


Perspectives on Everyday Life Challenges of Danish Young People With Duchenne Muscular Dystrophy (DMD) on Corticosteroids

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Abstract

This study aimed to investigate perspectives on everyday life challenges of young persons with Duchenne muscular dystrophy in Denmark treated with corticosteroids perceived by young persons and their parents to improve rehabilitation interventions. Nineteen semi-structured interviews were conducted: 10 individual interviews with 10 persons with DMD and six individual interviews with parents (five mothers and one father) and three couple interviews (three mothers and three fathers). The analysis was guided by interpretive description methodology and Antonovsky's Sense of Coherence theory. The results indicated that persons with Duchenne muscular dystrophy existed in a flux between experiencing greater Sense of Coherence revolving around normality and less Sense of Coherence exposing their vulnerability which unfolded in four opposing themes: (1) bodily ability and disability, (2) content and anxious, (3) sociable and lonely, and (4) independent and dependent. Future rehabilitation should aim at supporting resistance resources promoting bodily ability, being content, sociable, and independent.

Keywords

Duchenne muscular dystrophy, corticosteroids, rehabilitation, interpretive description, sense of coherence, Denmark

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Background

Duchenne muscular dystrophy (DMD) is a progressive, genetic neuromuscular disease that occurs in 1:3,600 to 6,000 boy births and almost never in girls (Birnkrant, Bushby, Bann, Apkon, Blackwell, Brumbaugh, et al., 2018; Emery et al., 2015; Silva et al., 2020). The disease is diagnosed in childhood and untreated leads to loss of ambulation before teens, a need for respiratory treatment in late adolescence or early adulthood, and a shortened life expectancy due to respiratory and cardiac complications (Emery et al., 2015; Yiu & Kornberg, 2015). Specific challenges are prevalent in persons with DMD, such as language and behavioral problems and cognitive difficulties like poor implicit learning (D'Angelo et al., 2011; Magri et al., 2011; Vicari et al., 2018).

In recent decades, the course of DMD has changed due to standard treatment with steroid therapy, respiratory support (Benditt & Boitano, 2005) and prophylactic cardioprotective medication (Emery et al., 2015; Hoffman, 2020); hence, persons with DMD are now expected to grow up and live an adult life (Landfeldt et al., 2020; Rahbek et al., 2015).

A recent review has shown that all disease progression milestone events were significantly postponed in persons with DMD treated with glucocorticoids; which was associated with reduced risk of losing clinically meaningful mobility and upper limb disease progression milestones across the lifespan as well as reduced risk of death (McDonald et al., 2018). Hence the medical treatment has delayed cessation of ambulation and postponed need for respiratory aids and back surgery (Houde et al., 2008; Koeks et al., 2017; Wong et al., 2017). However, this medical treatment has serious side effects like growth restrictions, increased appetite, weight gain, anxiety, development of moon face, and being emotionally unbalanced, risk of osteoporosis, lowered immune

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response to infections, and cataract (Birnkranz, Bushby, Bann, Apkon, Blackwell, Colvin, et al., 2018; Manzur et al., 2008). Adolescents treated with corticosteroids look younger than their age and go into puberty later than peers (Wood et al., 2015). The steroid medical regimen has led to improved physical function for persons with DMD (Emery et al., 2015; Hoffman, 2020), but the side effects of the treatment has also led to physical, mental, and social challenges that may affect their everyday lives. Research on how persons with DMD under medical treatment and their families experience and cope in everyday life—especially in relation to cognitive, behavioral, and emotional functions—is sparse (Hendriksen et al., 2017). It has been shown that persons with DMD rely increasingly on assistance and support as they age and lose function, but that they also strive to maintain as much independence and control as possible (Walker et al., 2021). Literature further underscores that persons with DMD experience challenges specifically related to transitions, like finding work, accommodate higher learning, experiencing social isolation, overprotectiveness or low expectations from family (Gibson et al., 2014). Parents to children with DMD need professional help and support to facilitate effective and open communication and to address emotional and palliative care when their child reaches adolescence (Saetrang et al., 2019). However, experiences in everyday life areas and their perspectives on challenges when living with DMD and the side effects from corticosteroid treatment are sparsely described. The aim of this study was to investigate perspectives on everyday life challenges of young persons with Duchenne muscular dystrophy in Denmark treated with corticosteroids perceived by the young persons and their parents to improve rehabilitation interventions.

Methods

Setting

National Rehabilitation Center for Neuromuscular Diseases is a nationwide specialist hospital that provides highly specialized knowledge to people with a neuromuscular diagnosis, their families, and health professionals from hospitals and primary care settings (RCFM, National Rehabilitation Center for Neuromuscular Diseases [RehabiliteringsCenter for Muskelsvind], 2017, 2021). In Denmark, all persons with DMD are referred to the National Rehabilitation Center for Neuromuscular Diseases by their neurologist or general practitioner. Persons with DMD are seen either in the outpatient clinic or at the patients' homes, once a year by health professionals working in multiprofessional teams of nurses, psychologists, physicians, occupational therapists, physiotherapists, and social workers.

