

Adults with Spina bifida - voices from everyday life and exploration of living conditions

Till VilmaRut och Vera

Örebro Studies in Medicine 225



HANNA GABRIELSSON

**Adults with Spina bifida - voices from everyday life and
exploration of living conditions**

Cover photo: Markus Naeslund
Portrait: Alexander Donka

© Hanna Gabrielsson, 2020

Title: Adults with Spina bifida - voices from everyday life and exploration of
living conditions.

Publisher: Örebro University 2020
www.oru.se/publikationer

Print: Örebro University, Repro 11/2020

ISSN 1652-4063
ISBN 978-91-7529-361-5

Abstract

Hanna Gabrielsson (2020):

Adults with Spina bifida - voices from everyday life and exploration of living conditions. Örebro Studies in Medicine 225

The overall **aim** of this thesis was to generate knowledge about living with Spina bifida, by mapping the condition and and together with the adults with Spina bifida explore their living conditions and experiences in everyday life. **Methods** Study I was a quantitative study with a cross-sectional design. In Study II, individual experiences of daily life were explored by deep interviews using a reflective lifeworld approach. Study III had a participatory approach including five members of a photovoice group who met for eight sessions. Photographs taken by the members served as a starting point for the dialogue about what was of interest in their daily life. A narrative analysis was conducted by the researchers, incorporating the analysis the group did together. Study IV focused on alignment with the methodology in which photovoice is grounded. By returning to the ideological cornerstones of photovoice, the empirical experiences from Study III and examples from the literature were elaborated through processes within photovoice. The **findings** show that those adults with Spina bifida who were ≥ 46 years old had less complex medical conditions and better physical and cognitive functions, and had attained a higher level of education. The main theme in Study II was presented as “The contradictory path towards wellbeing in daily life.” In Study III, the members’ experiences in everyday life showed that many solutions offered by society were “An adaptation for us, but it works for no one.” The findings are further presented under three themes: “Accessibility – a never-ending project,” “Tensions of a normative view” and “Power to influence.” By focusing on action and narrative in Study IV, it is shown that dialogue, action, and interaction are important aspects of using photovoice. In **conclusion**, not all adult persons receive the support they need in everyday life, something future generations of adult persons with Spina bifida may have a higher need for. The stories and experiences of adults with Spina bifida in this thesis paint a history of not being asked, concerning their own situation. This shows that there is insufficient integration of the persons’ experiences in society’s efforts to plan for, and support, these individuals. The photovoice method was feasible for this group, providing opportunity for being part of dialogue, action, and interaction.

Keywords: Spina bifida, adults, living conditions, health, photovoice, experiences

Hanna Gabrielsson, School of Health Sciences
Örebro University, SE-701 82 Örebro, Sweden

List of studies

This thesis is based on the following studies:

- I. Bendt, M.*, **Gabrielsson, H.***, Riedel, D., Hagman, G., Hultling, C., Franzén, E., Eriksson, M., & Seiger, Å. Adults with spina bifida: A cross-sectional study of health issues and living conditions. *Brain & Behaviour*, 2020. doi: 10.1002/brb3.1736
*shared first authorship.
- II. **Gabrielsson, H.**, Karlberg Traav, M., & Cronqvist, A. Reflections on health of young adults with spina bifida: the contradictory path towards well-being in daily life. *Open Journal of Nursing*, 2015, 5, 303–312.
- III. **Gabrielsson, H.**, Hultling, C., Cronqvist, A., & Asaba, E. Views on everyday life among adults with spina bifida: an exploration through photovoice. *International Journal of Qualitative Studies in Health and Wellbeing*, 2020, 15:1, 1830702. doi: 10.1080/17482631.2020.1830702.
- IV. **Gabrielsson, H.**, Cronqvist, C., & Asaba, E. Photovoice revisited: an analysis of process and methodology. In manuscript.

The studies will be referred to by the Roman numerals I–IV throughout this thesis.

Reprints were made with permission from the respective publishers.

List of Abbreviations

AIS	American Spinal Injury Association Impairment Scale
CBPR	Community-based participatory research
CIC	Clean intermittent catheterization
CRPD	Convention on the Rights of Persons with Disabilities
FIM	Functional Independence Measure
HC	Hydrocephalus
ILO	Independent Living Organization
IQ	Intelligence quotient
IQR	Interquartile range
LSS	(Swedish acronym for <i>Lagen om stöd och service till vissa funktionshindrade</i>) Support and Service for Persons with Certain Functional Impairments Act
MF	Muscle function
MMC	Myelomeningocele
MMCUP	MMC <i>Uppföljningsprogram</i> (MMC Follow-up Program)
NSCIR	Nordic Spinal Cord Injury Registry
NTD	Neural tube defect
RLR	Reflective lifeworld research
SB	Spina bifida
SD	Standard deviation
SoL	Social Services Act
UN	United Nations
UTI	Urinary tract infection
WHO	World Health Organization

Table of Contents

PREFACE.....	11
INTRODUCTION.....	13
BACKGROUND	14
Spina bifida	14
A rare condition	16
Independence and community participation.....	16
Navigating social contexts.....	17
Navigating the health-care system.....	18
Self-care in everyday life	19
Framing disability	21
Models of disability	21
Legal rights of persons with disability	22
Grey zone concerning legal rights.....	23
RATIONALE.....	26
THEORETICAL FRAMEWORK AND CONCEPTS	27
Health from a caring science perspective.....	27
Everyday life and lifeworld theory	28
Narratives as lived experience together with others.....	28
A participatory approach	29
OVERALL AIM.....	30
Specific aims.....	30
METHODS.....	31
Overall context	32
Study I.....	32
Participants	32
Data collection.....	32
Measurements/instruments.....	33
<i>Sociodemographic factors</i>	33
<i>Structural and medical characteristics</i>	33
<i>Physical function and assistive devices</i>	34
<i>Cognitive subtests</i>	36
Data analyses	36
Study II.....	36
Participants	36
Data collection.....	37
Data analyses	37

Studies III and IV.....	38
Photovoice.....	38
Participants	39
Data collection through photovoice	39
Narrative analyses, Study III	39
Methodological analysis, Study IV	41
Ethical considerations.....	41
Informed consent.....	41
Confidentiality	42
Ethical approval	42
FINDINGS	43
Study I.....	43
Study II.....	43
Study III.....	44
The exhibition	46
Study IV	46
REFLECTIONS ON FINDINGS.....	47
Lack of opportunities for independence and participation	47
Stories of not being asked affecting identity	48
Stories as action and ability.....	50
Action as health.....	51
METHODOLOGICAL CONSIDERATIONS.....	54
Validity, reliability, and generalizability	54
Aspects of trustworthiness and credibility	55
Reflexivity	56
Considerations in analyses.....	57
CONCLUSIONS AND IMPLICATIONS OF THE RESEARCH	59
CLINICAL IMPLICATIONS	60
TACK.....	61
SVENSK SAMMANFATTNING	64
Bakgrund.....	64
Syfte	64
Metod.....	64
Resultat	65
Konklusion.....	66
REFERENCES	67

Preface

Before I studied nursing, I worked at a nursing home for children with severe autism and intellectual disabilities. This was *home* for the children who lived there most of their time. The work rested on an anthroposophical philosophy, which imbued aspects such as the home like environment, the food served, and the activities carried out. In the end, the philosophy formed the actions and contributed to a pronounced humanistic view strengthened by involving the children and highlighting the importance of their surroundings. As I reflect, this work had a strong impact on me and led to my decision to become a nurse.

As this project evolved from my clinical work as a registered nurse and coordinator of a team working with persons with Spina bifida (SB), my point of departure was based on a professional perspective. As a nurse, I met persons with SB, a congenital spinal disorder, in a clinical setting, and I also met persons with acquired spinal cord injury. The clinic has responsibility for medical follow-up of all persons over 18 years with SB. This responsibility includes regular, multidisciplinary check-ups with a focus on the medical consequences of the disorder. Working as part of a multidisciplinary team expands the understanding of the patient's situation from several perspectives. The team working with this group of persons gradually became aware of needs that required a different approach compared to that for persons with acquired spinal cord injury. The fact that the clinic had, and still has, a considerable group of adults with SB, made us start thinking of mapping the group and their experiences, as this seemed to be missing from the literature.

Having met many of the persons yearly for their check-ups, and in between for other purposes, such as assessments, supportive talk, group activities, or wound dressing, I developed a notion about the group. This was a group with a wide range of outcomes, also in terms of societal support and consequently of how their everyday life was turning out. Through meeting many of them on a regular basis, even if at times a year went by between meetings, meant that relationships developed. Even though many in this group had medical concerns that needed to be monitored on a regular basis, these did not seem to be the main concerns of the persons themselves. As an example, a colleague and I started to organize group activities, at first with women with SB, with focus on healthy food and exercise. I gradually

became aware of how activities and events in everyday life, like cooking and exercise, were at times hard to achieve. Still, in many cases, they were expected to be taken care of by the persons themselves. The group activities were highly appreciated by the members of the group, mostly because they provided a social meeting space (the focus on cooking and exercise was of secondary importance). This, together with the general situation for adults with SB, contributed to my interest to learn more about adult persons with SB and their everyday lives through their own voices. It has been important for me to pay homage to voice, because who could know everyday life with SB better than those living it. By involving persons with SB in constructing knowledge concerning their situation, and their stories, I've come to learn so much. Some of this knowledge is shared in this thesis.

Introduction

This thesis aims to contribute to the knowledge about the everyday life of adult persons living with SB. It is important to note that my background and thinking is grounded in caring science, which has guided the choice of methods and concepts in this thesis. Grounding my reflections in caring science means embracing the view that persons are experts on their own lives, based on personal experiences in relation to health (Dahlberg & Segesten, 2010). This view and related concepts of relevance will be further developed under “Theoretical framework and concepts” and “Methods”. While stating my grounding in caring science, I also draw on literature from related disciplines such as medicine, disability science, law, occupational science, narrative theory, and community-based participatory research (CBPR).

The term “everyday life” in this thesis is used interchangeably with “daily life,” and is based on the concept of existence understood as lifeworld. To access a person’s lifeworld and openly and compliantly touch the life context and life project of that person means entering into that person’s world and experiences. The lifeworld theoretical approach includes the whole living context affecting people’s processes in health and caring (Dahlberg & Segesten, 2010). It is through everyday life that the lifeworld takes place. Living conditions are in this thesis viewed as both internal and external conditions for living, and the relationship between them.

In this thesis, different perspectives on living conditions and the everyday life of persons with SB will be presented. Persons with SB are not one homogenous group, since they represent a wide range of physical disabilities and cognitive outcomes. Still, the experiences of growing up in a specific society with a congenital condition constitute a common platform.

