contact him with questions whenever I got a problem. Niek always made time to discuss these things. Finally, I want to thank the participants in the interviews, without them there would simply be no results. I heard many interesting but also painful personal stories.

Thijs Som
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Abstract

Background Duchenne Muscular Dystrophy (DMD) care requires a high standard of multidisciplinary, integrated care and service coordination among hospital, community, social, and primary healthcare services. Since it is a progressive condition, the patient is in contact with different stakeholders at different stages of the care pathway. The organization and coordination of such complex care may cause healthcare professionals, patients, and parents to experience heavy administrative burdens and regulatory pressure.

Objectives The aim of this study was to identify the extent, sources, mechanisms or causes, and potential solutions to reduce administrative and regulatory burdens perceived by healthcare professionals, patients, and parents in the DMD care pathway.

Methods A literature review was conducted to identify available literature on administrative and regulatory burdens experienced by parents and patients. In addition, a day was spent at a pediatric neurology outpatient clinic to get a brief insight into this part of the care pathway. Finally, seven interviews with healthcare professionals from the DMD care pathway and four interviews with parents of DMD patients were conducted.

Results Administrative burdens and regulatory pressure are significant for parents. These burdens arise at multiple locations in the care systems, of which municipalities, suppliers of assistive devices, health insurers, healthcare professionals, and personal characteristics of parents were mentioned most often in the literature and interviews as a source of administrative and regulatory burdens. The extent of perceived administrative and regulatory burdens depends on a number of factors, related to personal capabilities, and capabilities of caregivers and payers, where large differences were mentioned.

Conclusion We find a multitude of sources of administrative and regulatory burdens, of which the provision of medical assistive devices by municipalities and suppliers was recognized as the most significant cause of administrative burdens and pressure. We advocate the centralization of DMD applications around the Social Support Act. This act covers assistance, support, facilities, and services for people with disabilities. Centralization will most efficiently reduce the perceived administrative burdens and regulatory pressure by healthcare professionals, patients, and parents. For patients and parents who experience great difficulty navigating the care pathway, a case manager could assist.

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1. Introduction

Healthcare systems are constantly changing due to the use of new technological developments, the expansion of biomedical and clinical knowledge, and aging populations (Nilsen et al., 2020). As a result, the healthcare demands of patients are increasing and becoming more complicated (Babalola, 2017). Patients with previously untreatable conditions experience more and more possibilities in terms of treatments. To meet patients' increasing and increasingly complex healthcare demands, healthcare organizations aim to provide patient-centered care. This approach respects the patient's values, needs, experiences, and preferences in the coordination and provision of care. The use of the patient-centered care concept has been proven to deliver high-quality care while resulting in less waste of resources, decreasing costs, and greater satisfaction among patients and healthcare professionals (Gluyas, 2015). However, complex personalized care provision could cause patients and healthcare professionals to experience administrative and regulatory burdens. Especially if care needs cross multiple domains, coordination and communication become increasingly complex. High administrative burdens and regulatory pressure could result in delayed or foregone care, resulting in increasing healthcare costs (Kyle & Frakt, 2021).

Extensive research has already been conducted regarding the administrative burdens and regulatory pressure experienced by healthcare professionals. For example, a study shows that Dutch healthcare professionals spend on average about 52 minutes of a workday on quality registrations. In addition, only 36% of these quality registrations can be used to improve care and the associated administrative burdens and regulatory pressure weigh on the motivation of healthcare professionals (Babbott et al., 2014; Zegers et al., 2022). Another study demonstrates that U.S. physicians consume one-sixth of the working hours on administrative tasks and that these tasks lower their work satisfaction (Woolhandler & Himmelstein, 2014). About a quarter of the total healthcare expenditures in the U.S. is spent on administrative costs, which amount to $1 trillion annually (Sahni et al., 2021). In the Netherlands, about 20% of total healthcare expenditures is spent on administrative costs, which is significantly higher compared to other European countries (Himmelstein et al., 2014). The existing literature on administrative burdens and regulatory pressure in healthcare primarily focuses on the burdens experienced by healthcare professionals. The perspective of patients is underrepresented in the literature, while certain patient groups could experience significant administrative burdens (Herd & Moynihan, 2021).

Patients with complex conditions or multimorbidities need to make a substantial amount of arrangements regarding the care they require. They have to organize and request necessary care, medical devices, and facilities throughout the care pathway, involving many different stakeholders.

Especially medical conditions that develop at a young age require a major role for parents (Rosland, 2009). Since there is limited literature available on the administrative burdens experienced by patients and parents, this study will focus on the administrative burdens and regulatory pressure experienced by all stakeholders in the care pathway of a complex condition. This study will focus especially on the care pathway of Duchenne Muscular Dystrophy (DMD).

DMD is a rare, severe inherited muscle disease in which the muscles slowly break down causing the patient to lose strength. It's a progressive, X-linked disorder which means that the disease occurs almost exclusively in males and is inherited through the mother line (Sun et al., 2020). The disorder is caused by mutations in a gene that encodes for the cytoskeletal protein dystrophin. Due to specific mutations in this gene, the dystrophin protein is not or is only produced in small quantities. Muscles without dystrophin are more susceptible to damage, resulting in the loss of muscle tissue and function over time (Duan et al., 2021; Nowak & Davies, 2004). DMD affects approximately 1 in 5.000 male births which amounts to 20.000 new diagnoses per year in the world (MDA, 2019). According to the Leiden University Medical Center (LUMC), about 400-500 patients are suffering from DMD in the Netherlands (LUMC, 2016). DMD affects many parts of the body as the disease results in the atrophy of skeletal, cardiac, and pulmonary muscles (VSN & NHG, 2006). The first signs of the disease are noticed around the third year of life. At this age, children with DMD have significant motoric development delays, gait abnormalities, difficulties with rising from the ground, and frequent falls. Around the age of 10-12, patients are confined to a wheelchair, and around the age of 18 and 20 years, they require artificial respiration (Venugopal & Pavlakis, 2022; Yiu & Kornberg, 2015). In addition, dystrophin is active in the brain, which can cause DMD patients to develop learning and behavioral problems (Anderson et al., 2002). There is no cure available for DMD, so the current interventions focus on the prevention and management of symptoms to improve the quality of life and longevity. The life expectancy of patients with DMD has improved over the last decades due to the use of corticosteroid therapy, antisense therapy, and the performance of scoliosis surgery. Today, the life expectancy for patients with DMD is between 30 and 40 years (Landfeldt et al., 2020).

A rare condition like DMD requires a high standard of multidisciplinary, integrated care and service coordination among hospital, community, social, and primary healthcare services (Ward et al., 2022). Throughout life, the symptoms of DMD get worse and the patient can do progressively less. As a result, the patient is in contact with many different stakeholders at different stages of the care pathway. Since DMD is expressed at an early age, parents are closely involved in the organization and request of care and services. Therefore, in this disease setting, administrative burdens and regulatory pressure would not be experienced by healthcare professionals, but also by patients and especially by parents. In 2018, the Dutch Ministry of Health, Welfare and Sport introduced the national action plan '(De)-

Regulate Healthcare' to reduce administrative burdens and regulatory pressure in healthcare. This plan contains concrete action points to reduce the administrative burdens and regulatory pressure experienced by healthcare professionals and patients (VWS, 2018).

However, the administrative burdens and regulatory pressure for healthcare professionals are still increasing and the pressure and burdens for patients and parents are barely addressed (Maris et al., 2020). As mentioned before, there is a gap in the literature regarding the administrative burdens and regulatory pressure experienced by patients and parents. Most literature focuses on the burdens that healthcare professionals experience. In addition, most of the literature originates from the U.S., which is not directly representative for other countries with different healthcare systems. For example, universal health systems generally provide comprehensive care and support especially for low-income people, a characteristic largely absent in the U.S. system. Therefore, one would expect patients and healthcare professionals experience fewer problems in universal health systems such as in the Netherlands. Furthermore, existing literature addresses administrative burdens and regulatory pressure in the healthcare system in general and not on specific healthcare pathways. This study focuses on the administrative burdens and regulatory pressure specifically experienced by all the stakeholders in the DMD care pathway. This provides an overall picture of the administrative and regulatory burdens associated with the care for a rare disease. Moreover, this study focuses on the DMD care pathway in the Netherlands. This is interesting because the Netherlands has a universal healthcare system in which the general practitioner plays an important role (Wammes et al., 2020). Finally, the Dutch healthcare system has recently seen a decentralization of policy to provide more customized care (VNG, 2014).

The purpose of this study is to identify the source and size of administrative burdens and regulatory pressure experienced by DMD patients, their parents, and the involved healthcare professionals in organizing and requesting DMD care and services. The mechanisms or causes behind these administrative burdens and regulatory pressure will be identified and potential solutions to reduce the burdens and pressure will be proposed. At last, recommendations will be made to help reduce the administrative burdens and regulatory pressure for DMD patients, their parents, and the healthcare professionals involved.

The following research questions will be addressed in this study:

- What is the extent of the perceived administrative burdens and regulatory pressure in the DMD care pathway?
- What are the sources of perceived administrative burdens and regulatory pressure in the DMD care pathway?

- What are the mechanisms or causes behind the perceived administrative burdens and regulatory pressure in the DMD care pathway?
- What are potential solutions to reduce experienced administrative burdens and regulatory pressure in the DMD care pathway?

2. Literature and conceptual framework

This section of the thesis presents the background and main theoretical concepts of this research. First, a brief overview of the Dutch healthcare system will be provided. Then, the DMD care pathway will be explained. Finally, two models used for the analysis of the data will be described.

2.1 Overview of the Dutch healthcare system

The Dutch healthcare system is based on several general principles: access to healthcare for all, solidarity through mandatory health insurance, and high-quality healthcare services (VWS, 2016). There are four laws of Dutch Healthcare: Firstly, all residents of the Netherlands should be insured according to the Health Insurance Act. This act is financed by premiums paid by all residents and guarantees short-term medical care such as general practitioner care, hospital care, mental healthcare, and medication. In addition, for a large proportion of DMD patients, it will be necessary to organize intensive and long-term care. This care is covered by the Long-term Care Act. This law provides high-level care for vulnerable individuals, such as people with severe disabilities. Patients and their parents can choose whether to finance this care contractually or through the personal healthcare budget. With the personal healthcare budget, patients or parents purchase necessary care and services themselves. The third act is the Social Support Act. This act covers assistance, support, facilities, and services for people with disabilities. The last act is the Youth Act, which covers help and care for young people and their families with growing up, parenting, and psychological problems and disorders. In 2015, the Social Support Act and the Youth Act were changed to provide more customized care for patients by providing care close to home. This change has decentralized government tasks, giving municipalities the responsibility for youth care and care for the long-term ill (Tweede-Kamer, 2021). So this kind of care and services are requested at the municipality and is, depending on the preference of the patients and parents, financed contractually or through the personal healthcare budget (VWS, 2016). Since DMD has a complex disease progression, the patient requires a diversity of care and services at different moments. Therefore, patients can be covered by all four laws of the Dutch healthcare system.

2.2 Overview of the Dutch DMD care pathway

The care pathway of DMD can be divided into three categories of care, namely primary care, secondary care, and tertiary care. Primary care for DMD patients is mainly provided by the general practitioner and paramedics in the patient's home environment. Most secondary care is provided by a rehabilitation center that is often integrated with a mytylschool, which is a special school for physically disabled children. Patients who are attending a mytylschool receive certain therapeutic services during school hours, eliminating the need for these visits after school. The rehabilitation centers are

usually located close to a patient's home. Patients that are not affiliated with a rehabilitation center receive secondary care from a local hospital. Most tertiary care is provided by centers of expertise. In 2015, the Netherlands Federation of UMCs (NFU) started to designate centers of expertise for rare diseases. These centers bring knowledge and experience together about a particular rare disease, they develop guidelines, coordinate research, ensure appropriate referral of patients, and can serve as the second opinion for patients. In addition, these centers provide patients and healthcare professionals with advice on effective care (Hendriks et al., 2016). The Radboudumc forms together with the Leiden University Medical Center (LUMC), the Kempenhaeghe center for neurological learning disabilities, and the organizations Duchenne Parent Project and Spierziekten Nederland the center of expertise Duchenne Center Netherlands (Duchenne-Becker-Expertisecentrum, n.d.). A multidisciplinary outpatient clinic is established in Leiden and Nijmegen. Here, diagnoses are made, the necessary checks and treatments are carried out, and scientific research is conducted to expand knowledge about DMD. Since many organs are affected by DMD, the centers consist of various professionals working together in the care pathway. Patients are advised to visit the center of expertise twice a year in the early stages of the disease. Later, a check-up once a year is sufficient (de Coo et al., 2016; Federatie-Medisch-Specialisten, 2021). During the outpatient visit, the patient is examined by a pediatric neurologist, pediatric rehabilitation specialist, physiotherapist, occupational therapist, speech therapist, and dietician. In addition, depending on the age and stage of the disease, assessments are carried out by a cardiologist, pulmonologist, and radiologist. On such a day, information is gathered about the patient's disease process (Radboudumc, n.d.). Based on patient questions, needs, and various results, advice is given after consultation with all healthcare professionals involved. The center of expertise also discusses results and advices with the healthcare professionals in primary and secondary care to arrange necessary care and services in the patient's home environment. Another stakeholder that provides tertiary care is the center for home respiration. When the respiratory function of the patient deteriorates too much, the patient needs to visit the Center for Home Respiration in Utrecht or Rotterdam (CTB) to initiate nocturnal noninvasive ventilation (de Coo et al., 2016; Federatie-Medisch-Specialisten, 2021).