Design, Methodology, and Theory

The design of the study was qualitative and guided by interpretive description methodology (Thorne, 2016) and Aaron

Antonovsky's theory Sense of Coherence (Antonovsky, 1987). Interpretive description aims to form empirically and theoretically substantiated knowledge that informs practice with a view to subsequent action (Thorne, 2016). Knowledge is formed inductively from the field, where data is generated and developed with an eye for the context. This approach is relevant in this project, where the focus is to create knowledge with a view to transferability and future rehabilitation practice for people with DMD. The theoretical lens Sense of Coherence signifies a coping ability of people to deal with everyday life stressors and consists of three elements: (1) comprehensibility, which reflects the understanding of the stimuli to which one is exposed, (2) manageability, which reflects the resources a person has available in himself and in the environment, and (3) meaningfulness, which reflects on what is significant to the individual (Antonovsky, 1987). The stronger a person's Sense of Coherence is, the greater the likelihood that the person will be able to master the stress-related stimuli that the person experiences in daily life (Antonovsky, 1979).

Sampling

The participants were recruited in August 2020 by purposive sampling (Thorne, 2016) through the register at the National Rehabilitation Center for Neuromuscular Diseases by Ann-Lisbeth Højberg. The inclusion criteria were young persons with DMD, aged 13 to 20 years, registered with the National Rehabilitation Center for Neuromuscular Diseases, cognitively able to participate in an interview, understand Danish, and who were or had been treated with corticosteroids. The age range was decided based on prior knowledge about young persons with disabilities (Maxey & Beckert, 2017). Persons who met the inclusion criteria received a written invitation to participate in an interview. Written consent was obtained directly from persons with DMD ≥ 18 years of age and through the parents of persons < 18 years. Parents representing persons with DMD who were < 18 years were invited directly, and parents of persons with DMD ≥ 18 years were invited after consent from their child. The parents chose whether they wanted to participate in the parental interview individually or as a couple.

Twenty-four persons with DMD and 30 parents were invited to participate, and 10 persons with DMD and 12 parents (representing nine persons with DMD) accepted the invitation (Table 1). Fourteen persons with DMD opted out of participation due to: not wanting to participate ($n=9$) or never replied to the invitation ($n=5$). One parent opted out due to not understanding Danish.

Data Collection

Nineteen semi-structured interviews were conducted by Ann-Lisbeth Højberg: 10 individual interviews with the 10 persons with DMD and six individual interviews with the

Table 1. Demographic and Clinical Characteristics of Study Population.

Participants in all	N=22
Participants with DMD*	10/10
Individual Interviews	10/10
Age	
15–20	10/10
Educational status	
Elementary	3/10
Youth education or other further education	4/10
Work from home or unemployed	3/10
Personal and practical assistance	
24 hours a day	5/10
Only at school	1/10
Only during leisure time (max 15 hours a month)	1/10
School and/or at home (3–5 hours a day) and/or during leisure time (max 15 hours a month)	3/10
Corticosteroid treatment start	
Started between 3 and 5 years of age	5/10
Started between 6 and 7 years of age	5/10
Ambulation	
<14 years age	5/10
14–17 years age	2/10
15–20 years age (wheelchair outside)	3/10
Participants—parents**	12/12
6 Individual interviews	6/12
3 Couple interviews	6/12
Sex	
Female	8/12
Male	4/12

*Duchenne muscular dystrophy.

**The parents chose whether they wanted to participate in the interview alone or as a couple.

parents (five mothers and one father) and three couple interviews (three mothers and three fathers). Due to the COVID-19 pandemic, participants were offered the possibility to be interviewed online. All participants chose to be interviewed in their homes. All interviews were recorded and lasted between 26 and 74 minutes for persons with DMD and between 22 and 95 minutes for the parents.

A semi-structured interview guide was developed based on existing evidence on DMD and experiences from colleagues working in the multiprofessional teams at the National Rehabilitation Center for Neuromuscular Diseases. The interview guide for the persons with DMD was pilot tested and adjusted by three young persons with DMD in their early 20s. Questions in the interview guides for persons with DMD and their parents were similar but adapted to being a person with DMD or being a parent. The interview guide aimed to unfold an understanding of comprehensibility, manageability, and meaningfulness (Antonovsky, 1979) in relation to everyday life challenges and coping with DMD and medical treatment.