Background

As the number of adults living with SB is increasing, it is important to improve knowledge about this population's health and living conditions, by mapping these and their situated lived experiences in everyday life through active community engagement. Most of the research about SB, so far, has been in the medical field, conducted in paediatrics and predominantly in North America. The research has had an emphasis on various medical aspects linked to the corresponding speciality in health care, and has been criticized for being focused on narrow medical outcomes, as well as being non-collaborative, based on small samples of convenience, with no comparison groups and a lack of consistent standards of measurement (Liptak & El Samra, 2010). Research in adults with SB is sparse but increasing, and includes mostly young adults (Boudos & Mukherjee, 2008; Buffart et al., 2008; Ridosh, Braun, Roux, Bellin, & Sawin, 2011; Verhoef et al., 2004; Young, Anselmo, Burke, McCormick, & Mukherjee, 2014). Very few studies have involved aging and health outcomes in people with SB (Liptak et al., 2016; Morley et al., 2020). A few recent studies have, however, included older adults. Examples are a study from Norway which included adults >50 years of age and studied aspects of participation, as well as health issues and physical function (Lidal, Lundberg Larsen, & Hoff, 2019), and a North American online survey, which included adults throughout the whole life span, and focused on demographics, health care access, and health conditions (Morley et al., 2020). One review protocol on psychosocial outcome in adults with SB has been published (Betz et al., 2019) and one Ph.D. thesis examined the relationship between self-determination and wellbeing in adults with SB, likewise through an online survey (Bruegel, 2017). The lived experience of adults with SB is sparsely researched (Kinavey, 2007). This information is relevant to the aim of this thesis.

Drawing on principles in caring science, the following paragraphs will present elements of relevance for the health context of adults with SB. The thesis starts with a brief description of SB from a medical point of view. Thereafter, aspects of meaning in relation to everyday life are presented.

Spina bifida

“Spina bifida” is a general term for a condition with a wide range of malformations, and is often used synonymously with “myelomeningocele (MMC)” (Alriksson-Schmidt, Josenby, Lindquist, & Westbom, 2018). It is

a congenital disorder that occurs early in pregnancy when the neural tube should, but does not, close, resulting in a spinal neural tube defect (NTD) (McComb, 2015). SB involves multiple body systems (Bakaniene, Ziukiene, Vasiliauskiene, & Prasauskiene, 2018; Wagner et al., 2015b) which at times cause extensive impact, leading to the description used by some, as the most complex congenital defect compatible with long term survival (Bowman, McLone, Grant, Tomita, & Ito, 2001; Talamonti, D'Aliberti, & Collice, 2007). The failure of closure of the neural tube causes a range in degree of outcome. Varying kinds of neurological loss are common (Mitchell et al., 2004). Classification is done by establishing the level of the spinal lesion and the presence or absence of hydrocephalus (HC) (Fletcher & Brei, 2010).

Most (75%) of the individuals born with SB reach adulthood today, due to improved medical and surgical treatments (Dicianno et al., 2008). However, the risk for early death remains throughout life (Oakeshott, Hunt, Poulton, & Reid, 2010), as do risks for secondary conditions from other organ systems. Impaired neurological functioning has consequences for motor and sensory functions, including urinary tract and bowel functions, resulting in higher incidence of urinary tract infections (UTIs), renal failure, obstipation, and pressure ulcers. Obesity is common. All of these are to some degree preventable secondary conditions with a high impact on health outcome. Experiences of depression and/or anxiety among individuals with SB are common (Dicianno, Karmarkar, & Houtrow, 2015; Wagner et al., 2015a; Veenboer et al., 2014), also issues with psychosocial adjustment compared to age-matched controls have been found (Holmbeck & Devine, 2010). The majority of persons with SB have shunts due to hydrocephalus (HC), and Chiari II malformation is common (Fletcher & Brei, 2010). It is well known that HC is negatively associated with cognitive function and therefore potentially interferes with many aspects of daily life (Dennis, Landry, Barnes, & Fletcher, 2006; Hampton et al., 2011; Wetzels, Heaner, Gabel, Tubbs, & Chern, 2018; Zabel et al., 2011). Many persons with SB have impaired cognitive ability with significant difficulties with executive functions (Rose & Holmbeck, 2007). "Executive functions" are defined as a group of higher order cognitive abilities including self-regulation, inhibition, planning, mental flexibility, and organization of behavior (Eslinger, 1996). Prospective memory can also be impaired and has been found to decline with age; those over 35 years of age had a larger increase in prospective memory impairments (Dennis, Nelson, Jewell, & Fletcher, 2010). Difficulties with executive functions and prospective memory

complicates contacts with health care and society in general, which becomes more evident in adulthood when demands increases.

A rare condition

According to the World Health Organization (WHO), approximately 15% (WHO, 2011) of the inhabitants in the world, 1 billion persons, are living with some kind of disability. In Sweden, the proportion is about the same. The number is increasing due to better medical treatment and the fact that the estimated age of survival of the human population is getting higher (Hallberg, 2017). Part of this is also true for persons with SB, most of whom are now reaching adulthood. However, the incidence of births with SB in Sweden is decreasing, not only due to improved prenatal diagnoses and subsequent abortion (Six out of ten pregnancies are interrupted when the fetus is found to have SB). Also, folic acid use in pregnant women results in decreased incidence (Socialstyrelsen / National Board of Health, 2014). In the 1970s, between four and eight births per 10 000 had SB. By comparison, in the last decade, the number was down to one to two/10 000 births (Bodin et al., 2018; MMCUP, 2017), with SB consequently considered as a rare condition in Sweden (MMCUP, 2020).

When looking at survival rates, or the percentage of those born with SB now living to adulthood, we see that a much larger percentage are today getting old than before. This can be viewed as a success. The society rescues more persons, i.e., those actually being born with SB, into adulthood. It does not, however, tell us anything about how the persons live their lives or their experiences of it.

Independence and community participation

Aging with a life-long disability increases the risk of negative health outcomes, and participation problems, especially when having an early-onset disability (Hilberink, van der Slot, & Klem, 2017). Many of the adult SB population lack work or employment (Törnbohm, Jonsson, & Sunnerhagen, 2014; Törnbohm et al., 2011). Studies show that even though more people with SB reach adulthood, many have difficulties meeting the expectations for, and demands on, adult individuals. This is often viewed as a lack of independence. Approximately one third of individuals with SB were found to be independent at the age of 30, one third needed supervision and occasional help, and one third needed assistance routinely for daily care

needs (Oakeshott & Hunt, 2003). Another example, shows low independence in typical “milestones of life”, described as moving away from home, studying at college, having romantic experiences and a number of friends. The executive functions such as planning, initiation and the ability to flexibly solve problems are considered to be measures of independence, which is a challenge for many in this group (Zukerman, Devine, & Holmbeck, 2011) or that are achieved later compared to others. It has been debated whether delays in skills development in youth with SB are developmentally appropriate or may be due to lack of expectations and support from home and at school, and from health care and the community as a whole (Spina Bifida Association, 2018).

One Swedish study found that children and adolescents with SB did not express motivations for becoming independent, and the mentioned motivations were based on a sense of consideration for the caregivers. This was suggested as a lack of internal drive for independence, and a possible connection to executive dysfunction (Strömfors, Wilhelmsson, Falk, & Höst, 2016). One study reporting low participation in community activities, such as employment and physical activities as well as living independently and dating in adults with SB, Boudos & Mukherjee (2008) identifies low motivation as the most common barrier to community participation. The authors (ibid.) reflect whether the low motivation could be due to: (a) never having experienced high levels of participation and therefore not considering this alternative for themselves; (b) limited peer interaction (which normally influences participation in activities) (Blum, Resnick, Nelson, & Germaine, 1991; Holmbeck et al., 2003 referred in Boudos & Mukherjee, 2008); or (c) reduced goal-directed or initiation behavior (Landry, Robinson, Copeland, & Garner, 1993 referred in Boudos & Mukherjee, 2008). The debate on possible delays in development and on low motivation and lack of internal drive is especially relevant in relation to the participatory approach in this thesis.

Navigating social contexts

Developing an identity and finding a place in a social context is, shaped not only by the surrounding communication (Cardillo, 2010) but also by interruptions in school and other social arenas affecting social development, by hospital visits and recurring bodily interventions: There are descriptions from back in time, of children with SB being hospitalized for weeks at a time, repeatedly, for orthopedic surgery or decubitus ulcer treatments (Tew

& Laurence, 1976). In a metasynthesis integrating and interpreting findings from twelve qualitative studies involving youths (9–26 years old) with SB (Lindsay, 2014), the author drew on the concepts of normalcy and biographical disruptions when presenting the following three themes: the medical management of SB; the importance of peer and family relationships; and identity and self-concept. The article suggests that youth with SB experience threats to self-conception and disruptions in biography. However, the investigated youth had strategies for maintaining normalcy within the constraints of SB. Some had social support from their family; but they still had challenges in making friends, and experiences of isolation and stigma (Lindsay, 2014). Research comparing persons with congenital disability to persons with acquired disability suggests that those with congenital disability are more stigmatized compared to persons with acquired disability, and get less personal help (Bogart, Rosa, & Slepian, 2018). In most cases, SB means being born as a patient, which can have a strong impact on identity. The development of an identity and finding one's place in a social context is part of the complexity of growing up with a congenital disability like SB. Another challenge is to handle the task of dealing with a complex disability, which is affected by the organization of care, the own ability, and/or the societal support granted (or provided by relatives) in everyday life.

Navigating the health-care system

As stated previously, SB has a wide range of both physical and cognitive outcomes. In severe SB, due to the involvement of many systems in the body, contact with many different medical specialties is needed (Fletcher & Brei, 2010). When persons with SB transition from pediatric care to adult care little is known about where they receive services (Webb, 2010). During childhood, care is often delivered through subspecialty, multidisciplinary, and coordinated settings; however, most of these programs only treat children and adolescents, but not adults (Liptak et al., 2016). Only a few clinics provide services to patients beyond adolescence, resulting in low representation of adult experiences (Thibadeau et al., 2013). Yet persons with SB continue to have elevated medical needs in adulthood (Morley et al., 2020; Webb, 2010), with significantly more hospitalizations per year compared to the general population (Armour et al., 2009; Dicianno et al., 2008). Moreover, older adults with SB have higher hospitalization rates and fewer primary care visits compared to young adults with SB (Liptak et al., 2016).

Several studies have highlighted the importance of coordinating the professional care for adults with SB (Dicianno, 2014; Young et al., 2014). To involve a health professional in the coordination of care has proved to be important in ensuring timely, effective mental health services in young persons with disability (Edwards, Patrick, & Topolski, 2003; Witt, Kasper, & Riley, 2003). A European survey with focus on living with a rare disease concluded that there was a clear lack of communication between service providers, leaving the coordination of care to the persons living with a rare condition and their main carers (most often women). This is a heavy time burden which affects the everyday life of the persons already facing severe challenges (Courbier & Berjonneau, 2017). The Swedish registry for persons with SB, the MMC *Uppföljningsprogram* (Follow-up Program) (MMCUP), states that coherent medical monitoring for adults with SB is lacking in several parts of the country (MMCUP, 2017). This is problematic for the individual with SB and is contrary to the United Nations (UN) Convention on the Rights of Persons with Disabilities (CRPD), which identifies equality and justice in health care services as a fundamental right of persons with disabilities (United Nations, 2008). Medical professionals' lack of knowledge about, and inability to recognize, common medical conditions related to SB (Spina Bifida Association, 2018) complicate the situation further, putting higher demands on the persons with SB. It has been reported for other rare conditions (and this equally applies to SB) that, due to the low prevalence of the condition, there is a lack of expertise among medical staff, which will only continue to worsen as the condition becomes rarer. This means that the person with the condition is forced to become knowledgeable about his or her own state (Courbier & Berjonneau, 2017) and communicate this knowledge to professionals in health care. This can be a challenging task which requires awareness of the organization of care and the ability to speak for oneself, alternatively having a functional proxy.