In addition to the three categories of care, there are other actors involved in the DMD care pathway. Important stakeholders are municipalities, suppliers of assistive devices, health insurers, sports clubs, employers, and regular schools for patients who do not go to a mytyl school. Also, the Duchenne Parent Project (DPP) and Spierziekten Nederland play an important role in the DMD care pathway. These are foundations that aim to encourage scientists, physicians, and companies to stimulate research for a treatment or cure for DMD. In addition, the DPP and Spierziekten Nederland offer a supportive role for DMD families (DPP, n.d.). Table 1 in appendix 1 represents an overview of the care

providers, the type of care and the frequency of use, the category of care, and the various healthcare laws involved.

2.3 Dahlgren-Whitehead and Levesque's model

The purpose of this research is to identify the source and size of administrative burdens and regulatory pressure, the mechanisms or causes behind these administrative and regulatory burdens, and solutions to reduce these pressures and burdens through the DMD care pathway. These items will be identified by conducting a literature review and interviews.

To map the sources and the mechanisms or causes of experienced administrative burdens and regulatory pressure related to the DMD care pathway, two commonly used models from the literature are used. The first model is the Dahlgren-Whitehead model of health determinants. With this model, the source of experienced administrative burdens and regulatory pressure can be partially identified. This model shows the relation between an individual, the environment, and health. As shown in figure 1, individual characteristics including age and sex are placed at the center surrounded by the various layers that could influence health in a positive or negative way (van Hartingsveldt, 2016). The first layer consists of lifestyle factors such as nutrition, smoking, and exercise. The second layer involves the social well-being of persons. This is formed by the influence of social and community networks which includes family, friends, and relatives.

Figure 1| Dahlgren-Whitehead model of health determinants (Dahlgren & Whitehead, 2021).

The third layer determines the extent to which people can maintain their health, this includes living and working conditions, food supply, and access to important resources and services such as education and healthcare. The component health care services contains stakeholders involved in the provision of healthcare, so the healthcare professionals from the different categories of care. Finally, the fourth layer shows that the individual's health is determined by socio-economic, cultural, and environmental factors.

This Dahlgren-Whitehead model of health determinants is widely used in research as it helps to consider different layers of influence on health, to broaden perspectives outward to consider the potential role of broader and wider health determinants, and thereby building a complete picture (Dahlgren & Whitehead, 2021). The model can be used to partially identify and categorize the source of perceived administrative burdens and regulatory pressure in the DMD care pathway as it encompasses the factors one may encounter while arranging care and services and thus the possible sources of administrative burdens and regulatory pressure.

To map the mechanisms or causes behind the administrative burdens and regulatory pressure, the Levesque conceptual model framework for healthcare access is used. This model provides an interesting and expansive perspective by outlining five dimensions of access and five capabilities of the population to access healthcare (figure 2).

Levesque's model shows the process of care demand and proposes a multidimensional perspective on access to healthcare in the context of health systems with dimensions of approachability, acceptability, availability and accommodation, affordability, and appropriateness. In addition, it considers the socioeconomic determinants of the population that results in the integration of the following five capabilities of individuals and populations: to perceive, seek, reach, pay, and engage in healthcare (Cu et al., 2021). This framework is useful because it encompasses both health systems' and patients' perspectives on access to care, allowing researchers to look at barriers to healthcare access that result from health systems' failures and people's abilities. Using this model, an attempt will be made to determine where in the process of care demand administrative burdens and regulatory pressure occur and what causes them.

The models described above are used as a starting point for analyzing the data regarding the source and mechanisms or causes behind administrative burdens and regulatory pressure. During the data-analysis, the models will be used alongside each other to keep an overview. These models can be considered as complementary in analyzing sources and mechanisms or causes of administrative burdens.

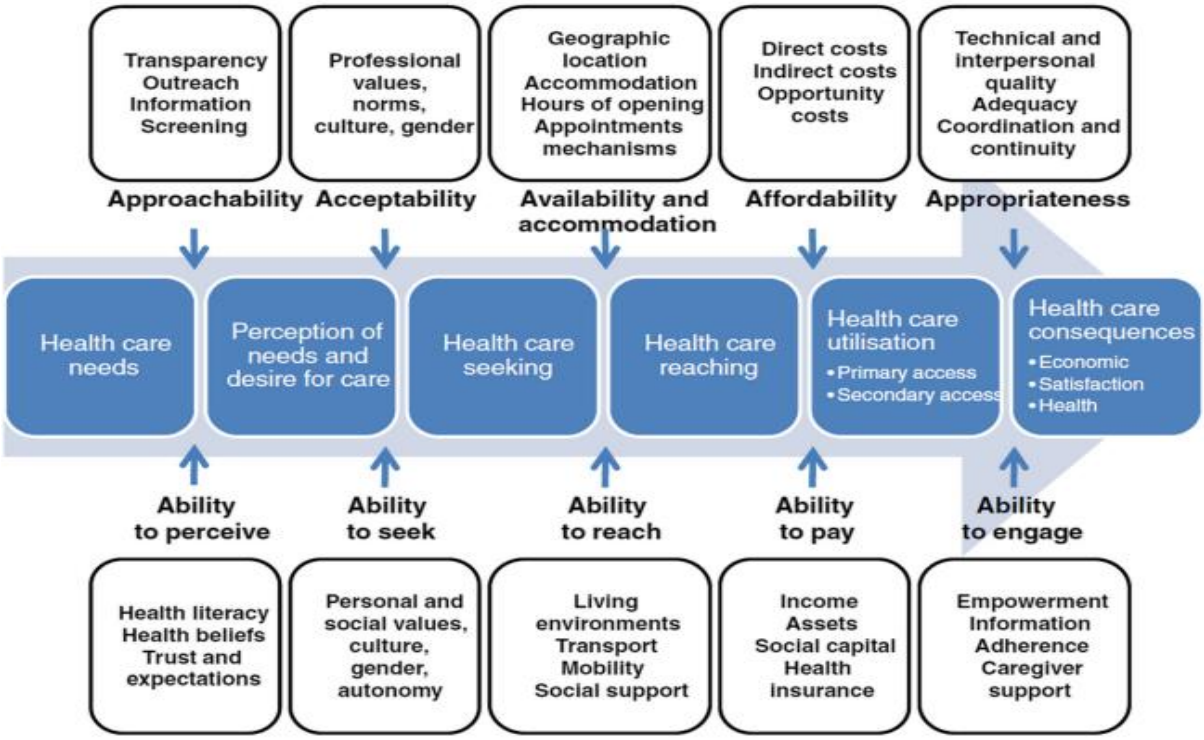

Figure 2| Levesque's Conceptual Framework of Access to Health (Cu et al., 2021).

3. Methods

At the start of this study, a literature review was conducted on the administrative burdens and regulatory pressure experienced by patients and parents. In addition, observational research was performed by spending a day at the pediatric neurology outpatient clinic of the Radboudumc in Nijmegen. Finally, interviews were conducted with four parents of DMD patients and seven healthcare professionals working in the DMD care pathway. A different topic list was prepared for the parents and the healthcare professionals. A semi-structured interview method was chosen, and the transcripts of the audio fragments were coded and analyzed.

3.1 Literature review

PubMed was searched for original, published studies on administrative burdens and regulatory pressure experienced by patients and parents. The PICOS tool was used to compile the search string (Brown, 2020). The search strategy contained the following main components: patients, administrative burdens, and qualitative research. There were no language or publication date restrictions applied. The complete search string can be found in Appendix 2. The search was performed on May 6, 2022. Because it was expected that there was limited literature available on this topic, it was decided to conduct the search only in PubMed and not to extend the search strategy to other databases. PubMed was chosen because it contains biomedical literature and MeSH terms can be used in the search strategy. The retrieved studies were screened on title and abstract in Rayyan by two researchers. References were excluded when they met at least one of the following exclusion criteria: (1) the study was not patient-, parent- or caregiver-centered, (2) the study did not cover administrative- or bureaucratic burdens, or (3) the study was not a qualitative study. In case of a doubt, a study was included. Then, the included references were screened on full text in Rayyan by two researchers. Studies were excluded when they met one of the previously mentioned criteria. The screening of both the title and abstract and the full text was performed by two independent researchers and conflicts were resolved by consensus after discussion.

3.2 Observational research

On April 11, 2022, a day was spent at the pediatric neurology outpatient clinic of the Radboudumc in Nijmegen. Two consultations with a DMD patient, their parents, and a pediatric rehabilitation specialist were attended to get a brief insight into this part of the care pathway. In addition, a multidisciplinary consultation with the involved healthcare professionals was attended. A short report of this day is shown in Appendix 3.

3.3 Interview protocol

The interviews were conducted in a semi-structured way using prepared topic lists. These topic lists were drawn up with contribution and feedback from two rehabilitation physicians, one physician assistant, and an expert in health economics and policy. It was decided to focus the topic lists for DMD patients and their parents entirely on parents, since DMD patients are often young, and the required care and services are organized and requested primarily by their parents. First, the objective of the research was introduced, and the design was explained. Then, parents were asked about the quality of provided care and the problems they experienced. Further, parents were asked about what needs to be organized and requested regarding DMD care and what possible solutions to certain bottlenecks could be. Healthcare professionals were asked about their role in the DMD care pathway, what they need to arrange DMD care and what solutions they envision to overcome certain bottlenecks. More semi-structured questions were prepared for the interviews with the healthcare professionals compared to the questions for parents because it was already known that healthcare providers experience administrative burdens and regulatory pressure. In this way, unbiased experiences and examples were obtained. The topic lists were tested with an initial interview, and after three interviews, minor adjustments were made to the topic lists to improve the coverage of certain items in upcoming interviews. The final topic lists can be found in Appendix 4. For the interviews, parents of DMD patients and healthcare professionals involved in organizing and requesting of DMD care and services were approached by pediatric rehabilitation specialists of the pediatric neurology outpatient clinic at the Radboudumc in Nijmegen since they are involved in the care pathway and have the contacts. When parents and healthcare professionals were willing to participate in an interview, they were contacted via email, and an appointment was scheduled. If the person did not respond to the first email, a reminder was sent after a week. The interviews were conducted in person unless the participant preferred an online interview. All interviews were recorded with a voice recorder.

3.4 Data-analysis

The audio fragments of the interviews were transcribed and reviewed for accuracy by the participants. The transcripts were imported into *Atlas.ti*. and coded using a deductive approach based on the conceptual framework. First, the transcripts of the interviews were analyzed by open coding, identifying the source and size of administrative burdens and regulatory pressure, the mechanisms or causes behind regulatory and administrative burdens, and the possible solutions to reduce the experienced pressures and burdens. After this, the codes were processed in *Microsoft Excel* to create structure in the coded items. During this stage of the analysis, the codes of the different interviews were related to each other. The models of Dahlgren-Whitehead and Levesque were used to categorize the sources and mechanisms or causes of administrative burdens and regulatory pressure around the

DMD care pathway. The codes related to the sources were categorized based on the different layers and components of the Dahlgren-Whitehead model. When a source could not be fitted into the Dahlgren-Whitehead model, an addition was made to the model to include the source. The codes related to the mechanisms or causes were structured based on the Levesque model. First, it was examined between which stages of the demand process the administrative burdens or regulatory pressure were caused. Then, it was examined whether this cause could be assigned to a term in the included dimensions or determinants in the model. When this was not the case, an additional term was formulated to include the causes in the model. The models from Dahlgren-Whitehead and Levesque were used in the same way to analyze the results of the literature review.

3.5 Research quality

The quality of this research was guaranteed in different ways. All the participants of the interviews were closely involved in the DMD care pathway, so they shared relevant information and experiences. The literature review was conducted to confirm the assumption that there is a gap in the literature regarding the administrative burdens and regulatory pressure experienced by patients and their parents.

The validity of this study was ensured in part by the use of method, investigator, and data source triangulation (Carter et al., 2014). In addition, member checking was used, so participants have the opportunity to correct misinterpretations in the transcripts of the interviews and to provide additions. Furthermore, peer debriefing was used to enhance the validity of the study.

The reliability of this study was ensured by the involvement of more than one researcher during each phase of the study. Two rehabilitation physicians, one physician assistant, and an expert in health economics and policy contributed to the preparation of the topic lists. The literature review, interviews, and coding of the interviews were conducted with cooperation of the expert in health economics and policy. Besides, the interviews were recorded so the data could be retrieved.

3.6 Ethical aspects

This study has been assessed for WMO compliance by the METC Oost-Nederland. They determined that no positive assessment from the METC Oost-Nederland or any other recognized medical-ethical review board was required to conduct this research. The confirmation is provided in Appendix 5. The participants of the interviews received an informed consent letter in advance, this letter is shown in Appendix 6.