Key interview questions for persons with DMD were: How does a typical day proceed in your life? What is it like to go to school/youth education/further education/work/stay at home? What do you enjoy the most during leisure time? How is your relationship with your classmates/colleagues? What other young people do you spend time with besides your classmates/colleagues (online/physical)? How would you describe your life now related to years ago? What personal or practical assistance do you need during a day? What would you describe as things you control in your everyday life? What matters the most to you during your everyday life? What is it like to have DMD? What is good/bad about receiving medical treatment for your DMD? How has the whole COVID-19 lockdown period affected you? What do you do when you have a bad day? How do you envision your future? What would be your most important advice to others with DMD? What advice would you give other parents with a child with DMD in relation to treatment with corticosteroids?

For the parents, the same interview questions were posed, but related to their son with DMD, like: How does a typical day unfold in your son's life?

Ethics

This study was conducted in accordance to the Helsinki Declaration of 1975 (The World Medical Association, 2016). According to the Central Denmark Region Committees on Biomedical Research Ethics, the project was not liable to notification [Request no. 1-10-72-1-20]. All participants were informed about the project by oral and written information and were guaranteed anonymity. Written and oral consent was obtained from all participants.

Data Analysis

The methodology interpretive description guided the analysis in a four-step iterative process conducted by all three authors (C. Handberg & Jensen, 2021; Thorne, 2016). First, all the interview data from the individual and the couple interviews were transcribed and uploaded to the qualitative software program NVivo™12. Initially, the data were content, and analysis coded separately by all three authors. Then, the authors discussed the initial analyses through constant comparison in a process of in-depth exploration by which specific elements in relation to the study aim were identified (C. Handberg & Jensen, 2021; Thorne, 2016). Second, the authors re-read the transcripts and scrutinized the data further for specific patterns and connections (C. Handberg & Jensen, 2021; Thorne, 2016). Third, a critical appraisal of relationships within data was conducted by the authors, and the relevance of the thematic options leading to our primary categorizations and interpretations was determined (C. Handberg & Jensen, 2021; Thorne, 2016). Fourth, an

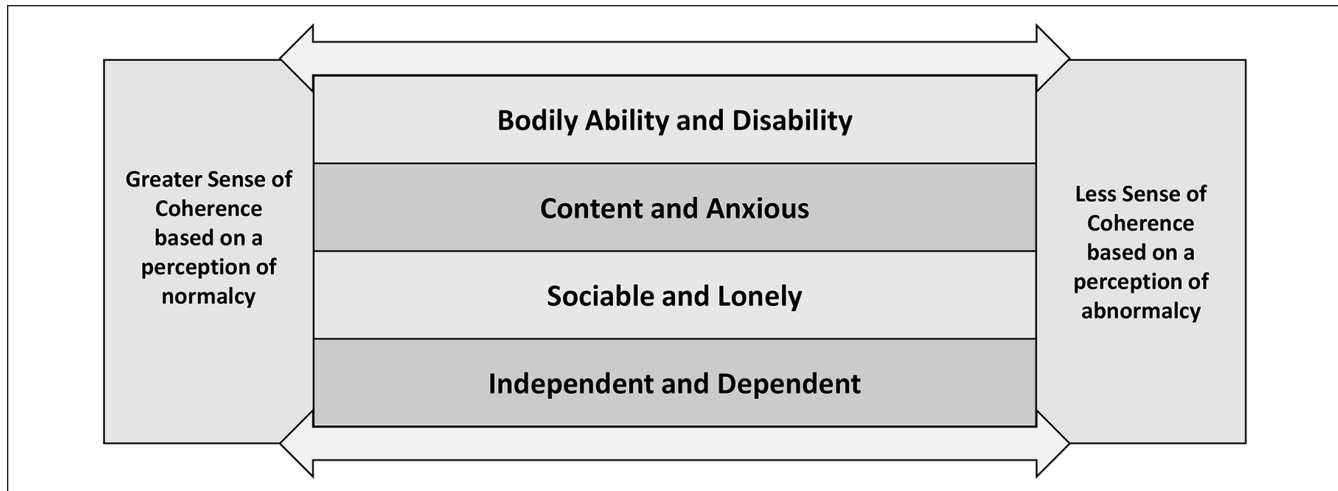


Figure 1. Understanding everyday life challenges in young persons with Duchenne muscular dystrophy treated with corticosteroids - a flux between experiencing greater Sense of Coherence and less Sense of Coherence.

extraction was made of the acknowledged main messages and key insights (C. Handberg & Jensen, 2021; Thorne, 2016).