Self-care in everyday life

As stated earlier, developments in the medical field have improved the outcome in terms of survival for persons with SB. Living with SB often requires self-care, as the condition in most cases involves medical conditions, some of which demand attention in everyday life to prevent life-threatening secondary conditions. Being aware of signs of shunt failure or pressure ulcers, and responding in a timely way when suspecting these complications will have an important impact on the outcome. Some adults

with SB receive home care for everyday life and, therefore, help to remember and perform these self-care activities. Others, however, are expected to manage this by themselves.

I here divert to deepen the discussion of one of many everyday concerns in SB. From birth, a lot of attention is paid to bladder function in SB to reduce the risk of pyelonephritis, urinary leakage, and UTIs. The most common dysfunctions of the bladder involve an overactive bladder, incomplete bladder emptying, and increased intravesical pressure during filling, which eventually can cause renal failure (Backhaus et al., 2002). For patients who are unable to properly empty the bladder, a common technique is clean intermittent catheterization (CIC), used in Sweden since the early 1970s (Lapides, Diokno, Silber, & Lowe, 1972). This treatment is crucial for survival and improvement of urological symptoms. Training for self-CIC is needed from an early age. To achieve the desired effect with avoidance of secondary conditions, this is a constant element of everyday life for many persons with SB. Some individuals use other techniques for bladder emptying, but whatever the technique, repeating the procedure daily is of importance. It should be noted that, due to neurological loss of sensation, the need to perform CIC is not necessarily linked to a feeling of a need to void. Rather, it is a procedure that needs to be routinely done by the watch, and checked, to avoid secondary conditions (Lindehall, 2007).

Other procedures of everyday life with SB include a bowel regimen and procedures for both bladder and fecal incontinence. Incontinence has a higher impact on social life than medical disorders, and can constitute a barrier for social participation (Lidal et al., 2019; Vu Minh Arnell & Abrahamsson, 2019). Addressing it demands both time and effort. Worth noting is that, in order to prevent serious complications, surgical treatments of urinary incontinence in SB often leave a way open for urinary leakage when the bladder overfills. This is done because it is known that, due to cognitive impairments, persons with SB may have problems achieving regular CIC (Lindehall, 2007), a requirement for the continence preservation surgery to work as intended. This is one example where medical priorities, albeit necessary, can impact negatively on participation in social life.

Framing disability

Lindberg, (2016) found that most research on disability in Sweden is done in the medical field, while research on legal, socio-political, and economic aspects on disability is lacking. He also found that there was little contact between the disability movement and disability studies in Sweden, and that most researchers did not have the experience of living with disabilities themselves (Lindberg, 2016). Congenital conditions, moreover, are reported to be less likely to be represented in research involving disability (Bogart & Matsumoto, 2010). The involvement of people with disabilities in issues that concerns them, is something that the disability movement has long pursued (Charlton, 1997; Hurst, 2003). This thesis considers, both the biological aspects and the experiences of living with SB, by involving the adult persons with SB in the co-creation of knowledge.

Models of disability

The medical model of disability has been criticized for reducing all aspects of disability to bodily impairments (Grue, 2011) and for sending the message that something needs to be “fixed.” This leaves an impact on the individuals who are exposed to this “fixing” perspective. The social model has contributed to shifting focus from “fixing” the individual to “fixing” the surrounding factors, such as accessibility (Hughes, 2010). The social model could be criticized for focusing only, or overly much, on the surrounding factors, and not addressing the biophysical factors that cause the disability (Grue, 2011). Given the benefits of the social perspective on disability, it has been influential in, together with a strong and active disability movement in the 1960s, preparing a new reform in the disability area in Sweden (Askheim, Bengtsson, & Richter Bjelke, 2014), mentioned further under the next heading.

The biopsychosocial model is an attempt to look at disability from different angles, and could occasionally work for this purpose. It can, however, be criticized for lack of knowledge on the interactions between the biological, psychological, and social factors (Cromby, Harper, & Reavey, 2013; Van Oudenhove & Cuyper, 2014). This is also a matter of language and discourse, according to Grue (2011). Grue questions whether the medical model is even a model, suggesting that it is more of a series of local examples of medicalization. As such, it can be understood as a historical model reflecting some of the inhumane treatments and experiences of disabled persons in different health care institutions over the last few centuries. The

medical model gives power to medical professionals by favouring medical treatment and “normalization” as suitable interventions, and denying agency to persons with disabilities. By continuing to refer to medicalization as a model, the risk is to keep feeding the deception that it provides theoretically valid perspectives on disability (Grue, 2011). In Swedish research on disability, the Nordic relational model of disability often is used. This model contextualizes disability as a relation between the individual with disability and the surroundings. The fact that the model had an impact on research was partly due to the public debate on the situation of people with disabilities, and partly to the introduction of an environmentally relative approach in official policy (Lindberg, 2016).

Legal rights of persons with disability

While researchers and activists may navigate the theoretical views of disability, these views also influence the discourse and as such impact legislation and social policy (Grue, 2011), causing consequences for the persons living with disability. In 2009, a worldwide process of law reform in the field of disability was initiated (Quinn, 2009), including foremost the UN CRPD (United Nations, 2008). The drafting of the CRPD was influenced by the international disability community with many disabled people’s organizations in the drafting committee (Degener, 2016). According to Degener (2016), the CRPD is based on the human rights model of disability, which at the time was a new model developed from the social model on disability and guided by values such as inclusion, equity, and human dignity.

In Sweden, the CRPD was ratified in 2008 (Utrikesdepartementet / Ministry of Foreign Affairs, 2008), but it has not been adopted as Swedish law. Article 19 of the CRPD refers to the aim for all disabled persons to achieve independent living and be included in society. The countries that ratified the CRPD, are required to ensure that disabled people can choose where and with whom they live, with access to a range of services including personal assistance (Brennan, Traustadóttir, Anderberg, & Rice, 2016).

Swedish welfare policy is considered to have had its golden years between the 1950s and 1970s when the social security system was implemented. As community care services, and social care were expanded, criticism increased, especially from the disability organizations, the so-called “Independent Living Movement” at the forefront. Persons with disabilities

claimed the right to decide on their situation, rather than let “the experts,” with their “expert knowledge,” do this. The development has continued, with a new emphasis on the individual and his or her rights to freedom of choice, self-determination, and participation in society (Lindqvist, 2012). In 1989, the second disability investigation by the government led to new disability legislation, the LSS (Swedish acronym for *Lag om stöd och service till vissa funktionshindrade* (Support and Service for Persons with Certain Functional Impairments)) Act (SFS 1993:387), in which personal assistance is incorporated as one of ten different interventions for specific support. The other nine are: counselling and other personal support, companion service, personal contact, respite services in the home, short stays away from home, short periods of supervision for schoolchildren over the age of 12, living in family homes or in homes with special services for children and young people, residential arrangements with special services for adults or other specially adapted residential arrangements, and daily activities. The local authorities, the municipalities, were given the responsibility to implement this reform (Askheim et al., 2014). LSS was developed in collaboration with Independent Living Organizations (ILOs) whose goals and principles were implemented in law and policy. To be granted services, you need to fit into one of three categories, according to the LSS Act (SFS 1993:387):

1. Persons with an intellectual disability, autism, or autism-related disability.
2. Persons with considerable and durable impaired intelligence after brain damage in adult years, caused by external violence or physical illness.
3. Persons with other durable physical or mental disabilities, which are evidently not caused by normal aging.

Regarding personal assistance, the municipalities cover the costs up to 20 hours each week, and the national authorities cover the additional costs for the persons with the most extensive needs (Askheim et al., 2014). Granted support, according to the LSS Act, should be complemented by support from the Social Services Act (Socialtjänstlagen (SoL)) (SFS 2001:453) (Hultman, 2018).

Grey zone concerning legal rights

When considering legal rights to support in daily life, many adults with SB find themselves in a grey zone, even though the majority of adults with SB

who have cognitive impairments often have executive difficulties that affect the ability to be independent. Their impairments are often not included in the specific categories of needs, so they have no access to the support offered by the welfare society. According to the Swedish National Board of Health and Welfare (Socialstyrelsen, 2011), approximately 88% of these efforts involved persons who were granted support under the LSS Act (any of the ten interventions and one person could have more than one intervention) belonged to the above category one, i.e., they had a confirmed intellectual disability or autism. In the current population living with SB, the estimated number of people diagnosed with an intellectual disability, and therefore covered by the LSS Act category one, varies from 10% (Werhagen, Gabrielsson, Westgren, & Borg, 2013) to 20%, (Verhoef et al., 2004). The figures being based on an intelligence quotient (IQ) <70 following a traditional IQ test, which is mandatory in getting the diagnose intellectual disability. A similar cause for lack of access to support has been reported in a North American study involving adults with SB (Liptak et al., 2016). The literature suggest that IQ is a poor predictor of general aspects of everyday functioning (Arvidsson & Granlund, 2018; Nisbett et al., 2012). Those who have been diagnosed with an intellectual disability receive more support in their daily lives, such as entitlement to daily employment and personal assistance, as they fall under category one. This does not mean that it is unproblematic to get support even for this group. As Fridström Montoya emphasizes, there is no clear distinction between weaknesses in cognition and intellectual disability. This highlights difficulties that arise when the “legal entity” cannot bring their own legal action (Fridström Montoya, 2015). Some persons with SB receive support according the LSS Act category three, at times after the physical aspects have been aggravated by something that could have been prevented with support at an earlier stage.

Research connecting HC, Chiari II and other associated brain malformations in SB, with following cognitive impairment has increased (Dennis et al., 2010; Dennis et al., 2016; Iddon, Morgan, Loveday, Sahakian, & Pickard, 2004). Eighty percent of children with SB (Talamonti et al., 2007) and approximately 60% of the adult SB population have HC (MMCUP, 2020). Although this is well known, SB is still viewed as mainly a physical disability when it comes to qualifying for social support. This is a contributing factor to the abovementioned grey zone problem, not least since the consequences of an alternative diagnosis can result in benefits from society, leaving the individual with different prerequisites. This also

influences how SB is framed. Aronowitz (2008) addresses this situation by looking at social factors that determine presence of health, vs. disease, in the bodies of individuals, and at the way society recognizes, defines, names, and categorizes disease (disability) states and attributes them to a cause or set of causes. Since the naming of these sets of causes (diagnoses) results in various services and benefits from society, the outcome of the naming process is of high relevance.

Rationale

In summary, based on this background, the number of adults living with SB is increasing, there is an extended need for knowledge concerning this group, throughout the whole life span. There are needs for both to establish knowledge on the natural history of SB (Lidal et al., 2019; Webb, 2010) and to involve adults with SB and their lived experiences in the research, in other words, studying the experience of having lived a long life with a congenital disorder in Sweden. This is in coherence with the WHO World Report on Disability, recommending more utilization of qualitative methods to investigate the lived experiences of persons with disabilities (WHO, 2011). Traditionally, persons with developmental disabilities have either been excluded from research, or have been treated as research subjects, rather than being included as partners or collaborators in research projects (Ward & Trigler, 2001). As this group has experiences of disabilities, a rare condition, and cognitive impairments, each of which status has a history of social exclusion and lack of knowledge from within, it makes it relevant to incorporate adults with SB as partners in constructing knowledge concerning them.