4. Results

In this study, first, a literature review was conducted and subsequently, 11 participants involved in the DMD care pathway were interviewed. This section will first describe the literature review's main results and then the interviews' results will be discussed.

4.1 Literature review

In total, 427 references were identified in PubMed with the search on May 6, 2022. After the screening on title and abstract, 38 references remained, and these were screened on full text. Of these studies, nine studies were included. The low number of included studies shows that there is a lack of literature regarding administrative burdens and regulatory pressure experienced by patients and parents in general. In figure 3, the number of studies in each stage of the review is shown.

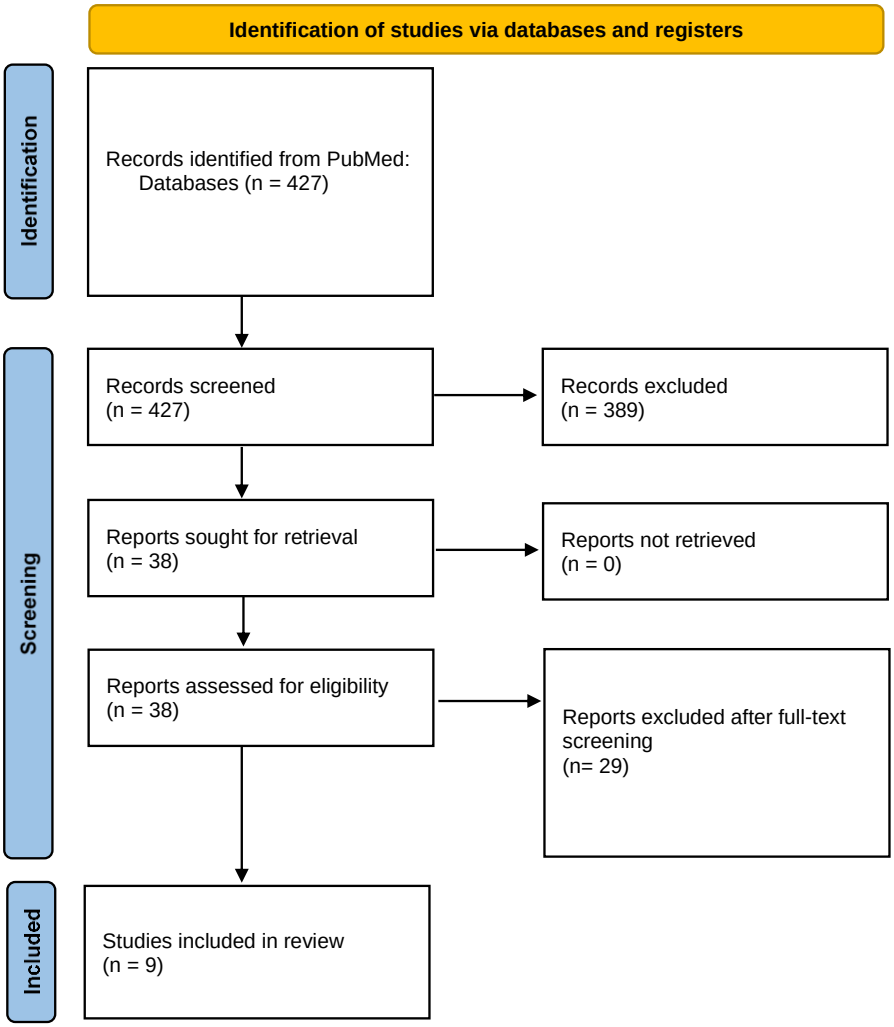

Figure 3| PRISMA 2020 flow diagram for new systematic reviews which included searches of databases and registers only (Page et al., 2021).

Almost all the included studies mentioned that most research about administrative burdens is focused on the problems that healthcare providers experience. This was also why the included studies shifted their scope of focus to patients and parents. For the analysis of the results of the literature review, the same construction was used as for the analysis of the interviews, the following items were extracted from the literature: the type of administrative burden or regulatory pressure, study population, the size of administrative or regulatory burden, the source of administrative burdens or regulatory pressure using the Dahlgren-Whitehead model, the mechanisms or causes behind administrative or regulatory burdens using the Levesque model and the described solutions. Only information about administrative and regulatory burdens experienced by patients and parents was extracted from the included studies since that was the purpose of the literature review. Table 2 provides an overview of the items extracted from the included literature.

Table 2| Extracted items of the included studies of the literature review.

Reference	Administrative burdens or regulatory pressure	Study population	Source (Dahlgren-Whitehead)	Size	Mechanism or cause (Levesque)	Solution
Black et al., 2010	Obtaining medical information and information about available instrumental support	Patients with a chronic disease	Personal characteristics Social and community networks Health care services: - Healthcare professionals	Not mentioned	Between needs and perception / desire - Ability to perceive (health literacy) Between reaching and utilisation - Ability to pay (income)	Patient navigator (PN)[1] giving social support. PN increases the understanding of the individual context of patients while strengthening social support networks for the management of the chronic condition
Henschke, 2012	Provision of assistive technology devices (ATDs) Financing of assistive technology devices (ATDs)	ALS and DMD patients	Personal characteristics Health care services: - Healthcare professionals Social services: - Supplier of ATDs Health insurer	Not mentioned	Between needs and perception / desire - Ability to perceive (health literacy) Between perception / desire and seeking - Ability to seek (information availability) Between utilisation and consequences	A multidisciplinary approach in the provision and financing of ATDs. Case managers[2] should coordinate this

[1] Lay people from the community, a social worker, or medical personnel. Provide transportation, schedule appointments, ensure that medical records are available, and provide social support
[2] Usually a professional. Focuses on coordinating complex, fragmented services to meet patients' needs while controlling costs

					- Appropriateness (adequacy, coordination and continuity, and timeliness)	
Janse et al., 2014	Understandability of provided information Degree to which people know which professional to call Paperwork	Informal caregivers	Health care services: - Healthcare professionals	Not mentioned	Between perception / desire and seeking - Ability to seek (information availability)	Case management. Professionals provide sufficient help with administrative tasks.
Kyle & Frakt, 2021	Appointment scheduling Information seeking Prior authorization[3] Billing issues Insurance premium problems	Nonelderly adults	Personal characteristics Health care services: - Healthcare professionals Health insurer	Not mentioned	Between needs and perception / desire - Ability to perceive (health literacy) Between reaching and utilisation - Ability to pay (income)	Improve measurement of patient administrative burdens to identify opportunities to improve quality, value, equity, and patient experience

[3] A process used by some health insurers in the US to determine whether they will reimburse for a particular treatment or service

Milosevic et al., 2014	Waiting for medical treatments/procedures Changes in administration procedures and admission in hospitals Patient empowerment Chronic disease management Financial costs of treatment	Patients and healthcare professionals	Personal characteristics Health care services: - Healthcare professionals	Not mentioned	Between utilisation and consequences - Appropriateness (adequacy, coordination and continuity and timeliness)	Greater partnership between a healthcare professional and patient associations to overcome the high burdens
Rowan & Shippee, 2016	Treatment approvals Finding information Customer services Paperwork Finding a doctor	People with mental illness	Health care services: - Healthcare professionals	Not mentioned	Between perception / desire and seeking - Ability to seek (information availability)	Efforts by plans to improve healthcare before and after the clinical encounter and by healthcare providers to develop treatments that meet patients' preferences

Sav et al., 2016	Arranging appointments Scheduling visits Arranging medical tests Financial issues	Patients with various chronic conditions	Personal characteristics Health care services: - Healthcare professionals Health insurer	Not mentioned	Between needs and perception / desire - Ability to perceive (health literacy) Between reaching and utilisation - Ability to pay (health insurance)	Health professionals should provide help to manage the burdens of which administrative burdens are a part.
Spencer-Bonilla et al., 2021	Negotiating health care services Affording medications Paperwork	Type 2 diabetes mellitus (T2DM)	Personal characteristics Health care services: - Healthcare professionals	2 hours/day paperwork	Between needs and perception / desire - Ability to perceive (health literacy)	Minimize the number of administrative tasks delegated to patients
Tran et al., 2019	Access to care High costs of care	Patients with a chronic condition	Health care services: - Healthcare professionals	Not mentioned	Between reaching and utilisation - Ability to pay (income, health insurance)	Give every patient an identifiable and accessible member of the treatment team as an entry point in the care system

4.2 Interviews

The main results of the analyzed interviews will be described per item as indicated in the conceptual framework: the size of administrative burdens and regulatory pressure, the source of administrative burdens and regulatory pressure, the mechanisms or causes behind administrative burdens and regulatory pressure, and the mentioned potential solutions to reduce the administrative burdens and regulatory pressure. An overview of the participants in the interviews can be found in table 3.

Table 3| Overview of the participants interviewed. This table shows the main characteristics of the participants.

Type of participant	Gender	Experience
Parent	Female	Patient with DMD aged 20-30
Parent	Female	Patient with DMD aged 10-20
Parent	Female	Patient with DMD aged 10-20
Parent	Female	Patient with DMD aged 10-20
Physician assistant	Female	Working in a center of expertise
Rehabilitation physician	Female	Working in a rehabilitation center
Occupational therapist	Female	Working in a center of expertise. Worked in secondary care rehabilitation
Physiotherapist	Male	Working in a center of expertise
Physiotherapist	Male	Working in a rehabilitation center
Pediatric rehabilitation physician	Male	Working in a rehabilitation center
Occupational therapist	Female	Working in a rehabilitation center

4.2.1 Size of administrative burdens and regulatory pressure

The healthcare professionals mentioned that they spend about half an hour to one and a half hours per patient on administrative and regulatory tasks. These tasks are conducted after direct contact with the patient and include a variety of activities. Indirect patient time is primarily spent on consulting with different stakeholders in the DMD care pathway and includes preparing reports of information, letters, authorizations, referrals, medical statements, and facility requests.

''The whole reporting is indirect patient time. But that also includes consulting with colleagues in and outside the center of expertise. That also means frequent correspondence with all kinds of

stakeholders, such as insurance companies, schools, and municipalities when requesting services.'' – physician assistant in a center of expertise.

The healthcare professionals mentioned that requesting and arranging assistive devices and services is one of the most time-consuming tasks. In most cases, the occupational therapist deals with this task. Applying requires a considerable amount of research, since each supplier of assistive devices, health insurance company, and municipality has different procedures.

''Everyone works differently, and everyone has their procedures. You'll find that out when you have to make a lot of requests. But that always requires some research, for example, whether a request should be sent directly to a supplier or a health insurance company.'' – occupational therapist in a center of expertise.

The interviewed parents of DMD patients mentioned that the administrative and regulatory burdens are very high as they have to arrange a considerable number of things by themselves. A lot of time and often negative energy goes into organizing all the care and services. The physical act of caregiving is perceived as tough, at times, but seems to be dwarfed by the impact of administrative and regulatory burdens.

''Administrative burdens are too high in the whole process. There is just a lot of time wasted on the regulatory pressure, and it often requires negative energy.'' – mother of a DMD patient.

One of the mothers mentioned that she and her husband spend about 25 days a year on organizing and arranging care and services for their son, which is equivalent to all their vacation days in a year. Another mother said that she spends about 4 hours a week on organizing care and facilities for her son. The organization of care and facilities consists of requesting for assistive devices, retrieving information, entering hours, and approving bills with health insurance companies. This is cited as time-consuming. Administrative burdens and regulatory pressure increase at certain moments in the disease process. For example, when school starts again, as a parent you have to get everyone back in the right mode, and when patients become older, they will need more complex care and services.

''You are constantly on the move, when one thing is finally settled, you need either extra care or something is broken again. There are constantly things, it is never finished.'' – mother of a DMD patient.

4.2.2 Source of administrative burdens and regulatory pressure

Figure 4 shows the number of codes per component in the Dahlgren-Whitehead model. The sources of administrative burdens and regulatory pressure for each component will be described below.