For the aim, interview guide, analysis, and writing up of the findings, the theoretical lens of Aaron Antonovsky's Sense of Coherence theory was applied (Antonovsky, 1979, 1987) to supplement the inductive interpretive description methodology. Antonovsky's Sense of Coherence theory relates to the idea that life unavoidably offers stressors to humans, and that some people handle these stressors better than others (Antonovsky, 1979, 1987). As mentioned above three core elements represent Sense of Coherence: comprehensibility, manageability, and meaningfulness (Antonovsky, 1979, 1987). In relation to the present study, Sense of Coherence theory was relevant to apply to help gain insight into perspectives on everyday life challenges and coping in young persons with DMD on corticosteroid treatment and their parents.

Results

The analysis of everyday life challenges when living with DMD and the medical treatment of DMD showed that the participants with DMD seemed to exist in a flux between experiencing greater Sense of Coherence revolving around normality and less Sense of Coherence that exposed their vulnerability (Figure 1). All participants (persons with DMD and parents) expressed the full continuum in various degrees. The participants with DMD seemed to exist in a state that shifted between displaying bodily ability but at the same time facing a reality in an everyday life living with a disability causing constant bodily challenges. The parents and children reported the same challenges, but for the most part the parents described their child's challenges in everyday life as worse or more extensive than the child would. Emotionally, both groups of participants described the persons with DMD

to be content during their everyday lives, but they also stated that they felt anxious about the existential conditions of life. Sociably, the persons with DMD explained that they had an active and outgoing life, but both groups of participants explained a different side of the persons with DMDs' everyday lives where they felt lonely and isolated. Finally, the persons with DMD explained that they felt independent and free to do what they wanted, but at the same time both parents and the persons with DMD displayed many challenges related to being dependent during most situations in everyday life (Figure 1).

Bodily Ability and Disability

The persons with DMD emphasized that they wanted to live a life like everyone else, using words like living “*an ordinary life*” or “*a normal life.*” The participants with DMD did not initially describe their DMD as a problem or a challenge, but as they revealed more details about their everyday lives, they did reach a meaningful explanation regarding how DMD and the side effects of their medical treatment prevented them from being like everyone else. Both groups of participants explained that the persons with DMD experienced the steroid treatment as both strengthening and at the same time weakening them. Related to strengthening, they explained that the medication prolonged their walking ability, helped retain strength in their arms and fingers for longer than anticipated, and that they in general felt stronger.

I think, that if I hadn't started taking my meds you know. . . I can't remember which year it was, but then I probably wouldn't have been able to walk for as long, and I wouldn't have the strength in my arms in the way I do now.

(Person with DMD, No. E).

Both the persons with DMD and their parents explained how side effects from the steroids caused the persons with DMD to be much smaller and younger looking than their peers due to growth retardation, and that an increased appetite often entailed weight gain. In addition, the medication caused them to become moon faced and led to osteoporosis and delayed puberty. The parents and the persons with DMD expressed that everyday life for the person with DMD was limited and challenged by the disease and the side effects and that the persons with DMD wanted to be like everybody else—especially like their friends.

Yes, and to be like others, it can. . . it can. . . that's pretty important actually, when you're in a wheelchair and are trying to be as normal as possible then. . . But it is pretty annoying that you get these fat chins because of the meds, I'm really sick of that.

(Person with DMD, No. F).

Or as another person with DMD said:

With my height you know. . . and my friends also have some insider jokes which are different from mine. And I don't really get them at all. . . get anything. . . it's like. . . I feel stupid compared to them. I know I'm not. . . , but you sort of get to feel that way. . . It's not a great feeling. . . in some sort of way. It. . . it really gets to me. . . a lot actually because I feel I have a lot to say. But I'm convinced they think: 'He's so fucking childish.'

(Person with DMD, No. C).

The parents and the persons with DMD elaborated on the challenges related to living with a disability, visible to everyone else. The physical side effects were described by both groups as a condition they had to learn to comprehend and manage in their everyday lives. Due to being small and looking younger, they often were mistaken for being younger, and they related how they fought stigmatization, bullying, and prejudices by, for instance, wearing bigger shoes, getting tattoos, and dressing up to appear and be regarded as older.

Yes, and let me just show you. . . , I got a tattoo on my arm. In that way I can also display that I'm old enough. . . old enough to get one of those.

(Person with DMD, No. F).

The parents acknowledged that physical appearance affected the behavior of their child and explained that they constantly had to push and support their child into taking more initiatives and be more outgoing. What seemed to matter to the persons with DMD was being able to be “normal,” for instance, in relation to their school, studies, or work. They described a desire to look like their friends and explained that they wanted to make “normal” confident choices in relation to their life. However, at the same time persons with DMD and their parents both described that the persons with DMD often had other interests than their friends when it came to sexuality and activities related to that. This might be

caused by a late puberty due to steroids. The older participants explained that as they got older, they found it easier to comprehend, manage, and accept that they were different from their friends in some ways but the same in so many other ways. They instead chose to focus on things that they found interesting like gaming, politics, food, socializing with their families. They also explained that as they got older, they became more open, outgoing, and curious regarding the future and possibilities and worried less about their appearance.