Theoretical framework and concepts

The theoretical framework in this thesis derives from caring science, and the research is based on the standpoint that the person should be involved as expert on his or her own situation (Dahlberg & Segesten, 2010). The voice of the person has gained an increasing interest in health care and research. Today, emphasizing the person's story is thought to be an important aim in caring (Omerov, Kneck, Karlsson, Cronqvist, & Bullington, 2020), partly because of the benefits of creating a relation with, and understanding what is important for, the person and thereby supporting him or her on his or her path towards health. But also, because listening in an open and accepting manner is an act of care that communicates to the person that he or she is viewed as a person worth listening to (Omerov et al., 2020).

Health from a caring science perspective

A concept of high importance in caring science is health. Dahlberg and Segesten (2010) argue for an empirical and existential view of health, meaning that health is a multidimensional experience. More specifically, they suggest that "Health could be understood in terms of feeling good and being able to implement what you consider to be of value in life, both large and small." (ibid, p. 53. Author's translation). This could be understood as the experience of health being related to a general feeling of wellbeing and an interweaving of the biological foundation of the human being with the existential dimension of being in a world together with others. The biological foundation is expressed through signs of disease, and when having a serious disease and/or a lifelong condition, this can constitute an outer perspective. Based on the existential dimension of health, we know that, despite the presence of disease, an individual can strive for wellbeing, which can result in some sort of health perception. In this thesis, the focus on health is from the above described existential perspective, incorporating an inner dimension, meaning that even though you have an injury, disability, or illness you can experience health. Concepts from the above description of health are used in the analysis in Study II.

Study I mainly provide insights into the biological foundation and psychosocial aspects of everyday life with SB at a group level, by mapping the condition and surrounding issues in a cohort. Studies II and III and parts of Study IV elaborate the existential dimension of life with SB, individually and together with others.

Everyday life and lifeworld theory

The living conditions and everyday life of adults with SB in Sweden are the focus of this thesis, with particular attention to the lived experience. Lifeworld theory has high relevance in caring science (Dahlberg & Segesten, 2010). In lifeworld theory, it is understood that all our feelings, thoughts, and what we do are experienced through the lifeworld. The reflective lifeworld research (RLR) approach, used in Study II, is an offspring of phenomenological philosophy and deals with life experiences (Dahlberg, Dahlberg, & Nyström, 2008). The lifeworld is our everyday existence through which we live and experience our lives. According to Husserl, the father of the phenomenological tradition where the lifeworld of humans takes a central place, the lifeworld is perceived without reflection and forms the foundation for understanding human beings, their lives, health, suffering, and states of wellbeing (Husserl, 1984).

Merleau-Ponty's ontological philosophy, as discussed by Dahlberg & Segesten (2006), states that the individual cannot be separated from her or his world context, in the sense that the world is part of the individual as much as the individual is part of the world. They need to relate to each other as aspects of "flesh of the world" (ibid). To be able to understand the everyday existence of others, we need to take part in what their lives contain. In this thesis, this is done in Study I by mapping living conditions such as housing, occupation, and health status, at a group level. In Study II, the focus is on daily life and experiences of this daily life from an individual perspective. According to Todres, Galvin, & Dahlberg, (2007), the surrounding world involves places, things, and situations that have meaning for how we live our lives and also for our health context. This can be further understood by reading Galvin & Todres' posit that it is in the meeting with other people's lifeworlds that a person can understand both him or herself and others. It is in the context of the lifeworld, together with others, that meaning is experienced (Galvin & Todres, 2013).

Narratives as lived experience together with others

The above condition, *together with others*, was met by using a participatory approach in Study III (and Study IV, which builds on Study III). Here, members of the photovoice group were collaborators in creating knowledge of the everyday life of adult persons with SB by sharing their experience through stories (narratives) and photos. Storytelling, or story sharing, is a

vital form of communication through which people share experiences and knowledge with others as well as make sense of lived experiences themselves (Clandinin, 2013). Narratives in Studies III and IV are understood as meaning making, where stories are lived but also constructed, shared, and created together. In research, narrative is a way of studying experience as story, inspired by a view of human experience in which humans, individually and socially, lead storied lives (ibid). Ricoeur refers to narrative as an inherent structure of (linguistic) expressivity, through which we recognize and understand ourselves in the world and within temporality. Despite temporality, Ricoeur links narrative to action, agency, and identity (Ricoeur, 1984;1985). The story can be viewed as a portal through which a person enters the world and by which their experience of the world is interpreted and made personally meaningful (Connelly & Clandinin, 2006). Since before the story is told, it is lived, constructing the story is an act of shaping lived experience (Mattingly, 1998). Whereas lifeworld theory tend to place a stronger focus on body and experience as something very visceral, narrative theory tends to place a stronger focus on language and action as something co-constructed. Applying both lenses in this thesis allows for different aspects of experience and everyday life to be foregrounded.

A participatory approach

In this thesis, *participation* is used to mean more than just being involved in situations. It involves (a) active and meaningful engagement/being part of; (b) choice and control; (c) access and opportunity/enfranchisement; (d) personal and societal responsibilities; (e) having an impact and supporting others; and (f) social connection, and social inclusion and membership (Borell, Asaba, Rosenberg, Schult, & Townsend, 2006; Hammel et al., 2008; Hemmingsson & Jonsson, 2005). This clarification is relevant for the method used in Study III and further elaborated in Study IV, where the methodology of a participatory method is explored. The theoretical underpinning of the participatory method photovoice derives from emancipatory education, feminist theory, and documentary photography. Emancipatory education, as posited by Paulo Freire (1993), assists the understanding of how co-constructing knowledge can lead to enhanced power to make change. Feminist theory elaborates the causes and processes of oppressive behaviours with the aim to end oppression and the centralization of male power in knowledge construction (De Chesney, 2015). It is in the nature of co-construction of individual and collective stories, through both doing and representation, that participatory approaches such as photovoice and narrative theory provide valuable tools in design and analysis.

Overall aim

The overall aim of this thesis was to generate knowledge about living with Spina bifida (SB), by mapping the condition and together with adults with SB explore their living conditions and experiences in everyday life.

Specific aims

Study I – To describe health issues and living conditions in a cohort of adults living with SB.

Study II – To describe the experience of daily life for young adults with SB through a theoretical lens of health.

Study III – To actively integrate expertise of persons living with SB, used to explore conditions embedded in their everyday life.

Study IV – To explore the meaning of dialogue, action, and group interaction as techniques that have methodological relevance for photovoice; further, using an empirical case, to illustrate the function of these techniques to explore the possibilities of narrative in collective construction of meaning and social action.

Methods

The aim was met by combining quantitative and qualitative methods. The first study had a quantitative approach and provides a knowledge base for the contextual understanding of the condition of being an adult person with SB and of the elements in the everyday life of adults with SB, on a group level. A cross-sectional design was used for data collection in Study I and the results are presented using descriptive statistics.

Experiences of daily life at an individual level were explored by deep interviews in Study II. The interviews were conducted with a RLR approach, and analyzed interpretively. Study III had a participatory approach including five members in a photovoice group who met for eight photovoice sessions; the generated material was analyzed narratively. Finally, Study IV focused on reflections on the methodology in which photovoice is grounded. The studies will be described separately below.

Table 1. Overview of the studies in the thesis

Study	Participants and materials	Method	Data analysis
I	The study included 196 persons with SB, ≥18 years old. Structured interviews, questionnaires, and clinical assessments.	Quantitative. Cross-sectional.	Descriptive statistics.
II	Six persons (four men, two women) aged between 28 and 44 years.	Qualitative, deep interviews using the RLR approach.	Interpretive, using concepts of health.
III	Five members (four men and one woman, aged 30–49 years) of a photovoice group who met for eight sessions.	CBPR; photovoice.	Narrative analysis.
IV	Field notes and transcripts, concerning processes in Study III; previous literature related to photovoice studies as well as to photovoice theory and methodology.	Exploring methodological processes.	Methodological analysis.

RLR = reflective lifeworld research.

Overall context

The Spinalis outpatient clinic, at Aleris Rehab Station (Previously Rehab Station Stockholm), specializes in spinal cord disorders, and has for 20 years been responsible for follow-ups of health care related to the spinal cord disorder in adults with SB living in the Stockholm region. Having access to many eligible professionals in one clinical setting is a unique opportunity for follow-up of adults with SB. In the follow-up procedure, persons with SB are met by a multidisciplinary team. It is in this Swedish sociopolitical and health care context, and a well-resourced center, that this thesis should be understood, as all the participants in Studies I–IV were enrolled at this outpatient clinic.

Study I

Participants

In Study I, individuals aged 18 years and over with SB (n=219) were invited to participate. One hundred and ninety-six agreed to participate, 104 women and 92 men, aged between 18 and 73 years. Persons included in the study had a diagnosis of SB. All three types of SB were represented in the study group: MMC; lipomeningocele; and SB occulta.

Data collection

Study I was quantitative and was conducted by a multidisciplinary team. The research group at the clinic comprised a physician, a registered nurse, a physiotherapist, a neuropsychologist, an occupational therapist, and a social worker. The aim of Study I was to describe health issues and living conditions in a cohort of adults living with SB.

A cross-sectional design was used. The data collection was conducted in conjunction with the participants' regular follow-up at the clinic. The structure of the data collection was based on the structure of the regular follow-up, but extended and systemized. Data was collected during individual face-to-face sessions through interviews based on questionnaires and clinical assessments. The interviews included sociodemographic factors, medical information, and associated conditions. Medical records (including records from the children's hospital, computerized medical records, and paper records, depending on availability) were used where necessary, to validate the information given.

Measurements/instruments

Each professional (a registered nurse, a physiotherapist and an occupational therapist) who met the participants, used a questionnaire covering issues usually related to the subject of their professional role. The questionnaire was used as a guide by the professional, who asked, and made assessments with, the participant, and/or checked the medical records where necessary. The questionnaires covered the topics presented under the four headings below.

Sociodemographic factors

The sociodemographic factors are given in Table 2.

Table 2. Sociodemographic data

Gender
Age
Household status
Biological children
Passed core subjects in primary school
Main occupation
Driver's licence
Independence and support in daily life (support in personal care, household activities, and/or through reminders)
Financial guidance
Assistance provider (municipality/state and/or family member/s)
Transportation services

Structural and medical characteristics

The structural and medical characteristics include the following parameters, measurements, and instruments:

Bladder and bowel regimens were established using Nordic Spinal Cord Injury Registry (NSCIR) data (Levi & Ertzgaard, 1998), covering the method for bladder emptying, voiding frequency, continence, use of incontinence pads, and complications.

Data on neuropsychological diagnoses, including Asperger syndrome, attention deficit disorder, attention deficit hyperactivity disorder, as well as psychological diagnoses (depression and anxiety) and intellectual disability, was collected from the medical records and through the interview.