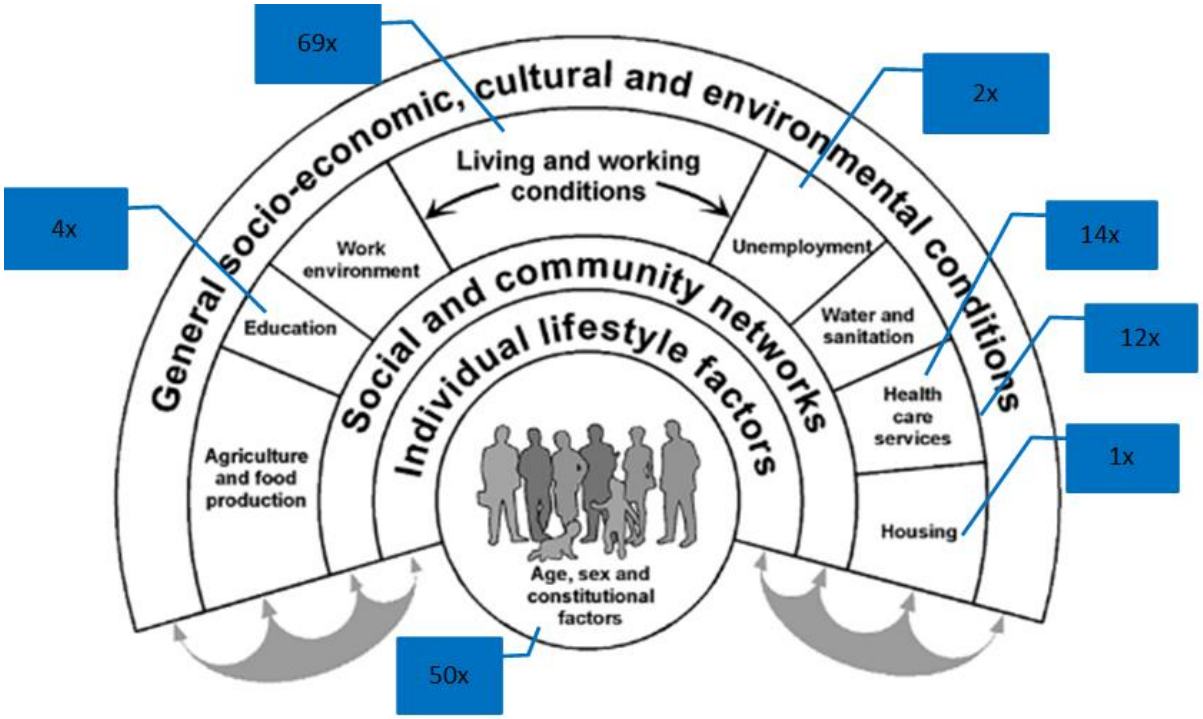

Figure 4| Overview of the numbers of codes identified for each component of the Dahlgren-Whitehead model.

Personal characteristics

The first source of administrative burdens and regulatory pressure can be attributed to the personal characteristics of patients but especially parents in the Dahlgren-Whitehead model. In total, this was mentioned 50 times as a source. Proactive parents get more things done compared to parents who are more wait-and-see or less daring. Less active or more timid parents may face more administrative and regulatory burdens in the healthcare system. This may lead to unwanted differences in care use between patients.

''You have people who are empowered. People who know what they want, they get what they want, or get very far. People who are less empowered and do not commit to anything just do not get what they need. You just have to keep actively pursuing everything. If you want something done you just have to call, email, and if you do not get a response, call, or email again.'' – mother of a DMD patient with a role in patient advocacy at the DPP.

''Parents who are very involved and active and have a good contact with municipalities need us much less than parents who do not really know or cannot keep the overview or are just overloaded in the care pathway''. - occupational therapist in a center of expertise.

On the other hand, proactive parents sometimes want to arrange so much by themselves that they experience high administrative and regulatory burdens. Moreover, these proactive parents may be arranging things that are not necessarily beneficial to the patient.

''On the other hand, if we are not involved, we sometimes encounter parents who have already arranged all kinds of things like a wheelchair and a wheelchair bus. Then they arrive with a wheelchair that is not appropriate for the size or demand for which it is intended.'' - occupational therapist in center of expertise.

Another determinant in experiencing administrative burdens and regulatory pressure attributable to the personal characteristics component is the education level of the patient's parents. Highly educated parents who are highly educated have more capabilities to think about possibilities and offer solutions and parents who are less educated, for example, are more seeking whom they can approach for certain issues. Higher educated people can more easily find information, take in the information, and use it to navigate through the care pathway, resulting in more and better care for their child.

''Parents who speak the language and have the networks and connections just get a lot of things done and get a lot of services and opportunities. Someone who does not have that also does not know what is possible.'' – occupational therapist in a rehabilitation center.

Another important factor is the parents' financial status. Patients with parents who have sufficient resources receive more and better care than others. In addition, long application processes can be bypassed by purchasing assistive devices themselves.

''If I need something, I buy it. I have the resources to buy it and that does make a big difference. But it is easy for me to say because we have our financial resources to get things done. If we were struggling with our finances, we could not do these kinds of things.'' – mother of a DMD patient.

<u>Social services</u>

A large number of quotes were related to social services, which was initially missing from the Dahlgren-Whitehead model. We added the category as part of the living and working conditions layer, since social services determine access and opportunities related to housing health care, and education. Municipalities and suppliers of assistive devices are covered by this part of the model. In total, municipalities were mentioned 46 times as a source of administrative burdens and regulatory pressure. Healthcare professionals, patients, and parents experience many administrative burdens and regulatory pressure around requesting assistive devices and home modifications under the Social Support Act. The request processes are long and complicated and there is a lack of knowledge within municipalities.

''If there is a wheelchair that needs to be replaced, I have to apply to the Social Support Act and write a whole report on why the wheelchair needs to be changed. Then you also have to substantiate

why a new chair is needed. You are entitled to a new chair after 5 years, but DMD patients do not always complete that period, they often need a new chair sooner.'' – occupational therapist in a rehabilitation center.

''I think we have spent 3 to 4 years working with our municipality to make the necessary adjustments to our house. We had a complete advice report supported by both occupational therapists from the rehabilitation center and a therapist from the center of expertise and that plan was rejected by the municipality as ''a wish of the parents''.'' – mother of a DMD patient.

Other administrative burdens and regulatory pressure related to municipalities are caused by poor cooperation with and frequent change of suppliers, lack of responsibility, unnecessary repetition of requests, no regular contact person, and poor accessibility.

''It is different every 3 years. We were first with [Name of supplier of assistive devices] in Nijmegen. Then after 2.5 years, it was settled and then they switched to [Name of supplier of assistive devices]. Now we have been with [Name of supplier of assistive devices] for 2 years, but I expect that in a year they will choose another supplier who will offer it cheaper. It is always about competition in healthcare. Why should there be competition in healthcare if all patients should be allowed to receive the same care?'' – occupational therapist in a center of expertise.

''At the municipality, you also have varying policies. One municipality comes to your home and determines what is needed and the other municipality only makes a phone call, and you have to deal with it. Yes, that care just changes every time.'' - occupational therapist in a center of expertise.

During the interviews, suppliers of assistive devices were mentioned 23 times as a major source of administrative burdens and regulatory pressure. In most of the cases, it was about extensive procedures for requests for certain assistive devices.

''If you are talking about a complex electric wheelchair, it can sometimes take up to six months.'' – physiotherapist in a rehabilitation center.

Other administrative burdens and regulatory pressure related to suppliers of assistive devices are caused by a lack of knowledge, lack of a regular contact person, unnecessary (annual) repetition of requests, poor cooperation with the municipality, and defective or no deliveries of assistive devices.

<u>Health care services</u>

Healthcare providers in primary and secondary care were mentioned 13 times as a source of administrative burdens and regulatory pressure. In most of these cases, it was about the lack of

knowledge or failure to adopt knowledge from the centers of expertise. As a result, processes for certain requests run slowly and laboriously.

''Despite all the reports from the Radboud center of expertise that we got and could show, they still wanted to judge for themselves.'' – mother of a DMD patient.

Moreover, there are differences between healthcare providers in rehabilitation centers in secondary care. One rehabilitation center has more expertise than another center, allowing for faster procedures and more appropriate devices can be delivered.

''This also varies from center to center, the more expertise there is, the easier it goes.'' – physiotherapist in a center of expertise.

Centers of expertise were mentioned once as a source of administrative burdens and regulatory pressure. The issue here is the laborious transmission of knowledge to primary and secondary care, leaving patients and parents to deal with this transfer.

<u>Health insurer</u>

A part of the quotes involved health insurers, which was initially missing from the Dahlgren-Whitehead model. We added this category between the health care services component and the general socio-economic, cultural, and environmental conditions layer since the health insurer plays an overarching role in the provision of care. In total, health insurance companies were identified 12 times as a source of administrative burdens and regulatory pressure. The burdens and pressure experienced here are mainly in the area of reimbursements, paperwork, lack of a regular contact person, rejection of requests, and unnecessary (annual) repetition of requests.

''Sometimes you have to write a referral for physiotherapy every year to the health insurer even though patients are entitled to chronic physiotherapy, and it is a condition that cannot be cured so I always think it is a bit unnecessary to write a statement every time.'' - rehabilitation physician in a rehabilitation center.

In addition, there are many differences between health insurance companies; some approve requests more easily and faster than others. This results in a difference in burdens experienced per health insurer.

''I always advise patients to look carefully at which package they take out for the next year. Experience shows that with one health insurance company, you will not be reimbursed for an arm

support device in November and with another, you will.'' - occupational therapist in a center of expertise.

Education

Schools were barely mentioned as a source of administrative burdens and regulatory pressure, they were only named four times. Finding a suitable high school and the adaptability of high schools and vocational colleges were mentioned here. In general, few problems were experienced regarding education.

Unemployment

The search for work was mentioned twice as a source of administrative burden and regulatory pressure, Few DMD patients work, and this is often because of all the regulations that exist. Arranging (voluntary) work and strict conditions around the various funding streams are barriers experienced here.

''We recently had a man who could work well, but because no taxi transport could be arranged, he still sits at home all day.'' - rehabilitation physician in a rehabilitation center.

Housing

Housing was mentioned once as a source of administrative burdens and regulatory pressure. It is difficult to find suitable housing for adult DMD patients because patients with physical disabilities often end up living in the same place as mentally disabled people.

''If those boys want to live independently then there are also those problems like finding good housing for those boys. It is very often the case that a physical disability is taken together with mentally disabled people and then you end up in such a group.'' – mother of a DMD patient.

4.2.3 Mechanisms of causes behind administrative burdens and regulatory pressure

Figure 5 shows the number of codes per component in Levesque's model. The mechanisms or causes of administrative burdens and regulatory pressure for each stage will be described below.

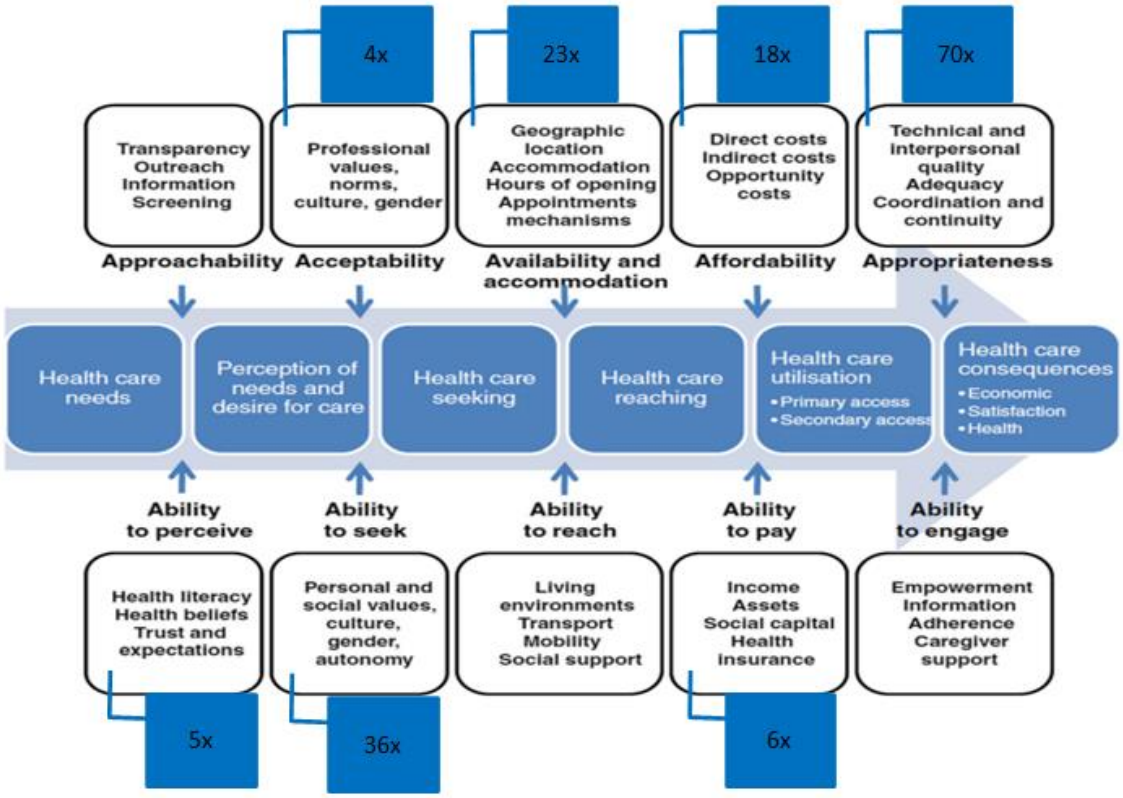

Figure 5| Overview of the numbers of codes identified for each stage of Levesque's model.

Between health care needs and perception of needs and desire for care

The mechanism behind administrative burdens and regulatory pressure is attributed five times between the stage of health care needs and perception of needs and desire for care in the healthcare demand process of Levesque. In these cases, burdens are caused by the ability of parents or patients to perceive. The codes associated are attributable to the term health literacy in Levesque's model.

''You notice that families from a lower social class have a bit more difficulty to arrange things, but also in accessing substantive healthcare information.'' – mother of a DMD patient.