I have learned what I can do and what I can't do. And that's. . . Imagine having a neuromuscular disorder and never realizing what you're able to do. You know, you just do what your parents tell you to do. No challenges. It sounds fucking boring. Well, you must stop regarding yourself as another kind of human being – a different species than everyone else. You're not! You're the same!

(Person with DMD, No. J).

Content and Anxious

The persons with DMD initially explained that they were happy, felt well, and were content in their lives, and that there were more good days than bad days. As the interviews unfolded, however, they described being sad, angry, afraid, and anxious. The persons with DMD and their parents described an everyday life for the persons with DMD with challenges related to comprehending and finding existential meaning in their condition in life.

We did realize that it was pretty difficult to get him to socialize during summer camp. It was rather difficult – you know – he preferred to spend time with me instead of the other children or he preferred to drive around camp alone. And it was like. . . come on. . . 'Get going'. . . until. . . and he gained weight and gained weight due to his meds and he stopped walking at the age of 8.

(Parent to person with DMD, No. N).

In general, the persons with DMD and their parents had an existential worry about life, knowing life with DMD would probably not be as long as that of other people. Some of the parents and persons with DMD described being worried, anxious, and afraid of early death. The persons with DMD explained that they did not know what to expect from life and that they could be hesitant to plan proactively because they did not know for how long they would live. Another worry for both groups of participants was the unknown progression of the person with DMDs' disease, and one participant worried about the possibility of ending in a ventilator. During the COVID-19 lockdown, these thoughts deteriorated, and they felt worse than before the pandemic.

It's difficult. . . how the future will turn out. That's something we think about constantly.

(Parent to person with DMD, No. R).

These worries about life, the future, and their relatives had in some ways psychologically matured the participants to an older age than they were, which contrasted with their young-looking bodies. The oldest participants with DMD reflected on these existential worries being easier to cope with as they got older and that they found it easier to comprehend the meaning of living with DMD and not let depressive thoughts take over too much. Furthermore, they described that they had learned to manage not to put themselves in a too vulnerable emotional position.

As one participant with DMD explained:

Yes, because a person with DMD – doesn't get to live as long as most people – it is therefore important to explore the things that you want to explore.

(Person with DMD, No. B).

In contrast to these participants, others explained that they planned regardless of their DMD and made plans for the future.

I think I've become more responsible you know, and ready to take on responsibilities and. . . I actually do feel. . . I have more knowledge I can use proactively and that. . . in general it is a good change in relation to my personality.

(Person with DMD, No. K).

Sociable and Lonely

The persons with DMD described their social life as very active, and that they were outgoing or had many online friends and enjoyed online gaming as a central event in their everyday lives. Nevertheless, as the interviews progressed, these statements changed somewhat, and both parents and persons with DMD shared a reality that was slightly different from the outgoing social life previously described by the persons with DMD. Both groups of participants described an everyday life for the persons with DMD—especially during the COVID-19 pandemic—but also before the pandemic that was less active and lacking in variety than the life they initially had described. As one parent explained:

Friends, buddies, and social life. . . He does have some really good friends with whom he hangs out, but not often enough.

(Parent to person with DMD, No. Q).

Or as a person with DMD said:

You can get lonely when living with a disability, because . . . they. . . the friends don't reflect on the fact that there is a reason why you cannot. . . why you're not able to. . . And they might think 'Well that's because he doesn't bother.' Then. . . then you might in a way get even more left out. . . that's what I think.

(Person with DMD, No. C).

Both groups of participants described that the persons with DMD spend a lot of time at home, mainly with their online

gaming community but also with their families. The persons with DMD explained that they were often left behind by their friends, who tended to forget to invite them to social gatherings or remember to contact them regularly. The persons with DMD found it difficult to maintain friendships and explained that their friends tended to seek new friendships at the start of new educational or leisure activity. On the other hand, the COVID-19 pandemic lockdown was also a relief for some because everyone was “*on equal terms*” in the sense that everyone was at home and socializing online whereas physical friendships were few. In school or during youth education programs, they spend time with their classmates, but often got left behind if there were social events after hours. The parents and the persons with DMD explained that the classmates seemed to forget about the persons with DMD and how much it could hurt to feel or see them deselected and alone, and how important friendships were to them and how they treasured the few “physical” friends they had.

He came home and told me. . . and at that point I have to admit it was difficult to be a parent. . . because he came home and told me that his friend had said: 'I'm throwing a party, but it's going to be somewhat wild, so I'm not inviting you.'

(Parent to person with DMD, No. Q).