Self-reported sleep habits were collected using a modified questionnaire based on the Basic Nordic Sleep Questionnaire (Partinen & Gislason, 1995) and were categorized as difficulty falling asleep, waking up in the middle of the night and not being able to go back to sleep, or sleeping too long.

Table 3. Structural and medical characteristics (and method of data collection)

Diagnosis (I+M) Length, cm (A) Weight, kg (A) Hydrocephalus, Yes/no (I+M) Shunt and number of revisions (I+M) Chiari II malformation (I+M) Diagnosis (type of SB) (M) Epilepsy (I+M) Blood pressure (A) Allergies (I+M) Drug treatment (I+M) Secondary complications - Pressure ulcer (on examination day; presence and location) (A) - Orthopedic surgery related to SB (I+M) - Tethered cord symptoms (I+M) (earlier in life and/or in the last year) - Pain (on the day of examination; presence and location) (I) - Sleep habits (I) Bladder symptoms (I+M) Bowel symptoms (I+M) Psychological and neuropsychological diagnoses (I+M)
A = assessment; I = interview; M = information from the medical records; SB = spina bifida.

Physical function and assistive devices

Measurements and instruments for assessing physical function and use of assistive devices are presented below.

Table 4. Physical function and assistive devices (and method of data collection)

Muscle strength, five-graded scale (A) Level of motor and sensory function, AIS (A) Contractures >20 degrees in hip, knee, or ankle joints, Yes/no (A) Mode of mobility Transfers from the chair or wheelchair to the bed: Independent, with assistance, or with the use of a lift. (A) Weekly physical exercise habits: No physical exercise, moderate exercise, or strenuous physical exercise (I) FIM (A) Hand function (A) Assistive devices, Yes/no (A+I)
A = assessment; AIS = American Spinal Injury Association Impairment Scale; FIM = Functional Independence Measure; Interview (I).

The participants were assigned to different muscle function (MF) groups classified as I–V (Bartonek, Saraste, & Knutson, 1999). An additional classification, MF 0 (Bendt & Bartonek, 2016), represents those with no loss of muscle strength, and MF V represents those with no muscle activity in the lower limbs and no pelvic elevation, based on a muscle strength examination of the lower extremities (Hislop, 1995).

An American Spinal Injury Association Impairment Scale (AIS) score (Kirshblum et al., 2011) determines the level of lesion, and sensory/motor function. Contractures >20 degrees in the hips, knees, and/or ankle joints were registered (American Academy of Orthopedic & Surgeons, 1988). Functional ambulation was registered according to the criteria by (Hoffer, Feiwell, Perry, Perry, & Bonnett, 1973).

The motor function part of the Functional Independent Measure (FIM) was used to measure the level of each participant’s disability and to indicate how much assistance was required to carry out activities of daily living (Dodds, Martin, Stolov, & Deyo, 1993). Dexterity was assessed with the Nine-Hole Peg Test (Mathiowetz, Weber, Kashman, & Volland, 1985), and hand strength was assessed with Grippit (Nilsen et al., 2012). Weekly physical exercise habits were registered: no physical exercise, moderate exercise (a

minimum of 30 minutes, one to two times weekly), and strenuous physical exercise (a minimum of 30 minutes, at least three times weekly), based on the participants' own statement.

Cognitive subtests

The cognitive subtests used were the coding test, the block design test, and the F-A-S verbal fluency test. The coding test measures associative memory, graphomotor speed, and processing speed; the block design test measures visual spatial processing, visual motor construction, and problem solving (Wechsler, 2002); and the F-A-S verbal fluency test measures verbal executive ability (Tallberg, Ivachova, Jones Tingheg, & Ostberg, 2008).

Data analyses

In Study I, descriptive statistics are presented as numbers and proportions. The Shapiro-Wilk test was used to analyze normal distribution. Mean and standard deviation (SD) were used for normally distributed variables; median, interquartile ranges (IQRs), and minimum–maximum values were used for non-normal distributions. Differences between the groups and the presence or absence of HC were analyzed among the variables for cognition and dexterity using the chi-square test for dichotomous variables, Student *t*-test for variables with normal distributions, and the Mann-Whitney U-test for variables with non-normal distributions. Statistical significance was set at $p \leq 0.001$. The analyses were performed using SPSS version 24 (IBM Corp., Armonk, NY, USA).

Study II

Participants

Eight persons were invited by telephone to participate. Six participants, four men and two women, aged 28–44 years accepted participation. All participants used a wheelchair for ambulation, although some walked short distances indoors. All had treated HC. One lived with a partner, two lived with their parents, and three lived on their own, one of them with personal assistance. One had a full-time job, one worked part-time in a family business, one was studying, and three had neither a job nor any other form of employment.

Data collection

Study II focused on daily life on an individual level. The study had a qualitative design and the aim was to describe the experience of daily life for young adults with SB through a theoretical lens of health. Deep interviews were conducted according to the principles of RLR, with flexibility and openness to the interviewees' answers, to capture their lived experience (Dahlberg et al., 2008). The interviews were directed towards the meaning of the phenomenon of which the study aimed to gain an understanding (ibid). The interviews lasted 60–90 minutes, and were recorded and transcribed verbatim. All the interviews were conducted at the clinic, based on the participants' own choice. Each interview began with the question *Can you please tell me about a good day?* Follow-up questions like *What does that mean?* or *Tell me more about it* were asked to deepen the interview.

Data analyses

In the first phase of the analysis, a naïve reading of the transcribed interviews was done to get a picture of the whole. Both in order to handle the rich material and because of the aim to focus on health, a tool was used for the second part of the analysis. The concepts used in the analyses stem from an emerging theory of health by Dahlberg & Segesten (2010) used in Gabrielsson, Traav, & Cronqvist (2015), see Table 5.

Table 5. Concept description: an emerging theory of health

Health	Human health is the overall goal in caring, including supporting and strengthening people’s health processes both biologically and existentially.
Existence and wellbeing	Biological health is intertwined with the existential experience.
Finding a place in existence	Living in the world in a way that both affirms the own way of being and an existential context.
Vitality	Life force is essential to be able to live a life full of obstacles and opportunities. Support and strengthen viability.
Life rhythm	Possibility of movement in life. An ability and sense of “being able to.” Life rhythm also includes stillness and rest.
Meaning and context	Human longing for meaning and context and how this desire is connected to health, suffering, and illness.

The concepts were used as codes utilized for grouping the text into categories. Codes with corresponding meaning units were brought together and constituted the material for the next level of analysis. A hermeneutic procedure was applied to gain an understanding of the whole in relation to the parts (Dahlberg et al., 2008). Approximately ten text units were selected and further analyzed, resulting in five themes and an overarching main theme.

Studies III and IV

Photovoice

Study III uses the method photovoice and Study IV explores the method’s inherent methodological processes. Photovoice is collaborative, action-oriented and is considered to belong to an approach in compliance with CBPR (Asaba, Laliberte Rudman, Mondaca, & Park, 2015; Wang & Burris 1994; 1997). A central tenet of photovoice is its characteristic of valuing and honouring knowledge of everyday people and everyday communities, highlighting what the community in focus deems of importance and not necessarily what the researchers may have predetermined to be important. In photovoice, the aim is to engage people

as experts on their own life situation through dialogue, narratively reinforced by photographs, which requires a clear commitment to involve people who, in different ways, live on the margins of society (Plunkett, Leipert, & Ray, 2013). Photovoice can be distinguished from other methods where photos or pictures are used, in that members of a photovoice group actively make decisions about what photos to generate and how to generate them, as well as being active in co-designing and revising the questions posed. In photovoice, “VOICE” as an acronym is intended to convey “Voicing Our Individual and Collective Experience” (Wang & Burris 1997, p. 381).

Participants

The original recruitment aim was to gather a group of between six and eight persons including both men and women. The final group consisted of five persons, four men and one woman, aged 30–49 years. Of these, three lived alone, one of them with 24-hour assistance, and two lived in serviced apartments. Two worked part-time and received a subsidy; two had sheltered work; and one had early retirement. Four of the group’s members used a manual wheelchair and one walked. Four were Swedish-born; one had moved to Sweden at around the age of 4. The size of the group was in keeping with photovoice methods and allowed for active dialogue among all group members (Asaba et al., 2015; Wang & Burris 1994; 1997).

Data collection through photovoice

In accordance with the methodology in photovoice, photographs served as a starting point for the discussion about what was of interest in daily life. The group met once a week for 8 weeks, for sessions lasting 2 hours. A theme, formulated by the group each week, served as a basis for the photos members took for the next session. The group agreed to bring in three photographs each per week, to work with during the sessions. The data consists of transcriptions from the sessions where stories, reinforced by photographs, were dialogued and co-constructed, as well as the processes within the project.

Narrative analyses, Study III

Phase I of the analysis actively involved the group through the dialogues and the sorting of photos and stories connected to the photos, in a process whereby the group was part of the visual thematic analysis presented in the exhibition. Phase II consisted of the narrative analysis conducted by the researchers.

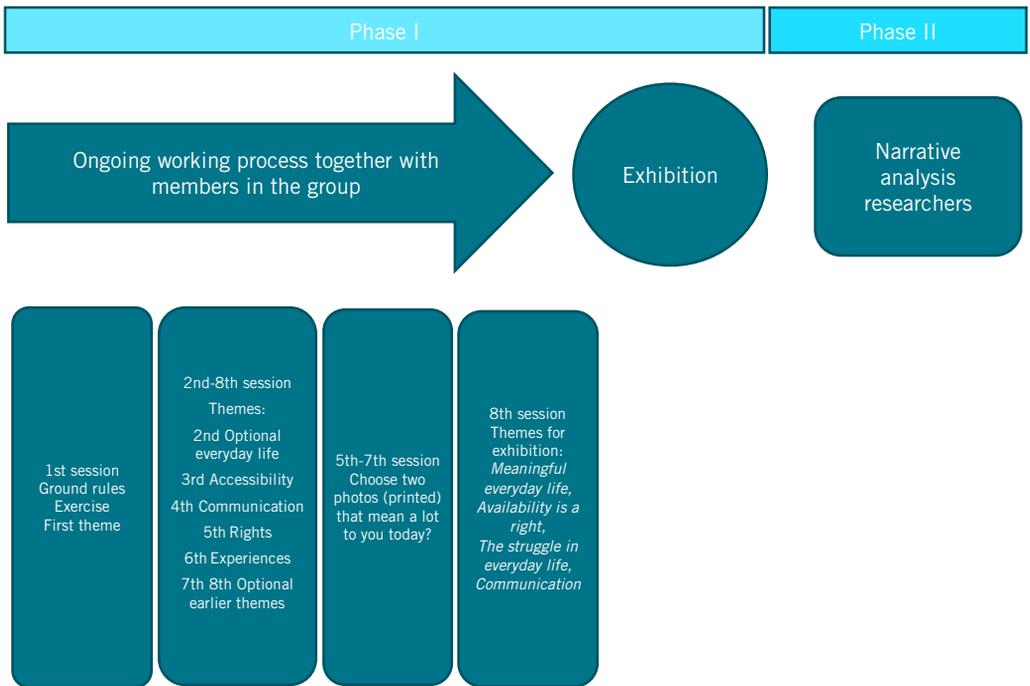


Figure 1. Flowchart of the research process

The narrative analysis was built on Phase I, and was based on transcripts, photographs, and field notes from the eight sessions. The initial reading of transcripts focused on identification of significant events and turning points in the actions and happenings provided by the data, to generate a story (Polkinghorne, 1995) on a group level. Significant events might be something distinctive, representative, or surprising. Surprise could include a finding that was contradictory to previous material, either from the intra- or interparticipant research data, or based on variation from within the literature. Altogether, this created material for the emergent narratives. Caution was taken to retain the wholeness of the narrative, rather than to fragment stories and experiences into parts or categories (Creswell & Poth, 2017). The themes were discussed back and forth among the researchers to reach consensus. The narratives generated and presented in Study III were

co-constructed by the participants and the researchers, even if the final analysis in Phase II was conducted by the researchers.