Between perception of needs and desire for care and health care seeking

In total, 40 codes are attributable between the stages of perception of needs and desire for care and health care seeking. Some of the burdens and pressures mentioned here are caused by patients' and parents' ability to seek. A part of this can be assigned to the term personal and social values from Levesque's model. Some parents send their child to a mytyl school, which means they experience less

administrative burdens and regulatory pressure because many examinations and applications are done during school hours. Moreover, parents of DMD patients tend to be very protective and find it difficult to give their children more responsibility at a certain point regarding the organization and application of care and services. As a result, the burdens and pressure on parents remain high. Another part is covered by a self-added term to Levesque's model: capabilities. As explained above, more active parents or patients get more things done in a faster way than passive or timid parents or patients.

''There are parents who are all over it and parents who are more relaxed about it. Being clear does make a difference. And if you are too timid then it might just be more difficult to get things reimbursed.'' – physiotherapist in a rehabilitation center.

Coping is another self-added term to Levesque's model between these stages. This covers the burdens caused by parents in particular struggling to face and accept their child's decline. Parents often need some time to accept.

''An acceptance that someone is constantly deteriorating, so continuous confrontation. So, the more assistive devices are needed, the more a person deteriorates. Parents want to deny that for a very long time.'' - occupation therapist in a rehabilitation center.

A small proportion of the codes is covered by the dimension acceptability of Levesque's model. These codes can be attributed to a self-added term to the model: information availability. Healthcare professionals have an important role to play in providing information. Administrative and regulatory burdens on patients and parents can be caused by not knowing the possibilities regarding work for patients and where to request what kind of things.

''There is a resource guide from Vilans[4]. There you can choose all sorts of things and then you are helped with which aids are best or may be appropriate. It also indicates, for example, when it is the right time to use an occupational therapist or physiotherapist. So all kinds of resources have been devised to help parents and patients. But you have to be able to find it, but I think for a lot of parents it's unfamiliar territory.'' – occupational therapist in a center of expertise.

<u>Between health care seeking and health care reaching</u>

There are a total of 23 codes assignable between the health care seeking and health care reaching phases. All these codes are covered by the dimension availability and accommodation in Levesque's model. Some causes of administrative burdens and regulatory pressure can be attributed, for

[4] A national knowledge organization for care and support

example, to the geographic locations of rehabilitation centers. When people live too far away from a rehabilitation center, organizing care, requesting assistive devices, and finding caregivers, for example, is a lot more difficult.

''We are not affiliated with a rehabilitation center. This means that we are the point of contact for assistive devices ourselves. It is always a quest to find the right one and that takes a lot of time and patience, but also a lot of phone calls and gathering knowledge.'' – mother of a DMD patient.

''As a local, you can get into trouble. You try to organize everything the best you can, but you get screwed because you organize it locally. Whereas, in my opinion, that is cheaper than through a rehabilitation center.'' – mother of a DMD patient.

Other causes in this stage of the care demand process are covered by a self-added term: stakeholder accessibility. Throughout the care pathway, the lack of contacts, lack of a regular point of contact, no responses, and poor reachability of stakeholders are cited as causes of administrative and regulatory burdens experienced by patients and parents.

''I have a different person on the phone every time.'' – mother of a DMD patient.

<u>Between health care reaching and health care utilisation</u>

In total, 24 codes are assignable between the stages of health care reaching and health care utilisation. A part of the quotes is attributable to the ability to pay for patients and parents. These quotes can be covered by the terms income and health insurance in Levesque's model. People who have a good income or good insurance can use more resources to unburden themselves.

''We have a good Long-Term care act indication so I can buy some care and services that will take some of the burden off myself and my husband.'' – mother of a DMD patient.

The other part of the codes in this area of the care demand process is attributable to the dimension affordability and can be covered by the self-added term reimbursement restrictions. The establishment of reimbursements for care and services is a major cause of administrative burdens and regulatory pressure experienced by healthcare professionals, patients, and parents. Also, repetitive submission of applications weighs as a heavy burden.

''From our point of view as practitioners, it is quite logical that a certain diagnosis simply requires long-term provision, but the municipality sometimes gets stuck with their rules. This is not always appropriate, I think.'' – physiotherapist in a rehabilitation center.

''For instance, I regularly have to make a follow-up authorization while I think that the concept does not change due to growth. In my opinion, it can be done without my intervention.'' - pediatric rehabilitation physician in a rehabilitation center.

Between health care utilisation and health care consequences

The vast majority of all the codes is assignable between these stages in the care demand process, 70 codes are located in this section of Levesque's model. All these codes are attributable to the dimension appropriateness. A part of the codes is covered by the terms adequacy and coordination and continuity in Levesque's model. The lack of knowledge and responsibility, difficult cooperation and information transfer between stakeholders, annoying mistakes, and moderate empathy and adaptability of different stakeholders in the DMD care pathway are causes of administrative and regulatory burdens for patients and parents.

The other codes can be covered by a self-added term to Levesque's model: timeliness. The extensive processing of applications for care and services is a major cause of perceived burdens.

4.2.4 Solutions to reduce administrative burdens and regulatory pressure

During the interviews, various areas for improvement and possible solutions to the high administrative burdens and regulatory pressure were given. First, healthcare professionals advocate for a shared patient record. Healthcare professionals in the different categories of care use different electronic patient records requiring additional letters and consultations between healthcare professionals in the different categories of care when transferring information. The same applies to the statements that need to be written. It would save a lot of work if a statement could be used multiple times by multiple agencies. A health insurance company, for example, should know that DMD is a chronic disorder and should not request a new statement every time. A statement should also be transferable from a previous health insurance company.

''The moment the health insurer receives a statement, they should just consider that as chronic and not ask for a statement again.'' – rehabilitation physician in a rehabilitation center.

Another frequently mentioned area for improvement is the collaboration and communication between the various stakeholders in the DMD care pathway. A patient can be visiting a rehabilitation center, center of expertise, and, if required, the CTB at the same time. This is fragmented throughout the care pathway and it is not always clear to all the different involved healthcare professionals when a patient has an appointment with another care provider. The collaboration and communication between these centers can be improved. In addition, the transfer of knowledge between a center of

expertise and healthcare professionals in primary care and rehabilitation centers can be improved. The center of expertise has the most experience with DMD patients, while primary care and rehabilitation centers are visited by fewer of these patients. Centers of expertise should be able to support primary care providers and rehabilitation centers, which can be improved by enabling and stimulating direct caregiver communication, e.g. through telephone or videoconferencing. Furthermore, it was indicated several times that the cooperation between municipalities and suppliers is dramatic. Because of this, patients or parents often have to mail and call, and application processes take more time.

''The cooperation between agencies such as [Name of supplier of assistive devices] and the municipality is literally to weep.'' **–** mother of a DMD patient.

Parents would also like to have a regular contact person at suppliers of assistive devices and municipalities. At some suppliers like company 1, it is not possible to get in direct contact with an advisor. Contact has to be made through a helpdesk, but the people there do not know what you are calling or emailing for. At some municipalities, there is no permanent consultant which is frustrating for patients and parents. This results in many calls, emails, and long, complicated procedures while services for DMD patients should be arranged quickly since a DMD patient deteriorates every day. Another solution that may reduce this problem is an expedited process for applying for assistive devices and services for patients with muscular diseases in general.

'' In my opinion, there should be an accelerated pathway for basically anyone with a muscle disease.'' – occupational therapist in a rehabilitation center.

The provision of information to DMD patients and parents can be improved. For example, they should be informed at an early stage about the tools that are available and where they can be requested. An overview with clear requirements that specific assistive devices must meet would also help patients and parents. In addition, it was proposed to develop a module with all information about DMD, so patients and parents do not have to explain it everywhere. A map with all available assistive devices could also contribute to reducing the workload of searching for patients and parents.

Since a large proportion of the perceived administrative burdens and regulatory pressure originates from municipalities, particularly in the area of applying for assistive devices and housing arrangements, there is a great area for improvement for the municipalities. First, the people who review applications should have knowledge about the progression of the disease. In addition, it would be easier if municipalities do not constantly change their suppliers. Also, changes could be made to

the Social Support Act. Currently, applications for services for young adults with complex disabilities are placed on the same pile as, for example, applications from elderly for domestic help.

''The majority of the Social Support Act use is attributable to the aging population so take the groups apart because that is just a different line of work. A request for domestic help because you are 80 should not be one the same pile as a request for young adult disability.'' – mother of a DMD patient.

One of the most frequently mentioned solutions by healthcare professionals and parents to reduce the high administrative burdens and regulatory pressure experienced by DMD patients and parent is the introduction of a care coordinator. This coordinator could keep an overview between all stakeholders and arrange care and requests. This person should then be the central node in the network and maintain short lines of communication with all the stakeholders in the DMD care pathway.

''What might help is a coordinator of care. So, someone who not only oversees the care side, but also the regulatory work such as contact with municipalities. So, someone who supervises the whole picture'' - rehabilitation physician in a rehabilitation center.

Parents also advocate for a central point where you can make requests. There, they would consider where the request should go and whether it should be financed through the personal healthcare budget, the Social Support Act, or a health insurance company.

''A point where you have the personal healthcare budget, Social Support Act, and the health insurance. A central point where you can make your request and they just figure out where that request should go. So that you as a parent do not have to figure out where a request should go.'' – mother of DMD patient.

5. Discussion and recommendations

The purpose of this study was to identify the extent of administrative burdens and regulatory pressure, the sources of the burdens, the mechanisms or causes behind these burdens, and the solutions to reduce these burdens and pressure.

From the results, it can be noticed that healthcare professionals do experience some administrative burdens and regulatory pressure but this is considerably less compared to the parents of DMD patients. When considering the sources of administrative and regulatory burdens using the Dahlgren-Whitehead model, it appears that personal characteristics, municipalities, and suppliers of assistive devices (social services), health insurers, and healthcare professionals (health care services) are the most frequently named sources. This was evident from both the interviews and the literature review. Multiple components were added to the Dahlgren-Whitehead model to accommodate all mentioned sources of administrative and regulatory burdens. The Dahlgren-Whitehead model originated in the US and there the system is organized differently than in the Netherlands, which may be an explanatory factor why municipalities, suppliers of assistive devices, and health insurers are not included in the original model. It should be noted that no firm conclusions can be drawn from the number of times a source was mentioned since no quantitative survey was conducted. When using Levesque's model to determine the mechanisms or causes behind perceived administrative burdens and regulatory pressure, the interviews reveal that the mechanisms or causes of the burdens early in the care demand process arise from parents' and parents' ability to perceive, but especially the ability to seek. Later in the care demand process, the mechanisms or causes are mainly because of failure of care systems. Many of the causes can be attributed to the dimension appropriateness. Similarly, no firm conclusions can be drawn from the number of times a mechanism or cause was identified since no quantitative survey was conducted. Most of the literature that emerged from the literature review cited health literacy, income, information availability, adequacy of stakeholders, timeliness of care requests, and coordination and continuity of the care pathway as causes of administrative and regulatory burdens. (table 2). In addition, we found that Levesque's conceptual framework of access to health is not complete. Several terms have been added to this model to fit all determinants of administrative burdens and regulatory pressure into the model and create a more complete picture.

Strengths and weaknesses

The quality, validity, and reliability are ensured in this study. Several methods were used, including a literature review and interviews with people closely involved in the DMD care pathway. The interviews were recorded, participants of the interviews had the opportunity to correct misinterpretations, and more than one researcher was involved in each phase of the study. However, this study has also

several limitations. Due to time constraints, only 11 interviews were conducted, of which only four were with parents of DMD patients. Because of this, the data is limited. In addition, only mothers of DMD patients participated in the interviews since the fathers were often at work at the time of the interviews. Besides, only healthcare professionals from secondary and tertiary care were interviewed during this study, and no other stakeholders associated with the DMD care pathway. However, during the last interviews, many of the same problems were raised so that may be a sign of data saturation.

Future research could interview more healthcare professionals and parents. In doing so, primary care healthcare professionals can share their experiences, and interviewing parents not seen by a center of expertise could also add value to a future study. In addition, it can be very valuable to engage with the stakeholders identified as a source and cause of perceived administrative burdens and regulatory pressure. It would be useful to discuss with municipalities and suppliers to obtain their side of the story. It will also be valuable to talk with the Association of Netherlands Municipalities to identify what they need to direct municipalities to make the application process more efficient. With this, a more complete picture can be made of the causes and possible solutions regarding administrative and regulatory burdens.

Recommendations

To reduce administrative and regulatory burdens for healthcare professionals, a shared electronic patient record could provide a solution (Kruse et al., 2018). This avoids duplication and repetition of the same questions for patients. With a shared electronic health record, all healthcare professionals and organizations involved could be notified. However, this is difficult to achieve because there are strict laws and regulations to ensure patient privacy. A personal health environment (PGO) may be a way to share information among all stakeholders. Through this system, the patient or parent can specify which information should be shared with which healthcare professional (Veldman, 2019). With complex care, such as DMD, it might help to be able to share this information also with suppliers of assistive devices, health insurers, and other involved organizations. This could both reduce administrative and regulatory burdens for healthcare professionals as for patients and parents since one does not have to continuously call, email, or write to transfer information between different stakeholders.