The persons with DMD described their need for something fun to happen and how much they yearned to be part of all the fun stuff that their schoolmates were having after school hours.

It's like. . . I think it's because I cannot. . . I cannot do the same things as my friends and that is really frustrating at times, because I want to be able to play soccer or go for a walk or whatever they tend to do. And that. . . that. . . I feel how frustrating that is to me, because I feel in way – I get left out.

(Person with DMD, No. C).

Both groups of participants explained that the persons with DMD also had friends with DMD but mentioned that it was important that they were on the same level of functioning as themselves or better. The parents explained that the reality of their child with DMD was that they appeared shy, unenterprising, and at times lazy.

I think that he would love to be much more outgoing, and be with friends, spend a lot more time with them, and participate when they're doing different things. But reality is that he often has something. . . or he was not present when they decided to do stuff.

(Parent to person with DMD, No. N).

Spending time with family and close relatives who were physically present was described by parents and persons with DMD as very meaningful and rewarding. Instead of having friends from school or youth education, participants

from both groups described how they became close to their personal assistant due to spending so much time together.

It's about creating a relationship. I don't consider my personal assistant as my assistant. I regard them as my friends AND assistants, but, for instance, when I need help to visit the toilet – then that's what we talk about, and then afterwards. . . The rest of the time I regard them as friends.

(Person with DMD, No. J).

Independent and Dependent

The persons with DMD started the interviews by talking about the independent life they led, but also about an everyday life where they were dependent on others much of the time. They expressed a wish to be independent like their friends, and they explained how this was possible to some degree through help and support from mainly their parents. They described being dependent on help and support from family, friends, personal assistants, and from the public healthcare and social and/or educational systems. On an individual level, the persons with DMD also explained the dependency as a burden when wanting to be young and independent. They wished to be independent but at the same time chose to spend time with their parents because they were there and knew their everyday challenges.

But I do want them to talk to, because I don't feel I can have personal conversations with anyone but my parents. And that is sometimes pretty annoying.

(Person with DMD, No. C).

The parents emphasized the burden it could be to always guard their child, who in some contexts was an adult in terms of age, but still highly dependent on them as parents to help them physically and psychosocially to become more mature and independent. The parents moreover stressed how important it was to make their child independent and resourceful, like, contacting the authorities in relation to applying for help and assistance.

He has to make that call because. . . and that's one thing I really think I need him to learn. From the first time when he sat there by the phone and didn't have the courage to call those social workers.

(Parent to person with DMD, No. U).

The participants with DMD and their parents were very much reliant and dependent on the public “system” for the help they needed to be able to manage an everyday life in the best possible way. Help from the system could be in form of for instance personal assistance, assistive devices, support in relation to work or education. In general, the persons with DMD and their parents did not display much confidence in the system and laws in relation to getting the needed help from public authorities. The participants

(especially the parents) had negative experiences with a public system in which everything was very complicated and time-consuming.

The care coordinator has made a rule only to look at appeals from [name] once a month because she feels that the number of appeals from [name] are to such a degree where she cannot keep up. Whereas we think we don't make appeals for fun. We make them because [name] has needs which have not been met.

(Parent to person with DMD, No. Y).

The parents described how difficult it was to navigate in the system and to know what help and support was possible and/or could be expected. Additionally, they explained how the personal assistants on which they were highly dependent sometimes quit, and they therefore had new people in their houses and that it took time to trust and rely on these people. In relation to this, the older participants with DMD described how they sometimes found it difficult to have the role as an employer for their personal assistants when at the same time they were in a close relationship or even friends with the personal assistant.

Discussion

The findings on everyday life challenges when living with DMD and being treated with corticosteroids showed that the persons with DMD seemed to exist in a flux between experiencing greater Sense of Coherence through their narratives on normality and less Sense of Coherence, exposing their vulnerability. Both groups of participants explained that the persons with DMD were in a state that shifted between: Bodily Ability and Disability, being Content and Anxious, feeling Sociable and Lonely, and being Independent and Dependent. It is a paradox that persons with DMD are treated with corticosteroids that they know will suppress progression of their disease, but at the same time entail serious side effects like growth restrictions, weight gain, and being emotionally unbalanced (Birnkranz, Bushby, Bann, Apkon, Blackwell, Colvin, et al., 2018; Manzur et al., 2008). The persons with DMD and their parents explained challenges related to everyday life in general with DMD and corticosteroid treatment but did not specify which challenges related to the medicine and which related to DMD.