Methodological analysis, Study IV

Photovoice has undergone methodological development and is being increasingly used; yet concerns regarding the method have been raised. In Study IV, issues with and adaptations of the methods used, and how they affect methodology, are discussed and reflected against the theoretical underpinnings on which photovoice rests. By returning to the ideological cornerstones of photovoice – emancipatory education, feminism, and documentary photography – the empirical experiences from Study III and examples from the literature are elaborated through processes within photovoice.

Ethical considerations

The three empirical studies in this thesis were conducted in accordance with the Declaration of Helsinki (World Medical Association, 2018). The ethical principles of benefit for others, justice and respect were adhered to by performing actions that promote wellbeing in a person, respectfully listening to the other person's view, and recognition of the person's dignity (Beauchamp & Childress, 2013), for example by obtaining informed consent for research in a fair manner (Judkins-Cohn, Kielwasser-Withrow, Owen, & Ward, 2014).

Informed consent

As this group had cognitive impairments of varying degrees, special considerations were taken when explaining the purpose of the research project and the possibility to withdraw the consent at any time. The information regarding each study aim, what was required from the participants, voluntary participation, and how collected data would be used, was provided orally and in writing. One aspect of importance was whether the members understood what they agreed to be part of. The participants were encouraged to ask if there was any information or question that appeared unclear. When the nurse researcher has the dual roles of clinical nurse and researcher, it is especially important to explain to participants that their participation is voluntary and does not influence the provided care (Judkins-Cohn et al., 2014).

The informed consent process in the photovoice project (Study III) was repeated continuously through the study, as was the possibility to withdraw from the project at any time. Since the group decided to share the results from the study through an exhibition, they also agreed on breaking anonymity. This was given special attention, by discussing the potential risks and benefits of doing so (Judkins-Cohn et al., 2014). The members of the group signed another informed consent regarding the exhibition. Another aspect of importance, when it comes to ethical concerns in a photovoice project, is whether other persons are visible in the photographs. This issue was handled continuously by asking permission from the person visible in the photo, for using the photo in the project. When the photographs for the exhibition were chosen, the facilitators made sure that if an outside person was visible in a photograph, he or she was asked for permission to have the photo shown publicly, to secure that person's right for anonymity and privacy.

Confidentiality

Collected data were handled in such a way as to protect confidentiality. Personal data were coded and were only available to the research group. The data is stored in a locked document cabinet at Aleris Rehab Station (Previously Rehab Station Stockholm), and digital data files are stored on a secure server protected by a password.

The data from Study III used in the scientific report has been anonymized, meaning that breaking the anonymity was primarily for the exhibition. This can impact the possibility to be recognized as a member of the group also in the scientific report. This issue was discussed within the group, when handling questions that arose regarding the decision to create an exhibition.

Ethical approval

Studies I, II, and III have been approved by the Regional Ethical Review Board in Stockholm (dnr. 2014/1111-31, 2010/761-31/5, and 2017/992-31/2). Study IV is a methodological and conceptual analysis of empirical data from Study III, and material in Study IV is covered by the initial approval for Study III.

Findings

A summary of the main findings of Studies I–IV is presented below; thereafter, an analysis of these findings will be presented under “Reflections on findings.”

Study I

In Study I, data are presented for the total cohort (N = 196) and separately for four age groups (Group 1: 18–30 years, n = 90; Group 2: 31–45 years, n = 60; Group 3: 46–60 years, n = 38; and Group 4: ≥ 61 years, n = 8). The participants had a median age of 33 years, 37 years for women (n = 104) and 30 years for men (n = 92). There was a significantly higher proportion of women in the two older groups. Those who were ≥ 46 years old had less complex medical conditions, and better physical and cognitive function, and had attained a higher level of education. Those who were < 46 years old had a greater number of secondary conditions such as HC, Chiari II malformation, tethered cord symptoms, and latex allergy. In the whole group, 7% lived in sheltered housing, 41% in a single household, and 31% with parents or a friend/friends; 3% were single living with a child or children, and 18% were living with a partner (with or without children). Sixteen percent had biological children. Forty-four percent of the study population received municipal or state assistance; of those, almost half (48%) relied on additional help from relatives. Twenty-two percent had a full-time job, while 15% had part-time work, and 8% worked in a protected workshop. Nineteen percent were unemployed and 20% were in school or other education, 16% were pensioner. Seventy-eight percent had urinary incontinence and 77% had fecal incontinence. The majority (54%) used CIC for bladder emptying. Seventy percent underwent the cognitive assessment. There were no significant differences in age, gender, or prevalence of HC between those who underwent the cognitive assessments and those who declined. The participants performed one SD below the reference values for the general population on the cognitive sub-tests, with significant differences ($p < 0.001$) between those with and those without HC.

Study II

Five subthemes were formed in Study II, the first of which was “Not understanding and taking responsibility for the lower body” and was related to the participants’ struggle to understand, and react to, signals from their body. “Having had people stand behind me – not having been allowed

to grow up myself,” as the second subtheme was called, included feelings of being pitied and receiving help without being asked, a lack of challenges, and constantly having someone at hand to help, leaving the participants feeling overprotected and unable to get things done on their own. The third subtheme, “Compared to people like me, I usually do well,” reflects a twofold experience of meeting others in a similar situation as oneself. On the one hand, it was a source of fellowship through sharing experiences; on the other, looking back on childhood experiences uncovered an ambivalence about socializing with others with SB. This partly had to do with the discovery that they were not being treated fairly by the environment, as the following quote shows: “... *They get an image of the world that is a bit wrong, I think [...] those kids were treated in a way that I did not actually like ... I thought that was so hard to watch.*” The fourth subtheme, “I thought it would work out by itself,” expresses a desire for independence and shows the many challenges on the way to getting there. These often were physical barriers, but participants also described the difficulty to handle situations by, for example, asking for help in time, when not being able to fix a problem on their own. Such situations might be directly related to health, or they could involve an activity like doing the dishes, washing, or preparing daily meals. The last subtheme, “A lack of structure in daily life,” describes the lack of a natural rhythm and balance in life created by unemployment, and the isolation following this. Those who had employment described the job as a platform for social contact and as providing the feeling of context. Having routines and a pattern in life was considered important. The feeling of being in a context could also be achieved by being with family or friends, or going to the gym or attending another form of organized exercise. The five subthemes were merged together under the main theme “The contradictory path towards wellbeing in daily life.”

Study III

In Study III, the findings are incorporated with the discussion and presented with an overarching theme that was identified in the members’ experiences in everyday life, namely, that many solutions in society are “An adaptation for us, but it works for no one.” The members’ stories showed that many of the systematically developed services and adaptations did not work and contributed to a *struggle in everyday life* and a sense that the design of services had not included any people living with different kinds of disabilities. One of the stories presented under this theme involved a photo

of a closed door, representing the inaccessibility to an absent supervisor and a narrative about a job to attend, but not really to participate in. This example of suboptimal work tasks, in terms of personal meaning, has been raised as an example of structural hindrances that need to be considered in reconceptualizing the idea of work in the context of disability (Asaba, Aldrich, Gabrielsson, Ekstam, & Farias, 2020). The findings are further presented under three themes: “Accessibility – a never-ending project,” “Tensions of a normative view” and “Power to influence.”

The theme “Accessibility – a never-ending project” was about dealing with accessibility in everyday life. This included physical barriers at work, such as a broken elevator and a supervisor not really taking the problem seriously and fixing the elevator in a timely manner. A photo of a broken seat belt initiated a dialogue on whether having to choose between safety and actually having transportation. Participants described that at times this meant either traveling unsafely to get to an appointment on time, or traveling safely but arriving late. The next theme, “Tensions of a normative view” was likewise illustrated by a photo of a closed door, where the door had a metaphorical meaning as the members felt they met with closed doors everywhere, when for example talking to the authorities or traveling somewhere. The communication from their surroundings affected the members. As a physical example, the closed door without any possibility to open it, or stairs but no elevator or ramp, communicated a lack of welcome and made the participants question whether they were desirable customers or not. This brought up the question of being valued vs. being devalued. The participants also shared experiences of having to handle degrading comments, which were either direct or indirect, in personal communications.

The last theme, “Power to influence,” is about opportunities for participation by being asked about your views. Participants shared their memories from childhood, describing early experiences of not being asked, not even in situations concerning their health or their own body. They expressed the desire to be independent and to participate on their own terms where this was possible. This was, as has been mentioned, sometimes complicated by people around them trying to be helpful. Not being asked and not counting led to a sense of social exclusion and of not being part of a context in mutual respect.

The exhibition

Part of the results from the first phase of Study III (see Figure 1) were communicated with the surroundings through an exhibition. The first opening of the exhibition took place on World Spina Bifida and Hydrocephalus Day, October 25th, 2018, at the Spinalis clinic, Rehab Station Stockholm (now Aleris Rehab Station). All the members of the group as well as local politicians and members of patient organizations were present and were part of a public conversation that followed. This event served as an initiation of dialogue about everyday life with SB, and as a platform for meetings that would otherwise not have taken place. The exhibition toured during the following year and has been shown in ten separate locations including a university college, university, rehabilitation centers (one in Sweden and one in Norway), a library, a hotel, and patient organizations. The members of the group were active in suggesting locations for the exhibition and many of the members have been present at the various openings of the exhibition. The exhibition has received some attention in the media, including an article in *Disability Policy* magazine (*Funktionshinderpolitik* in Swedish), another in a local newspaper (named *Akershus Amtstidende*) in Nesodden, outside Oslo, Norway, and a report on the local radio in Stockholm, Sweden.

Study IV

The function and meaning of dialogue, action and interaction in relation to co-constructing knowledge are elaborated in Study IV. The concepts are argued as being unique strengths in photovoice methodology and that these strengths are possible to build on in studies involving persons with SB. By active involvement, encouraged by the methods used, interactions in the group (Study III) became self-sustaining and thus increased participation. The repeated group sessions were an important prerequisite for involvement and the will to develop, and for relations to grow and be interrelated with trust, as a base for dialogue and story sharing. The group discussions and group interaction provide an arena for the dialogue and reflection to develop, including action, as in “doing together” with others. Storytelling occurs in the process of group discussion, where both individual and collective experience is shared and created (Wang & Burris, 1997). By focusing on action and narrative, it becomes evident that dialogue, action, and interaction are important aspects of photovoice projects. Drawing on these aspects provides an arena for storytelling and story making, which have not previously had an explicit part in photovoice.