Meanwhile, a motion was passed in the House of Representatives of The Netherlands opposing the (annual) re-indication of chronically ill patients. The government should discuss with municipalities and health insurers to extend the indication for these conditions and in some cases make it lifelong (Tweede-Kamer, 2022). This could reduce the perceived burdens and pressure on healthcare professionals, patients, and parents.

For patients and parents, administrative burdens and pressures could be reduced if there are regular contacts at municipalities and suppliers of assistive devices who are easily accessible. In case there is a new contact person at the municipality or supplier, there should be a proper transfer of information, a PGO could contribute to this as well. In addition, these contacts should have sufficient knowledge about the condition. This could be achieved by informing suppliers and municipalities at an early stage about disease progression and the needs involved. This information should ideally be provided by the government as this should be a nationwide improvement.

During the interviews, parents expressed a need for more information provision since they now spend a lot of time searching for and figuring out the possibilities regarding care and facilities. For example, parents requested several times for a manual listing all possible assistive devices for DMD care. However, a tool has already been developed on behalf of the Ministry of Health, Welfare, and Sport where all the assistive devices can be found. This tool is called the Vilans resource guide. Since some of the parents are not familiar with this guide, there is a role for centers of expertise and rehabilitation centers to provide patients and parents with such information. At an early stage of the disease, these centers should help patients and especially parents on their way to obtain the necessary information.

The results show that the administrative burdens and regulatory pressure regarding the application of assistive devices are partly due to the long and complicated processes at municipalities, health insurers, and suppliers. In 2015, there were already advocates for an improved process for requesting facilities for amyotrophic lateral sclerosis (ALS). Because, like ALS, DMD is a progressive disease, and assistive devices must be provided on time so that they are appropriate, and patients are not constantly catching up. Therefore, it was suggested that a national protocol should be established and implemented to make the application process for progressive diseases more efficient (Das et al., 2015). This task was assigned to municipalities but today there are still municipalities that can not manage to provide facilities for progressive diseases on time (VWS, 2022). The government has agreed with the Association of Netherlands Municipalities that the latter supervises municipalities on the provisioning process, but it remains to be seen if and how soon things will improve.

The interviews show that the medical care around DMD is well organized. The medical side is concentrated with several centers of expertise throughout the country that were mostly praised by parents. Knowledge transfer to primary and secondary care does need to be improved to ensure high-quality local care as well. Complex social care is underserved throughout the care pathway. During the interviews and in several studies included in the literature review, the potential role of a coordinator to achieve burden and pressure reduction was mentioned. Since the administrative burdens and regulatory pressure are so high for patients and parents, it should be viable to offer all families a

coordinator whom they can approach with all questions regarding DMD care. This coordinator should need extensive knowledge of the disease and the healthcare system to bring the questions and applications to the right person. The burdens on patients and parents will thereby decrease significantly. This will give parents more time and energy to focus on the physical care of their child instead of all the regulation around it. Such a case manager or coordinator has also been introduced for people with Alzheimer's disease and post-COVID patients (Radboudumc, 2022). This professional has usually a paramedic background such as a physiotherapist or occupational therapist. A case manager or coordinator should be able to take administrative and regulatory burdens off parents' shoulders when they need this. An important benefit of a case manager is equalization, patients receive similar care, irrespective of capabilities and socioeconomic position. However, some reservations apply when opting for a coordinator or case manager. Adding a layer between patients and healthcare professionals could also result in additional burdens and communication. Specifically, since medical care is perceived as organized relatively well, it could be more efficient to focus the solution on streamlining municipal provision of assistive devices.

A promising approach to reduce the administrative and regulatory burdens experienced by healthcare professionals, patients, and parents is to centralize complex social care. A national center that patients and parents can contact with all questions and requests could improve appropriateness and timeliness of provision of assistive devices. Case- specific knowledge would be easier and more efficient to acquire. This solution would apply to complex diseases that require specific knowledge in combination with very low annual patient numbers in municipalities. Besides DMD, other complex rare diseases may apply. Similar centers have been introduced for the aftercare of Q fever and post-COVID (de Koster, 2019). The House of Representatives of The Netherlands has already passed a motion urging the government to investigate what would be needed to introduce a central point of contact for applications with personal healthcare budgets as soon as possible (Tweede-Kamer, 2022). This would be a useful step in reducing the administrative and regulatory burdens, but we believe the problem should be addressed more broadly by also regulating all DMD applications around the Social Support Act nationwide.

Healthcare professionals, patients, and parents experience heavy administrative and regulatory burdens regarding DMD care. We found several sources of administrative and regulatory burdens, of which provision of medical assistive devices by suppliers and municipalities was mentioned most frequently. The most frequently cited mechanisms or causes behind administrative and regulatory burdens were adequacy of stakeholders, timeliness of care request, and coordination and continuity of the care pathway. In summary, we advocate the centralization of DMD applications around the Social Support Act. The progression of DMD is obvious so it is known when a patient needs certain

assistive devices or services. In our opinion, the administrative and regulatory burdens can be most efficiently reduced by centralizing the applications via the Social Support Act. In addition, differences in patients or parents capabilities to coordinate care can be a source of administrative and regulatory burdens as well as introduce differences in care use; in these cases, a case manager could offer solutions to provide support to patients and parents navigating through the care pathway.

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7. Appendices

7.1 Appendix 1: Overview of the stakeholder in the DMD care pathway

Table 1| Overview of the stakeholders involved in the DMD care pathway. This table is constructed using the guidelines for the treatment of DMD (de Coo et al., 2016; Federatie-Medisch-Specialisten, 2021).

Healthcare provider	Type of care	Frequency of use	Category of care	Financed
General practitioner	Support and guidance in patient's home environment	On demand	Primary care	Health Insurance Act
Pediatric neurologist	Diagnosis, Corticosteroid treatment, Monitoring blood count, glucose, and adverse events	At the start of the treatment, 4x in the first year, then 2x a year (glucose control 1x a year)	Secondary or Tertiary care	Health Insurance Act
Pediatric physician	Guidance and treatment of acute complaints not specifically related to DMD	On demand	Secondary or Tertiary care	Health Insurance Act
Clinical geneticist	Diagnosis, also test mother and siblings of patient	1x	Secondary care	Health Insurance Act
Radiologist	Diagnosis, Monitor bone quality, and development of deformities	1x a year	Secondary or Tertiary care	Health Insurance Act
Ophthalmologist	Eye check	On demand	Secondary care	Health Insurance Act
Dentist / Orthodontist	Correcting teeth and jaw defects or abnormalities	On demand	Secondary care	Health Insurance Act
Pulmonologist	Pulmonary function test and cough support	1x a year	Secondary or Tertiary care	Health Insurance Act
CTB	Non-invasive (nocturnal) ventilation	On demand, when involved 1x a year. Close contact. When this is started, patient visits pediatric neurologist 1x a year	Tertiary care	Health Insurance Act

Cardiologist	Measure blood pressure ECG and Echocardiography	Each control 1x per 2 years, after 10 years 1x a year	Secondary or Tertiary care	Health Insurance Act
Orthopedist	Correction of scoliosis, Treatment of spine fractures and contractures	On demand. Yearly control	Secondary care	Health Insurance Act
Endocrinologist	Analysis of problems with growth and puberty	On demand	Secondary care	Health Insurance Act
Rehabilitation physician	Monitoring body function and structures, activities and, participation related to disease progression. Establishes therapeutic plan and goals and considers which assistive devices are needed	At least 2x a year	Secondary or Tertiary care	Health Insurance Act
Occupational therapist	Advices and trains patients to perform daily activities. Requesting and fitting of assistive devices	At least 2x a year	Primary, Secondary, or Tertiary care	Health Insurance Act
Physiotherapist	Maintain functions, conditions, and coughing techniques. Can collaborate with occupational therapist	At least 2x a year	Primary, Secondary or Tertiary care	Health Insurance Act
Dietician	Monitor intake of adequate calcium and Vitamin D to prevent osteoporosis, and monitor weight loss/gain. Evaluation of nutrition	1x a year	Secondary or Tertiary care	Health Insurance Act
Psychologist	Focus on coping. Support patient and family with acceptance of diagnosis. Analysis and diagnosis of	On demand	Primary or Secondary care	Health Insurance Act

	behavioral and/or learning problems			
Social worker	Focus on coping. Support family with acceptance of diagnosis	On demand	Primary or Secondary care	Social Insurance Act
Nursing specialist	Can serve as point of contact for patient, family, and care providers	On demand	Secondary care	Health Insurance Act
Nurse	Take care in hospital or home care in specific situations	In case of hospitalization	Secondary care	Health Insurance Act
Pharmacist	Provides the prescribed medications	On demand	Primary care	Health Insurance Act
Gastrointestinal liver doctor	Analysis/diagnosis of problems with the stomach, intestines, and liver	On demand	Secondary care	Health Insurance Act
Urologist	Remedies urinary tract problems	On demand	Secondary care	Health Insurance Act
Speech therapist	Examine level of language comprehension, language production, and intelligibility. Monitoring of oral motor skills and chewing and swallowing function	On demand	Secondary or Tertiary care	Health Insurance Act
Home care / district nurse	Assistance in daily living (especially in adult patients)	On demand	Primary care	Health Insurance Act, Social Insurance Act, or Long-term Care Act
Municipality	Handling applications for home modifications and assistive devices	Upon need	-	-

Supplier of assistive devices	Provides the necessary assistive devices	Upon need	-	-
Health insurance provider	Involved in what types of care are and are not reimbursed and what law reimbursed the care.	At all times through the care pathway	-	-
(Mytyl)School/ Sports club / Employer	Provide appropriate education, sports, and work for DMD patients	At all times through the care pathway	-	-
Dutch Parent Project	Foundation dedicated to more and better research on DMD and brings DMD families together	Upon need	-	-
Spierziekten Nederland	Brings families and people with DMD together to share experiences.	Upon need	-	-

7.2 Appendix 2: Search string

PubMed

Population filter

("Caregivers"[MeSH Terms] OR family caregiver* [Title/Abstract] OR Patient* [Title/Abstract] OR family carer* [Title/Abstract])

Outcome filter

(Administrative task* [Title/Abstract] OR administrative burden* [Title/Abstract] OR administrative problem* [Title/Abstract] OR administrative time* [Title/Abstract] OR administrative work* [Title/Abstract] OR bureaucratic task* [Title/Abstract] OR bureaucratic burden* [Title/Abstract] OR bureaucratic problem* [Title/Abstract])

Study type filter

(''Surveys and Questionnaires''[Mesh] OR ''Focus Groups''[Mesh] OR ''Interviews as Topic''[Mesh] OR ''Interview'' [Publication Type] OR Interview* [Title/Abstract] OR survey* [Title/Abstract] OR questionnaire* [Title/Abstract] OR focus group* [Title/Abstract])

7.3 Appendix 3: Report observational research

Meeloopdag multidisciplinaire polikliniek Duchenne spierdystrofie in het Radboudumc

Op 11 april 2022 heb ik een dag meegelopen op de poli voor kinderen met Duchenne spierdystrofie in het Radboudumc. Het doel van deze poli is om kinderen jaarlijks op basis van multidisciplinair- en functieonderzoek, advies te geven en vragen over klachten en problemen te beantwoorden. Tijdens een dag op deze poli wordt de patiënt onderzocht door onder andere de volgende zorgprofessionals: een kinderneuroloog, kinderrevalidatiearts, fysiotherapeut, ergotherapeut, logopedist en diëtist. Daarnaast worden er meestal, afhankelijk van de leeftijd en het stadium van de ziekte, onderzoeken uitgevoerd door een kinderorthopeed. cardioloog, longarts en een radioloog. Over de hele dag wordt er informatie verzameld rond het ziekteproces van de patiënt. Deze informatie wordt aan het eind van de dag doorgesproken door de betrokken zorgprofessionals en vervolgens worden de patiënt en ouders bijgepraat over de resultaten van de verschillende onderzoeken. Daarnaast wordt er advies gegeven over de volgende stappen. Dit laatste gaat allemaal in samenspraak met de patiënt en de ouders.

Op deze dag ben ik aanwezig geweest bij twee consulten met een kinderrevalidatiearts en twee patiënten met ouders. Vervolgens ben ik bij het multidisciplinaire overleg geweest waar de bevindingen door de betrokken zorgprofessionals werden besproken.

<u>Consult 1</u>

Tijdens dit consult kwam er een patiënt binnen die de professionals een ‘’goede Duchenne’’ noemen omdat hij bijvoorbeeld nog steeds (kleine) stukjes kan lopen, iets wat bij patiënten van zijn leeftijd maar weinig voorkomt. De patiënt liet vanaf het begin van het consult al merken dat hij er weinig zin in had. Zo zat de patiënt het gehele consult onderuitgezakt op zijn telefoon een spelletje te spelen en gaf hij op alle vragen van de kinderrevalidatiearts korte antwoorden. De patiënt gaf aan dat alles goed ging en dat hij nergens last van had. Hij merkte weinig tot geen achteruitgang ten opzichte van een jaar eerder.