Considering our findings in relation to Aaron Antonovsky's salutogenic model and Sense of Coherence, the coping capacity of persons with DMD is related to dealing with everyday life stressors in relation to comprehensibility, manageability, and meaningfulness (Antonovsky, 1979, 1987). The salutogenic model indicates two mechanisms of importance: a *behavioral* mechanism that highlights the opportunity to empower people to use their resistant resources in stressful life situations and a *perceptual* mechanism related to dealing with life situation stressors by reflecting and understanding stressful and the available resources (Antonovsky, 1987,

1996; Super et al., 2016). Resistant resources in life situations are important in regard to creating conducive experiences in life supporting development and promotion of Sense of Coherence, whereas resistance deficit can undermine a person's Sense of Coherence (Antonovsky, 1987).

The life a person leads and the patterns in that life are defining for how well that person does in relation to experiencing Sense of Coherence. These patterns are founded in the balance which exists between resistant resources and resistant deficit during childhood and adolescence (Antonovsky, 1987). Sense of Coherence is often considered to be a stable entity which is developed from young adulthood and onward, stabilizing around the age of 30 (Super et al., 2016).

The persons with DMD in our study were 15 to 20 years old, and adolescence is, in general, known to be a sensitive and susceptible period of life where one's personality is founded (Antonovsky, 1987). This sensitive period becomes even more vulnerable when a person is living with a disability and visible side effects of medical treatment (Maxey & Beckert, 2017), which was also shown in our study. Adolescents with a disability face many challenges in common with their typically developing peers and are more aware of a wish to be like their peers (Maxey & Beckert, 2017). Research like the present study shows that during certain periods of life persons with DMD are exposed to stressors. In our study, stressors for the persons with DMD seemed to promote the resistant deficit elements related to the findings of being anxious, lonely, and dependent.

Adolescents with disabilities face many of the same challenges and desires as their peers, for education, friendships, and social participation, but often there are fewer opportunities for participation (Maxey & Beckert, 2017); this was also an area that both groups of our participants described as being especially challenging. These findings are underlined by research showing that after completing primary school, young persons with disabilities often experience periods of insecurity and loneliness (Hansen, 2008; Maxey & Beckert, 2017). The persons with DMD emphasized that they wanted to be normal and treated like everyone else but experienced that other people at times perceived and treated them differently than they did to young people without disabilities. In a study on adolescents with neuromuscular disorders, it was shown that the adolescents often experienced being rejected by peers and that they often felt misunderstood (Vuillerot et al., 2010). Some of the findings in our study were similar, the adolescents with DMD being challenged physically, psychologically, and socially by many life situation stressors at such a fragile time of life. Earlier stressors often will continue to occur in life in different shapes and forms, and the experience of Sense of Coherence will be affected by the ability to comprehend and manage stressors (Antonovsky, 1987). Negative attitudes toward people with a disability persist in society and based on the statements from our participants' explaining how they fought stigmatization, bullying, and prejudices by, for instance looking different such

attitudes might impact the person with DMDs' potential integration into society. Thus, the persons with DMD would indeed benefit from developing a Sense of Coherence in their lives but also, they need support because resilience is contingent on both inner and outer resources. Additionally, persons with DMD have been shown to report significantly lower quality of life than their healthy peers (Uzark et al., 2012), but despite decreased physical functioning, older boys seem to attain a better psychosocial quality of life than perceived by their parents or achieved by younger boys (Otto et al., 2017; Uzark et al., 2012). Therefore, it is important to promote the resistant resources that can help the participants with DMD to cope with comprehending, managing, and finding meaning in life.

The elements that were promoted by resistant resources among our participants seemed to be related to enhancing bodily ability, and being content, sociable, and independent. The persons with DMD talked in positive terms about everyday life, describing life with DMD as meaningful and manageable, and even though their daily life appeared to be more challenging, a behavior that seems like a healthy coping mechanism in accordance with Sense of Coherence theory (Antonovsky, 1987). Feelings like hope and excitement (compared to hopelessness and apathy) will promote the experience of Sense of Coherence, and among both groups of our participants, it seemed like particularly the older participants with DMD were more driven by their resistant resources, which to some degree caused them to project a stronger Sense of Coherence. This is in accordance with Sense of Coherence theory, underlining how the personality is formed along the way and that the resistant resources normally increase with age. These findings relate well to the disability paradox that revolves around the fact that many people living with disabilities report that they experience a good or excellent quality of life dependent upon finding a balance physically, psychologically, and socially (Albrecht & Devlieger, 1999). Therefore, it will be especially important to support the younger group of boys with DMD at an early state, strengthening their behavioral mechanisms. Support from parents and/or health professionals will enhance the boys' resistant resources in stressful life situations and their perceptual mechanism related to handling life situation stressors by reflecting and understanding what the available resources are for the specific person in a precise situation (Taylor et al., 2016). Future research should investigate the developmental patterns unique to adolescents with DMD in relation to relatives and the specific needs related to social support regarding the different age groups within adolescence. In addition, it would be relevant to conduct parent-child dyads and dyadic analysis which could hold a potential contribution to the insights of the challenges related to the persons with DMD and their parents. Finally future research should explore gender-related influences on the persons with DMD to extend an understanding of responses to life circumstances in this context.