Reflections on findings

This thesis aimed to generate knowledge about everyday life with SB as an adult. The knowledge was generated, in collaboration with adults with SB, by exploring their living conditions and experiences in everyday life. The reflections are presented in four themes, the first of which is a synthesis of the findings in Studies I–IV. The findings are reflected by returning to the concepts of relevance from the theoretical framework guiding this thesis.

Lack of opportunities for independence and participation

The findings in Study I indicate that participants aged 46 years and over, the “survivors,” had better prerequisites for living independently and participating in society. They had less complex medical conditions, as well as better physical and cognitive functions, and had attained a higher level of education. Those under 46 years appeared to have lower prerequisites for living independently and participation in society at a younger age, indicating a need for social support in the future. This suggests a connection between bodily and cognitive aspects, on the one hand, and participation in society, on the other.

The results in Study II reveal another dimension of independence and participation, where the surroundings played a decisive role. The theme in Study II, “Having had people stand behind me – not having been allowed to grow up myself,” showed that having had support from others had negatively impacted on independence and possibilities to learn by making mistakes. Similar results were evident in the theme “Tensions of a normative view” (Study III), where a desire for independence and participation on own terms was at times opposed by people in the surroundings who were being “helpful” without asking. One dimension here concerns the way the surroundings view a person and his or her capabilities, which is constantly present in these situations. The theme “I thought it would work out by itself” (Study II) describes challenges in getting things done, and self-expectations not being met. The participants described how the desire for independence was dampened by experiences of pacifying events affecting both household, health and confidence. Taken together, the living conditions concerning independence and participation were affected by internal and external factors, leading to experiences of pacification, social exclusion, diminished health (through participants not taking care of themselves), and feelings of failure. It is important to acknowledge the lack

of opportunity for independence and, consequently, participation, shown by the members' experiences. This lack of opportunities stands in contrast to the values elaborated through a participatory method (Study III). Building on this, the meaning of participation was further developed in Study IV.

Stories of not being asked affecting identity

While some of the stories embedded in this research show a lack of opportunities for participation in everyday life, other stories describe experiences of not being asked concerning issues regarding the participants themselves. In both Study II and Study III, experiences of receiving help without being asked were described. The condition of not being asked was part of the participants' lifeworld. Similar findings were reported in a study of women with SB, which explored the lived experience of anger using focus groups (Daalen-Smith, 2014), where anger was linked to the women's treatment by the health care system, the educational system, and by society in general. Especially one quote from the article, describing painful medical procedures that were done without explanation, and further describing being paraded naked in front of medical professionals without consent (Daalen-Smith, 2014), was highly recognizable to the participants of our study, who told a similar story under the theme "Power to influence" (Study III). The story revealed another dimension of not being asked as the member of the photovoice group looked back on a memory from childhood, an exposed situation when, at 8–10 years old, he found himself lying on a mattress with a group of medical students sitting next to it, who turned him and strapped down his legs, and so on. No one had asked him if it was ok for the medical students to attend the medical procedure. Drawing on Mattingly's view on narrative as a way of constructing experiences (Mattingly, 1998), but also as a way to reconstruct the view on self, this early condition of not being asked, and not being heard, not only leads to the story construction itself and to the possibility for the person's story to be constructed at all, but also affects the person's view of self. The potential power in story and voice is further put into words by Frank (1995), who writes:

Seriously ill people are wounded not just in body but in voice. They need to become storytellers in order to recover the voices that illness and its treatment often takes away ... When any person recovers his voice, many people begin to speak through that story. (pp. 12–13)

Language also plays a crucial role as an important aspect of the social environment, since the language we use reflects our view on the phenomena in focus. According to the developmental perspective, young children are socialized in language, as well as through language (Nelson, 2000; Ochs & Capps, 2001). Language provides the basis for a self-identity to develop in childhood (Piaget, 1951). This includes the language provided by the surroundings, e.g., health care, which has been an actor of importance by providing a language to the families with a child born with SB, and, as time has gone by, to the adult person with SB. Holland, Lachicotte, Skinner, and Cain (1998) view identities as primarily being constructed through interactions firstly with the social environment, and thereafter on a more intrapersonal level. The environment, and the contexts, in which people express their identities are not neutral. Rather, they are socially charged by discourses and ideologies shaping the boundaries which continuously reposition the individual (Holland et al., 1998). This is in consistence with the results of both Studies II and III, where others in the members' lives and in their actual physical surroundings communicate values. These are communicated both through direct language (addressed to the person with SB) and indirect language (used in the treatment of, and language used for, others with SB or any other disability), and by the physical surroundings (through being accessible or not). Yet another view on identity is that it is not only affected by social discourses, identity is also expressed and reconstructed through everyday doing situated in a particular social-cultural context (Asaba & Jackson, 2011). This is of relevance in relation to the result in Study I, II and III, showing low employment rate and a longing for meaningful employment and for being in a context.

In light of the above, the story of “not being asked” leaves the person with SB in exclusion and ignorance and positions him or her in the box of incapability, all of which affects surrounding attitudes and personal identity. A similar conclusion was reached in the study involving women in focus groups to explore anger (Daalen-Smith, 2014), where the experienced oppression was linked to ableism, including constantly being compared to others, devalued, made to feel unimportant, and not being listened to or taken seriously, with little or no agency in their lives. This treatment of being made to face inaccessibility both in the surroundings and in communication, by not being asked, affects a person's self-image and opportunities to make the own voice heard. There seem to have been few opportunities in the past, for persons with SB, to be heard, and therefore

they have had little practice in putting experiences into words. A lifeworld understanding always involves the relational contexts in which persons act, and how these relational contexts have the possibility to change them (Todres, Galvin, & Dahlberg, 2014). One possibility for change and healing is to “reach for insidersness,” as its absence can become a form of iatrogenic suffering (ibid). Another possibility can be to enable persons to feel part of a larger whole, and being given the opportunity to try out alternative identities, through engaging in occupations and actions (Asaba & Jackson, 2011). This can be enhanced by policies that create inclusive employment opportunities.

Stories as action and ability

By being listened to, the ability to tell the own story, in itself an action, is communicated. As Mattingly puts it, if there is a story, there is action and you cannot understand action without stories (Mattingly, 1998). Stories involve action and experiences, involving someone trying to do something, and what happens to her and to others as a result. Narratives are not merely about experiences, but in a sense experiences are about narratives. Action and experience are both personal and socially constructed (ibid.). Narrative, in itself, can be a way of reconstructing or rewriting the own understanding of life and self (Alsaker, Bongaardt, & Josephsson, 2009; Mattingly, 1998). Consequently, participation in the photovoice project (Study III) constitutes an opportunity for change on an individual level by members reconstructing the view of self. Via members’ participation through story and action, as described in Study III and further elaborated in Study IV, the relationship of story and action becomes evident as an opportunity to construct stories together with others. Apart from story, action in this context is intimately related to dialogue and group interaction as a foundation for reflection. The members of the group performed actions individually in between the sessions (photographing their everyday life) as well as together by visual thematization of the meaning of the photographs. The thematized photographs later formed an exhibition, which served as a space to start a dialogue concerning issues of relevance for the group. In the language used in photovoice, this is action as in promoting social change. Everybody has a voice; at times, the question of importance is whether anyone is listening, and who? The opening exhibition served as an arena for dialogue between stakeholders (local politicians), the members of the group, and members from patient organizations. Thereby, the voice of the group as experts on their lives was shared and illuminated in a direct and clear manner.

As this thesis reports lack of possibilities for participation and possibilities to be heard and to put experiences into words and, thereby, construct valuable insights on life, the photovoice project constitutes an important experience of participation. Descriptions of the attitudes of the surroundings, in Studies II and III, showed questionable and excluding behaviors, which often led to feelings of powerlessness. Powerlessness can be viewed as a result of being objectified and not believing that the own participation makes a difference (Wallerstein & Duran, 2008).

The meaning of participation was elsewhere explored among persons with disability (Hammel et al., 2008). Participation was viewed as a right predicated on access, opportunity, respect, and inclusion. According to the authors, it includes a personal and societal responsibility requiring determination, advocacy, and empowerment, as well as a means to experience social connectedness with other people and communities. Developing advocacy, control, and power is part of the process. Power, on either an individual or a group level, was also related to societal access and opportunity (ibid). The photovoice project confirmed some of these meanings of participation, while the stories of everyday life showed a clear absence of such participation. Even in situations such as being employed, which suggests being part of a social context and participating in society, participation was lacking (Study III). Absence of opportunities for experiencing participation in everyday life equates to absence of an opportunity for health and wellbeing through being part of, and being in a context, and act (Dahlberg & Segesten, 2010).

Action as health

The concept of health in caring science involves more than biological functions. Biological health is incorporated in the existential experience of health, including meanings of being in a world together with others. Health is also understood as a sense of “being able to,” concerning different aspects of life (Dahlberg & Segesten, 2010). Health, when it is viewed as an ability to act, implies that the opposite, i.e., ill health, represents a kind of failure of action. “Action” is here understood in the sense of an intentional action, including “ordinary doing” (Nordenfelt, 1995). As elaborated from the action theory on health in Study IV, the environment can either make action possible, or the opposite. To be healthy is to be able to act (and/or pursue the own goals) provided that the environment is accepting. The environment is expected to be conducive to enabling people to act (ibid).

The environment can in this sense be viewed as an opportunity, afforded by surrounding factors, for a person's possibility, affecting the person's ability (Nordenfelt, 2009). The photovoice project can be seen as such an opportunity, in a supporting environment that positively affected the members' abilities. While this was done in a small context, during a limited time period, with the possibilities to create a supportive environment, it should be remembered that the lack of a supportive environment in everyday life is a reality, and that the consequences both for the possibility for actions and the ability to act can affect a person's health.

Another important base for a person's ability to use acquired abilities and competencies is motivation, by which is meant the ability to form intentions and act on them (Nordenfelt, 2009). Motivation is viewed by some as energy, drive, or will (Tengland, 2020). Energy, drive, and will also constitute the executive functions (using a more general definition), which in persons with SB are typically affected as a consequence of cognitive impairments. These known impairments were reasons for concern regarding the possibility of conducting a photovoice study (III), which in the end turned out to exceed the researchers' expectations, by increasing the motivation and will of the members. The positive aspects of being in a group contributed to activities that the group accomplished together by members using their abilities together, which resulted in something larger than individual abilities and accomplishments.

Everyday life is existence as we know it, and can be understood as something we are familiar with as an obvious arena for happenings in life (Dahlberg & Segesten, 2010). The biological reality for persons with SB, a congenital condition, can be viewed as another dimension of accessing the world, the existence, through a body "as we know it." One aspect of living with a congenital condition is that it involves no experience of losing functions. This could of course happen at some point during life, but unlike many models of adaptation to disability, which refer to acquired disability involving grieving over lost functioning (Bogart, 2014) and getting to know a "new" body and a new situation, in SB there is no coming to terms with losing something that was previously taken for granted. More importantly, having a congenital condition seems to be linked to aspects in history of having been exposed from early on to different views on disability, affecting treatment, health care, housing, whether being included in regular education, having an occupation, and so forth. Looking back at the results

of this research, especially Studies II and III, the quotations conveyed a sense of accepting things as they were, for example in statements involving bodily functions. The suggestion was that these situations were familiar, well known, and taken for granted among the members of the group. On the other hand, what was explicitly spelled out during the interviews and sessions was a wish for a meaningful context filled with meaningful actions. As reflected above, this should be manageable with the right support and suitable actions.