De ouders van de patiënt lieten weten dat ze de nabespreking liever online via een videocall op een ander moment plaats zouden willen laten vinden aangezien de energie wel op aan het raken was. Ze waren, na een lange reis, al vanaf 8:30 op locatie en normaal zou de nabespreking tot ongeveer 16:00 duren. De patiënt was voorafgaand aan dit consult bij de cardioloog en de paramedici (fysio- en ergotherapeut) geweest. Hier had hij meerdere tests ondergaan waaronder de 6 minuten looptest bij de paramedici. Toen de kinderrevalidatiearts vroeg of de patiënt een stukje wilde lopen, weigerde hij dat omdat hij dat net ook al gedaan had. Tijdens het consult werden verder vragen

gesteld over school, slapen, medicijnen, puberteit en dagelijkse handelingen en de hulp die de patiënt krijgt. De patiënt vertelde nog niks te merken van de puberteit. De ouders van de patiënt merken aan het gedrag dat de puberteit wel gestart is. Bij een kort lichamelijk onderzoek werd er gekeken naar de voeten, benen en rug van de patiënt. De rechterknie leek niet helemaal gestrekt te kunnen worden bij staan.

De patiënt ging er na het consult direct vandoor terwijl de ouders nog even bleven praten. Zo gaven de ouders van de patiënt aan of er in de toekomst tussen de afspraak met de paramedici en het consult met de revalidatiearts wat tijd zou kunnen zitten omdat dit volgens hen de reden was dat de patiënt er geen zin in had en geïrriteerd reageerde. Na het consult met de revalidatiearts ging de patiënt naar de radioloog voor een röntgenfoto en vervolgens naar de diëtist.

Consult 2

Tijdens dit consult kwam er een jonge patiënt met zijn ouders voor het eerst langs op de polikliniek in het Radboudumc. Hiervoor was hij alleen behandeld in een ziekenhuis in zijn eigen omgeving. Deze patiënt zat, voor Duchenne begrippen, al vroeg in een rolstoel. Daarnaast heeft deze patiënt een jaar geleden een operatie aan zijn voet ondergaan waarbij de achillespees verlengd is om zijn voeten in de juiste stand te laten staan.

Tijdens dit consult werd er een ingevulde vragenlijst door de patiënt overhandigd aan de revalidatiearts (dit werd in het vorige consult vergeten). Deze vragenlijst bevat de meeste noodzakelijke informatie. Hierdoor kan er meer tijd besteed worden aan de knelpunten en vragen van de patiënt aangezien de tijd van een consult beperkt is. Elk consult duurt maar 30 minuten en dat is best weinig voor de hoeveelheid informatie die er besproken wordt. Na de algemene vragen over school, slapen en dagelijkse handelingen werd er gevraagd naar de voeten van de patiënt. De patiënt gaf zelf aan hier liever niet over te praten. De moeder van de patiënt vertelde dat 6 maanden na de operatie de rechtervoet van de patiënt alweer in de verkeerde stand stond. Een nieuwe operatie werd aangeraden door de orthopeed die de patiënt voorafgaand aan dit consult had gezien. Echter, de moeder van de patiënt was erg huiverig voor een nieuwe operatie aangezien niet het gewenste resultaat werd geleverd met de vorige operatie. De vorige operatie werd uitgevoerd in het ziekenhuis in de omgeving van de patiënt.

Verder vertelden de ouders dat het een lange dag was voor de patiënt, die al vanaf 5:15 wakker was. De mogelijkheid om te overnachten in een hotel dicht bij het ziekenhuis werd niet gebruikt. Deze mogelijkheden krijgen patiënten en ouders altijd voorafgaand aan het jaarlijkse onderzoek op de polikliniek.

Een ander knelpunt wat door de moeder van de patiënt werd aangekaart was het transport van de patiënt. Momenteel kan de patiënt alleen vervoerd worden in de eigen auto wat erg zwaar is voor de ouders en ook voor de patiënt geen optimale houding oplevert. De aanvraag voor een busje werd door de gemeente afgewezen en de familie heeft geen geld om een eigen bus aan te schaffen. Een crowdfunding leverde te weinig geld op waardoor er nog niks veranderd is. De moeder van de patiënt gaf wel aan dat ze in beroep gaan tegen de afwijzing van de gemeente. Hiervoor vroeg ze aan de revalidatiearts of ze medische verklaringen op kan stellen zodat die bijgevoegd kunnen worden. Doordat er weinig mogelijkheden zijn qua vervoer kan de familie weinig tot geen spontane uitjes ondernemen wat de familie wel dwars zit.

Op de röntgenfoto van de rug van de patiënt was lucht te zien in de maagstreek. De patiënt gaf ook aan vaak te moeten boeren en de kinderrevalidatiearts gaf aan dat hij dit ook moet blijven doen omdat hij hier anders last van kan krijgen. De patiënt heeft ook moeite met ontlasting. De moeder wil dit probleem eerst proberen op te lossen door de patiënt meer te laten drinken en wanneer dit niet voldoende helpt te starten met bepaalde medicijnen om dit probleem te verhelpen.

Doordat de rechtervoet van de patiënt niet meer recht staat kan de patiënt niet meer staan en kan hij ook geen statafel meer gebruiken. Hierdoor ligt of zit de patiënt alleen maar wat de ontlasting ook minder stimuleert. De revalidatiearts gaf aan dat een eventuele operatie misschien wel nodig kan zijn om dit probleem te verhelpen maar dat dit misschien niet te snel al gedaan moet worden. Er zijn meerdere zaken die eerst geregeld moeten worden zoals het transport van de patiënt, de zithouding van de patiënt en de ontlasting problemen.

Multidisciplinair overleg

Tijdens het multidisciplinaire overleg werden beide patiënten besproken door de betrokken professionals: fysiotherapeut, ergotherapeut, diëtist, kinderrevalidatiearts en longarts. In het dossier konden foto’s en andere resultaten teruggevonden worden. Op basis van deze informatie werden bepaalde adviezen voorgesteld. Deze adviezen worden tijdens de nabespreking met de patiënt en ouders doorgenomen. Tijdens het multidisciplinaire overleg werd er meermaals benoemd dat er contact opgenomen zou gaan worden met de zorgverleners die in de eigen omgeving van de patiënten betrokken zijn om bepaalde zaken af te spreken.

Voor de patiënt van het eerste consult gaat nagevraagd worden of een aantal van de testen van de paramedici volgend jaar al in de eigen omgeving van de patiënt uitgevoerd kunnen worden omdat de patiënt het hier erg zwaar mee had. Uit de testen van de paramedici was een flinke achteruitgang te zien terwijl de patiënt zelf tijdens het consult aangaf weinig achteruitgang te merken.

Verder merkte ik op dat tijdens het consult de kinderrevalidatiearts vragen op de patiënt richt en dat de ouders toevoegen waar dat nodig is of wanneer de patiënt zelf geen antwoord wil geven zoals in het eerste consult het geval was. Hierdoor wordt de patiënt al vroeg in het proces actief betrokken. Dit is onder meer belangrijk om later de transitie naar meer zelfstandigheid te kunnen maken.

Ten slotte hoorde en merkte ik dat het voor de patiënt van het tweede consult misschien voordeliger was geweest als hij in een eerder stadium in contact was geweest/gekomen met de poli in het Radboudumc. Deze patiënt kwam nu pas voor de eerste keer langs en, naast dat hij zo niet meer kan deelnemen aan een net gestart wetenschappelijk onderzoek, had de multidisciplinaire poli misschien meer voor de patiënt kunnen betekenen met betrekking tot de voetoperaties en de nabehandeling hiervan.

7.4 Appendix 4: Topic lists interviews

Topic list interviews zorgprofessionals

Datum: Namen interviewers:

Introductie

Welkom [Naam],

Ik wil u alvast bedanken voor uw deelname aan dit interview. Ik zal mezelf eerst even kort voorstellen. Mijn naam is Thijs Som en ik ben op dit moment bezig met mijn master Science Management Andy Innovation aan de Radboud Universiteit in Nijmegen, hiervoor heb ik mijn bachelor Medische Biologie afgerond. Op dit moment ben ik bezig met mijn afstudeeronderzoek. *Tweede interviewer voorstellen*.

Doel van het onderzoek
Het doel van dit onderzoek is het in kaart brengen van de regeldruk en administratieve lasten die patiënten/ouders en zorgprofessionals ervaren bij het organiseren van de zorg voor Duchenne. Daarbij willen we de barrières en knelpunten boven water krijgen en achterhalen of er al bestaande oplossingen zijn of wat er nodig is om bepaalde knelpunten op te lossen. Het doel is om tot aanbevelingen te komen om de regeldruk van patiënten/ouders en zorgprofessionals te verminderen.

Allereerst de vraag of u bezwaar heeft als de audio van dit interview wordt opgenomen. Dit zal enkel gebruikt worden om dit interview uit te werken en zal daarna op een beveiligde schijf bewaard worden. Het interview zal maximaal 60 minuten duren en zal volledig anoniem verwerkt worden. Het transcript van het interview kan ik u mailen zodat u daar eventueel nog wijzigingen in kan laten doorvoeren. Deze interviews kunnen ons helpen om belangrijke knelpunten te identificeren en mogelijke oplossingen te onderzoeken om de regeldruk te verlagen.

- **Algemene vragen**
    - Voorstellen
        - Zou u zich kort voor kunnen stellen?
            - Waar werkt u?
            - Hoe lang bent u werkzaam binnen het zorgpad van Duchenne?
        - Wat is uw functie binnen het zorgpad van Duchenne?
            - Welke taken voert u uit binnen het zorgpad van Duchenne?
            - Voert u taken uit die buiten uw officiële takenpakket? Zo ja, welke en waarom?

Om de zorg van patiënten met Duchenne goed te organiseren, moet mogelijk veel geregeld en afgestemd worden met verschillende partijen.

- **Wie regelt wat**
    - Hoeveel tijd bent u kwijt met organisatie en afstemming van zorg rondom Duchenne (registraties)?
        - Zijn er registraties die volgens u onnodig zijn met het oog op de kwaliteit van de zorg?
            - Heeft u hier voorbeelden van?
        - Zijn er andere registraties die volgens u onnodig/overbodig zijn?

            - Heeft u hier voorbeelden van?
        - Heeft u een voorbeeld van zaken die (onnodig) veel tijd kosten om te organiseren?
    - Welke zorg organiseert u voor of namens de patiënt en ouders?
        - Worden er door de ouders/patiënten taken bij u neergelegd met betrekking tot het regelen van bepaalde zorg of voorzieningen?
        - Wat moet u doen om dit voor de patiënt en ouders te regelen?
        - Welke barrières ervaart u tijdens het organiseren en aanvragen van deze zorg en voorzieningen?
    - Ervaart u verschillen in zorgvraag bij patiënten?
        - Zo ja, waar ligt dit aan (patiënt/beloop ziekte, systeem)?
    - Ervaart u verschillen in zelfredzaamheid bij patiënten met Duchenne en hun ouders met betrekking tot het organiseren van zorg?
        - Zo ja, in hoeverre leidt dit tot (ongewenste) verschillen in zorggebruik?

- **Mogelijke oplossingen**
    - Wat zou volgens u anders kunnen of moeten?
    - Heeft u al ideeën of oplossingen voor bepaalde barrières?
    - Wat is er nodig (welke middelen) om bepaalde barrières weg te nemen?

Afsluiting

- Is er nog iets wat we nog niet besproken hebben en wat van belang kan zijn?
- Zijn er vanuit u verder nog vragen of suggesties, bijvoorbeeld met betrekking tot het onderzoek?
- Heel erg bedankt voor uw tijd!

Topic list interviews patiënten en ouders/verzorgers

Datum: Namen interviewers:

Introductie

Welkom [Naam],

Ik wil u alvast bedanken voor het meedoen aan dit interview. Ik zal mezelf eerst even kort voorstellen. Mijn naam is Thijs en ik ben op dit moment bezig met mijn master Science Management and Innovation aan de Radboud Universiteit in Nijmegen, hiervoor heb ik mijn bachelor Medische Biologie afgerond. Op dit moment ben ik bezig met mijn afstudeeronderzoek. *Eventuele tweede interviewer voorstellen*.

Doel van het onderzoek
Het doel van dit onderzoek is het in kaart brengen van de dingen die geregeld moeten worden door Duchenne patiënten/ouders en zorgprofessionals. Daarbij willen we de problemen die ervaren worden boven water krijgen en achterhalen of er al bestaande oplossingen zijn of wat er nodig is om bepaalde problemen op te lossen. Dit alles om de regeldruk van patiënten/ouders en zorgprofessionals te verminderen.

Vindt u het goed als ik dit gesprek opneem? De informatie zal buiten mijn onderzoeksgroep met niemand anders worden gedeeld en zal worden opgeslagen op een beveiligde schijf. Het interview zal maximaal 60 minuten duren en zal volledig anoniem verwerkt worden. De uitwerking van het interview kan ik u mailen zodat u daar eventueel nog wijzigingen in kan laten aanbrengen. Deze interviews kunnen ons helpen om belangrijke problemen te identificeren en mogelijke oplossingen te onderzoeken om de regeldruk te verlagen.