Methodological Considerations

To ensure rigor and credibility in the study, we will address four elements of importance in relation to evaluating an interpretive description study: epistemological integrity, analytical logic, interpretive authority, and representative credibility (Thorne, 2016).

The *epistemological integrity* in our study was based in Aaron Antonovsky's Sense of Coherence theory and our approach was founded in the inductive scaffold of interpretive description (Thorne, 2016). Sense of Coherence influenced the aim, the interview guide, the analyses, the interpretation of data, and the writing up of our findings (Antonovsky, 1979, 1987). Being consistent with the use of Sense of Coherence made our *interpretive authority* clearer and more transparent. We acknowledge that there could be a potential risk of the analysis being deductive when applying Sense of Coherence as a theoretical lens, however it was used as inspiration and guide in the analysis, which can explain that it is not so distinct or apparent in the results section. In relation to our *analytical logic*, we moved from raw data to the four analytical steps and our interpretation guided by Sense of Coherence, but still in an inductive manner. This process was ensured and substantiated by applying quotes throughout the results section. Even though the concept of Sense of Coherence aims to promote health, recent studies have questioned this stability of Sense of Coherence due to an unclear understanding of the mechanisms underlying Sense of Coherence (Super et al., 2016). This entails challenges related to determining what is specifically needed in health promotion activities to strengthen Sense of Coherence for the groups under study (Antonovsky, 1996; Super et al., 2016).

The *representative credibility* in this study can be argued to be uncertain due to the small sample of persons with DMD. Only boys were included in the study, since the disease almost never occurs in girls, and no girls with the diagnosis were registered at National Rehabilitation Center for Neuromuscular Diseases (Silva et al., 2020). Nevertheless, our sample even though small represented almost half of the persons with DMD in Denmark in the studied age group. Moreover, triangulation of data was ensured by interviewing both the persons with DMD and their parents. Research using the interpretative description methodology, does not have an endpoint with an assumption of data saturation which would imply that all information from the participants had been heard so frequently that it could be anticipated (Thorne, 2016). The representation of participants and data ensured a nuanced and broad data set that supported our findings well. The parents and persons with DMD explained the challenges in general but did not however go into detail on whether the challenges they described were related to corticosteroids or DMD even though our initial intention was to point out the specific role of corticosteroids versus the DMD. The findings reflected challenges in everyday life for young

persons with DMD and their parents, and such challenges might be similar to and relevant for other groups of young people living with a disability and their parents in other contexts, countries, and populations.

Conclusions

This study provided insight into everyday life challenges of young persons with DMD in Denmark treated with corticosteroids perceived by the young persons and their parents. The participants in our study seemed to exist in a flux between experiencing greater Sense of Coherence through their narratives on normality and another reality with less Sense of Coherence, exposing their vulnerability. To improve rehabilitation interventions and underpin future health promotion efforts for persons with DMD, it seems important to aim at supporting resistance resources promoting bodily ability, and being content, sociable, and independent. The behavioral and perceptive mechanisms of support should be designed to meet physical, psychological, and social needs, which, we suggest, will be relevant for health promotion and strengthening Sense of Coherence in the persons with DMD. The successful application of behavioral and perceptive mechanisms will empower the persons with DMD to deal with stressors that can have a positive influence on their health, and furthermore contribute to constant and meaningful life experiences that increase and reinforce the experience of Sense of Coherence.

Practice Implications

To improve rehabilitation interventions for young persons with DMD, nurses and other health professionals and peers can support an experience of Sense of Coherence through provision of knowledge and counseling. Rehabilitation initiatives must support biopsychosocial resistance resources promoting bodily ability and being content, sociable, and independent in young persons with DMD. Online services seem like a good possibility for young persons with a disability to be more actively involved, and the practice implications therefore point to further development of online rehabilitation services such as information, advice, and knowledge from health professionals on how to manage everyday life's physical, psychological, and social challenges related to DMD and medical treatment. Likewise, structured professional and peer-support rehabilitation online groups for persons with DMD and their parents should be provided, where young persons with DMD and their parents are given the opportunity to meet and share experiences with peers online.

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Author Contributions

All authors have made substantial contributions to all of the following: (1) the conception and design of the study, or acquisition of data, or analysis and interpretation of data, (2) drafting the article or revising it critically for important intellectual content, (3) final approval of the version to be submitted.

Data Availability Statement

The data that support the findings of this study are available on request from the corresponding author. The data are not publicly available due to privacy or ethical restrictions.

Declaration of Conflicting Interests

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