- **Algemene vragen**
    - Zou u zichzelf kort voor kunnen stellen?
    - Hoe ervaart u de zorg voor Duchenne in het algemeen?
    - Ervaart u problemen met betrekking tot regeldruk en administratieve lasten?
    - Ervaart u problemen in het algemeen met betrekking tot de zorg voor Duchenne?

- **Wie regelt wat**
    - Hoeveel tijd bent u kwijt met het organiseren van de zorg?
        - Ervaart u wel eens problemen bij het regelen van de zorg?
        - Welke aspecten van de zorg rondom Duchenne kosten u het meeste tijd en moeite?
    - Welke zorg en voorzieningen moet u zelf organiseren en aanvragen?
        - Wat moet u doen om dit te regelen?
    - Hoe ervaart u de samenwerking tussen verschillende partijen betrokken bij de zorg van Duchenne?
    - Hoe ervaart u het aanvragen van voorzieningen, denk aan rolstoel, transport, passend onderwijs etc.?
        - Waar loopt u tegen aan?
        - Waarom loopt u hiertegen aan?
    - Is het wel eens niet gelukt om bepaalde zorg te regelen?

    - Met wie kunt u contact opnemen wanneer aanvragen en organiseren niet (goed) lukt?
        - Hoe ervaart u deze hulp?

- **Mogelijke oplossingen**
    - Hoe kan het voor u makkelijker worden om de zorg te regelen?
    - Waar of wanneer had u meer behoefte aan informatie omtrent het organiseren en aanvragen van zorg?

- **Afsluiting**
    - Is er nog iets wat we nog niet besproken hebben en wat van belang kan zijn?
    - Heel erg bedankt voor uw tijd!
    - Zijn er vanuit u verder nog vragen of suggesties met betrekking tot het onderzoek?

7.5 Appendix 5: Email METC-Oost Nederland

Titel van het onderzoeksprotocol: Optimization of the care pathway for patients with Duchenne Muscular Dystrophy (DMD): an observational case study*

Dossiernummer METC Oost-Nederland: 2022-13811

Naam hoofdonderzoeker: Thijs Som

Naam onderzoekscentrum: Radboudumc

Naam indiener: N. Stadhouders

Datum indiening: 10 mei 2022

Geachte meneer Stadhouders,

U heeft de commissie verzocht een uitspraak te doen over of bovengenoemd onderzoek onder de Wet medisch-wetenschappelijk onderzoek met mensen (WMO) valt en op grond daarvan door een erkende medisch-ethische toetsingscommissie beoordeeld moet worden.

De onderzoeksdeelnemers worden niet aan WMO-plichtige handelingen onderworpen en aan hen worden geen WMO-plichtige gedragingen opgelegd.

Op grond hiervan verklaart de commissie dat het onderzoek niet onder de WMO valt. Voor de uitvoering ervan is derhalve geen positief oordeel vereist van de METC Oost-Nederland of een andere erkende medisch-ethische toetsingscommissie.
De commissie heeft uw onderzoek alleen beoordeeld op WMO-plicht en niet aan een inhoudelijk oordeel onderworpen (in de proefpersoneninformatie kan daarom niet worden vermeld dat het onderzoek is goedgekeurd door een METC).

Dit oordeel is tot stand gekomen na bestudering van de volgende documenten:

- Aanbiedingsbrief d.d. 10 mei 2022
- Onderzoeksprotocol d.d. 5 april 2022
- Informatiebrief d.d. 10 mei 2022
- Topic list interviews zorgprofessionals d.d. 10 mei 2022

Voor zover u dit nog niet heeft gedaan raad ik u aan in de deelnemende centra na te gaan of voor de uitvoering van uw niet-WMO-onderzoek een beoordeling door de niet-WMO-toetsingscommissie ter plekke vereist is (zie voor het Radboudumc de website METC Oost-Nederland: niet-WMO-plichtig onderzoek).

Graag attendeer ik u voorts op het Integraal Kwaliteitssysteem wetenschappelijk onderzoek (IKS) voor wet- en regelgeving en het beleid van het Radboudumc t.a.v. niet-WMO-Onderzoek. Hier vindt u onder het kopje Radboudumc SOPs o.a. het 'Normenkader statusonderzoek'. Mocht u vragen hebben over informatie in het IKS dan kunt u contact opnemen met het RTC Clinical Studies (zij

bespreken dan uw vra(a)g(en) met ter zake deskundige in het Radboudumc en kunnen u dan adviseren).

Ik vertrouw erop u met dit bericht van dienst te zijn.

Met vriendelijke groet,

Prof. Dr. P.N.R. Dekhuijzen, voorzitter

METC Oost-Nederland
METCoost-en-CMO@radboudumc.nl

T (024) 3613154

Radboud universitair medisch centrum
Tandheelkunde gebouw
Philips van Leydenlaan 25 (route 348), Nijmegen

www.radboudumc.nl

www.metc-oost-nederland.nl

*Note that the final title of the research has changed.

7.6 Appendix 6: Informed consent letter

Informatiebrief voor interviews voor onderzoek regeldruk bij de ziekte van Duchenne

1. Algemene informatie

Het Radboudumc zet in op continue kwaliteitsverbetering en persoonsgerichte zorg. In dit kader is een onderzoek gestart naar de ervaren regeldruk rondom de ziekte van Duchenne. Het is belangrijk om meer te weten te komen over welke problemen patiënten met de ziekte van Duchenne, ouders en zorgprofessionals ervaren met betrekking tot het organiseren en aanvragen van zorg en voorzieningen. Daarnaast is het belangrijk om te weten welke oplossingen er al bestaan voor bepaalde problemen, wellicht elders in het land, en op welke manieren andere problemen opgelost kunnen worden om zo de regeldruk te verlagen.

2. Doel van het onderzoek

Het doel van dit onderzoek is om door middel van interviews in kaart te brengen welke problemen Duchenne patiënten, ouders en zorgprofessionals ervaren bij het organiseren en aanvragen van zorg en voorzieningen. Op basis hiervan worden aanbevelingen gegeven die bijdragen aan het verminderen van de regeldruk.

3. Achtergrond van het onderzoek

Dit onderzoek maakt deel uit van een masterscriptie onder supervisie van de afdelingen kinderrevalidatie van het Radboudumc en IQ healthcare. IQ Healthcare is een wetenschappelijke afdeling van het Radboudumc die zich inzet voor kwaliteitsverbeteringen binnen de zorg. De afdeling kinderrevalidatie in het Radboudumc maakt deel uit van de multidisciplinaire polikliniek voor Duchenne patiënten. Het Radboudumc is gespecialiseerd centrum voor zorg bij de ziekte van Duchenne. De ziekte van Duchenne is een ingrijpende ziekte met een hoge ziektelast en een ongunstige prognose. Patiënten hebben intensieve zorg en ondersteuning nodig vanuit verschillende zorgverleners en domeinen. Dit vergt een grote mate van onderlinge afstemming, administratieve last en regeldruk. Het is niet eerder in kaart gebracht hoe de tijd en moeite om de zorg te regelen en af te stemmen wordt ervaren door patiënten, ouders/mantelzorgers en zorgverleners, en of er mogelijkheden zijn om deze regeldruk te verminderen of efficiënter te organiseren. Dit onderzoek beoogt dit door middel van interviews met betrokken partijen in kaart te brengen. Het onderzoek loopt van 7 maart 2022 tot en met 29 juli 2022.

Voordat u besluit deel te nemen aan een interview is het belangrijk dat u weet wat wij van u vragen.

Neemt u even de tijd om de volgende informatie goed te lezen. U kunt altijd vragen stellen voorafgaand aan, of tijdens het interview.

4. Waarom bent u benaderd voor een interview en wat houdt meedoen in?

U bent benaderd om deel te nemen omdat u betrokken bent bij het zorgpad van Duchenne of dit pad zelf doorloopt. Mogelijk heeft u ervaringen met regeldruk en barrières bij het organiseren van de zorg. Wij hopen dat u uw ervaringen met ons wil delen.

Waar gaan de interviews over?

- Uw rol binnen het zorgpad van Duchenne
- Uw ervaringen met organiseren van zorg
- Uw ervaringen met administratieve lasten
- Uw inschatting van welk deel van de administratieve lastendruk mogelijk kan worden verminderd
- Mogelijke oplossingsrichtingen voor ervaren knelpunten

5. Wat wordt er van u verwacht?

Als u deel wilt nemen aan een interview, maken we een afspraak op een tijd en plaats die voor u goed uitkomt, dit kan eventueel online. Het interview duurt maximaal 60 minuten en er wordt (met uw toestemming) een audio opname gemaakt.

6. Als u niet mee wil doen met het onderzoek

Het deelnemen aan een interview levert inzicht in de problemen en knelpunten die optreden binnen het zorgpad van Duchenne met betrekking tot het organiseren en aanvragen van zorg en voorzieningen. Uw antwoorden kunnen ook anderen helpen om zo de regeldruk binnen het zorgpad van Duchenne te verlagen. U bent uiteraard niet verplicht deel te nemen maar we zouden het wel erg op prijs stellen. U kunt uw deelname ten alle tijde stoppen zonder opgave van redenen.

Zijn er risico's aan deelname aan het interview? Er zijn geen risico's verbonden aan deelname aan het interview. Als u hier toch uw twijfels over heeft kunt u contact opnemen met de onderzoekers (voor contact informatie, zie hieronder).

7. Gebruik en bewaren van de gegevens

De opnames worden bewaard op een beveiligde netwerkschijf, waar alleen projectleden toegang toe hebben. Alle gegevens worden 15 jaar bewaard. Binnen vijf werkdagen wordt een korte samenvatting van de belangrijkste bevindingen uit het interview overlegd ter goedkeuring.

Wat gebeurt er met de resultaten? De resultaten worden gebruikt om een uitspraak te doen over de verschillende problemen en knelpunten die ervaren worden omtrent het aanvragen en organiseren van zorg en voorzieningen. De resultaten worden in de vorm van aanbevelingen in een rapport verwerkt dat wordt aangeboden aan de multidisciplinaire polikliniek voor Duchenne in het Radboudumc en aan IQ Healthcare. Daarnaast worden de resultaten gepubliceerd in een wetenschappelijk tijdschrift. Er zullen geen citaten uit de interviews gebruikt worden zonder uw specifieke toestemming.

Wanneer u voldoende bedenktijd heeft gehad, wordt u gevraagd te beslissen over deelname aan dit onderzoek. Indien u toestemming geeft, zullen wij u vragen deze op de bijbehorende toestemmingsverklaring schriftelijk te bevestigen. Door uw schriftelijke toestemming geeft u aan dat u de informatie heeft begrepen en instemt met deelname aan het onderzoek.

8. Heeft u vragen?

Bij vragen kunt u contact opnemen met Thijs Som of Niek Stadhouders. Voor onafhankelijk advies over meedoen aan dit onderzoek kunt u terecht bij de onafhankelijke onderzoeker, Gert Westert. Hij heeft kennis van het onderzoek, maar is niet inhoudelijk betrokken.

Thijs Som
06-30505282
thijs.som@radboudumc.nl

Dr. Niek Stadhouders
0614467004
niek.stadhouders@radboudumc.nl

Contactgegevens onafhankelijk onderzoeker

Prof. Dr Gert Westert

Gert.Westert@radboudumc.nl

TOESTEMMINGSVERKLARING – interview

Te tekenen voor aanvang van het interview
Leest u onderstaande stellingen zorgvuldig en zet een kruisje in het vakje op elke lijn.
Zet daarna de datum, uw naam en uw handtekening onderaan de pagina.

Titel onderzoek	**Regeldruk bij de ziekte van Duchenne**	
1	Ik bevestig dat ik het informatieformulier over de interviews gelezen en begrepen heb.	
2	Ik bevestig dat ik de mogelijkheid heb gehad om vragen te stellen over het interview en dat ik tevreden ben met de ontvangen antwoorden.	
3	Ik begrijp dat het mijn eigen keuze is om deel te nemen aan het interview en dat ik op ieder moment kan stoppen zonder opgave van reden.	
4	Ik begrijp dat ik geen vragen hoef te beantwoorden als ik dat niet wil.	
5	Ik begrijp dat de interviews opgenomen zullen worden.	
6	Ik begrijp dat alle informatie die ik tijdens het interview geef, vertrouwelijk behandeld en veilig opgeslagen wordt, en dat de naam van de organisatie en het project niet anoniem zullen zijn.	
7	Ik begrijp dat de informatie die ik tijdens het interview geef met de leden van het projectteam wordt gedeeld.	
8	Ik geef toestemming om de interviewgegevens 15 jaar na afloop van dit onderzoek te bewaren.	
9	Ik ga er mee akkoord dat mijn antwoorden gebruikt kunnen worden voor onderzoeksdoeleinden (zoals rapportages, publicaties en/of presentaties) en ik begrijp dat er geen citaten uit mijn interview gebruikt zullen worden zonder mijn specifieke toestemming daarvoor.	
10	Ik ga akkoord met deelname aan dit interview	

↑ Naam ↑Datum ↑Handtekening

↑ Naam onderzoeker ↑Datum ↑Handtekening