Methodological considerations

This section presents the weaknesses and strengths of this research project regarding design and how the research was conducted. The combination of quantitative and qualitative methods is considered a strength. This made it possible to highlight different perspectives: the perspectives of the group level, those of individuals, as well as perspectives gained through interaction in the group. Having a variety of methods, besides offering different perspectives on a research question, also reduces the risk of errors linked to a specific method (Patton, 2015). On the other hand, using different approaches may have contributed to a lack of deepened knowledge about one specific approach, which could have been achieved by using the same approach twice.

Some of the limitations in each study regarding the methods used will be briefly discussed below. My objective throughout has been to offer detailed descriptions of the methods, contexts, research process, and methodological limitations, to enable the reader to evaluate the results and possible applications. The small sample size in Studies II and III can be viewed as a limitation. However, qualitative evaluations refer to integrity of data, such as rich data, thick descriptions, and a balance between meaning and research interpretations as part of trustworthiness. As such, trustworthiness involves more than sample size (Williams & Morrow, 2009).

Validity, reliability, and generalizability

In the quantitative cross-sectional study (Study I), concepts of validity, reliability, and generalizability are of interest (Polit & Beck, 2010). The findings are based on the clinical assessment interviews and assessments collected during individual face-to-face sessions with the persons with SB. The questionnaires used were developed in cooperation with multi-disciplinary staff from the clinic to strengthen reliability and validation. Parts of the questionnaire were already in use in regular follow-ups of persons with SB. In moments of uncertainty regarding specific issues, medical records including those from the relevant children's hospital, computerized medical records, and paper records, depending on availability, were used for validation.

Meeting the persons with SB face to face is considered a strength, compared to the alternative of using self-reported questionnaires. Meeting three

professionals who are interviewing and performing assessments can be exhausting. The cognitive tests were therefore performed on a separate occasion to diminish the consequences of potential fatigue. At the time of starting the project, there were no existing validated instruments for this group. Some of the chosen instruments are validated for persons with spinal cord injuries or the general population.

Since the findings are based on an SB population in Stockholm, with a specific structure for follow-up, it is not certain whether the results can be generalized to other cultural contexts or other disabilities. However, it can be assumed that some of the findings are generalizable to SB populations (and other disabilities) in other areas in Sweden and some of the elements to other geographical areas in the world. Since Study I is a cross-sectional study, the results for this study are valid only during the time of data collection. Study I generated a large set of data, some of which are still not reported. In the analysis of the data, new questions arose, making it clear that some issues were not included, which would have been desirable. This knowledge is valuable when planning studies ahead. Had we reported more of the collected data, the effort of reporting so much information in one article might have led to losing a deeper understanding for certain parameters.

Aspects of trustworthiness and credibility

In Study II, openness was strived for as one of the important aspects of RLR. This applied throughout the data collection, starting with the question posed, since an answer is always shaped by the content and meaning of the question (Dahlberg et al., 2008) and, as such, is important for both interviewing and discussing validity in scientific research (ibid). In Study II, the trigger question was, *can you please tell me about a good day?* and follow-up questions depended on what the person said. In Study III, the goal was to involve the participants in shaping the questions that were formulated. This was in accordance with a wide aim, stated at the beginning of the research process, following the recommendations in photovoice (Asaba et al., 2015). The said aim was formulated into the question: *How do persons with SB experience opportunities and obstacles in everyday life?* which was communicated to the members of the group as *We are interested to know more about your everyday life.* Thereafter, the questions were formulated by the group, as themes, based on which they photographed and shared their experiences. The above illustrates an effort to be compliant

with the persons with SB and their expertise. An ideal situation in Study III, following photovoice methodology, would have been for the group to introduce the project from the start, bringing the research question along (Wang & Burris, 1997).

Since the members in the photovoice group were active in the first phase of the analysis, a check off was continuously done in the process, where the facilitators summarized what the group had said during the session to see if the group agreed, and if not, the group adapted the summary where needed. This also reflects the collaborative relationship between the researcher and the members of the group, both in the narration and in reflexivity (Clandinin, 2013) and the dialogical interpretation which is important in photovoice (Plunkett et al., 2013). The second part of the analysis was, however, not checked with the members of the group, since that analysis is considered as the researchers' own. The members were asked repeatedly during the project if they, as a group or individually, were interested in being part of the academic report, but no-one chose to be part of this process. Since there were eight photovoice sessions, there was time for relations within the group to grow, and for the stories and data to thicken and get rich (Williams & Morrow, 2009). In Study III, we worked actively with participation through communications methods intended to develop relations of trust and open communication, elements known to increase rigor and trustworthiness (Lennie, 2006).

Reflexivity

While the researchers in quantitative research traditionally strive for objectivity, researchers in qualitative research increasingly view truths as multiple and value the subjectivity of both researchers and participants (Williams & Morrow, 2009). This has been described as being on a continuum with the post-positivism view emphasizing objectivity on the one hand, followed by interpretivism, constructivism, social constructionism, and critical/ideological and postmodern/post-structural theories on the other, giving emphasis to the values of the researcher in the research process (ibid). With this view in mind, my roles and reflexivity in Studies I–IV have been diverse along this continuum. During data collection in Study I, I met the persons with SB as a clinical nurse. Once the data was input into Excel, I could start to practice objectivity in relation to the data. In Study II, I was guided by principles for lifeworld research, including going to the “things themselves” by being observant, and attentive and sensitive (Dahlberg et al.,

2008) to the experiences shared by the participants. My preunderstanding was handled by *bridling*, meaning trying not to understand too quickly or carelessly, waiting for the phenomena to present themselves to avoid making definite what is not definite (ibid). This was strengthened by reflections and discussion on the findings between the co-authors. In Study III, my role shifted from being facilitator, to being observer, and then to being part of the group. As a facilitator, you are observing what is happening in the room; at the same time, you are a part of the group and of what is going on in the process. To minimize the risk of the voices of the facilitators becoming too strong and impacting negatively on ideals of participation (Asaba et al., 2015) in the research process, reflections were conducted by the facilitators after each session. These reflections had an emphasis on what the group seemed to find important and worked as a validation strategy, to secure the focus on the group's voice. By encouraging voices in an ethical relationship with research members and raising the level of awareness of the members of the photovoice group and other stakeholders, encouraging action and being self-reflective, Study III (and also Study IV) offers authenticity and therefore validity, as described by Lincoln, Lynham, & Guba (2011).

Considerations in analyses

In the analysis of Study I, the descriptive statistics were discussed with co-authors who had expertise in statistics. As Study I had a response rate of 89%, a sample size calculation was not relevant. P-values were presented for six variables that were considered to be relevant, with statistical significance. There were no significant differences in age, gender, or prevalence of HC between those who underwent the cognitive assessments and those who declined.

The interpretative analysis in Study II was a departure from the analysis in RLR. After the first naïve reading, concepts from an emerging theory of health, described in the “Methods” section in this thesis, were used. The theory was selected with the study's aim, of describing the experience of daily life for young adults with SB through a theoretical lens of health, in mind and after reading through the rich material elicited. This can in one way be viewed as the theory having been chosen by the data (Dahlberg et al., 2008). The concepts discussed and their meanings were used to categorize the text in the initial phase of the analysis. However, in RLR, using a theory too early in the interpretive process is considered to disturb

the interpretation more than it helps (Dahlberg et al., 2008). There is a possibility that the theory input was too early in the process of interpretation, and that a different analysis procedure would have led to different results. The purpose of using a theory in RLR is to help see the data and its meanings better (Dahlberg et al., 2008). As such, I would argue that the theory we used helped to find the meanings embedded in the rich material. The theory was only used during this early phase of the analysis; thereafter, the analysis proceeded in line with the hermeneutical spiral towards a new whole (ibid).

The first phase in the analysis in Study III has already been discussed under the heading “Aspects of trustworthiness and credibility” above. The second phase of the analysis was a narrative analysis conducted by the researchers. To ensure credibility of the analysis, the data set was checked for contradictions, juxtaposing final themes and narratives, building on the work of Mattingly (1998) and Polkinghorne (1995). The themes were discussed in the research group to achieve richness and to check for representativeness of the raw data. By weaving together, the findings with the discussion, which was presented with relevant references and theoretical aspects, based on the discussions among the researchers, triangulation was used in the interpretation.

Conclusions and implications of the research

This thesis contributes to the body of knowledge of everyday life for adult persons with SB. The results emphasize that persons with SB have a complex set of medical, physical, cognitive and social needs that need to be addressed to sustain health and desired living conditions.

One important conclusion is that not all adult persons with SB receive the support they need in everyday life. Future generations of older adults with SB may need more attention, due to the complexity of outcome in various areas at a younger age which are expected to sustain through the lifespan and impact on living independently and participating in society. The results from the studies in this thesis is a call for systematic follow-up of services, as well as further exploration of how to accomplish individual and social support throughout life. To receive appropriate support in everyday life can enable possibilities for independence and participation in society, and thereby contribute positively to health. This is a field that needs further exploration.

The stories and experiences of adults with SB in this thesis shows a history of not being asked concerning their own situation. This includes insufficient integration of the persons experiences in society's efforts to plan for, and support these individuals. Altogether, different aspects contribute to low prerequisites for participation. Despite challenges in participation, by for example cognitive impairments with executive difficulties, the participatory method photovoice was feasible in this group. The photovoice project provided an opportunity for being part of, act and interact. This illustrate possibilities for collaboration and co-constructing knowledge, which can be used in future studies and when planning for support efforts in society. Photovoice is a feasible research method for this group, which also can be used as an intervention in future studies.

Clinical implications

There are a lot of processes in everyday life that need to be functional to actively support health. This needs to be acknowledged and considered in the clinical setting to enable appropriate support and sustainable actions, including giving the conditions for participation in decisions concerning care and treatment.

Photovoice can be used in a clinical setting, as an educational effort organized within a specialized rehabilitation clinic aiming to develop and actively train participation in decisions and co-working with others. This can also be organized to support self-care and may accordingly contribute to improved health and wellbeing.

Tack

Det finns många att tacka för att den här resan överhuvudtaget blev av, och för alla erfarenheter och lärdomar jag fått på vägen. Först av allt vill jag säga ett stort tack till alla som deltagit i studierna, som generöst gett av sin tid, kunskap och sitt engagemang samt delat sina berättelser. Utan er hade det inte blivit någon avhandling.

Mina handledare har varit avgörande i det här arbetet. Mats Eriksson, vi introducerades på en lunch (som du egentligen skulle ha med Malin Karlberg Traav och Agneta Cronqvist). Jag är tacksam och glad över att den där lunchen blev av och att vi därefter jobbat bra tillsammans. Tack för ditt lugn och din kunskap som hjälpt mig att navigera i den akademiska världen. Åke Seiger, du har varit en motor i kartläggnings-projektet (Benny) och aldrig sviktat i din övertygelse att det här kommer att genomföras. Tack för din kunskap, ditt engagemang och hängivenhet, i möten såväl i kommentarer på texter. Agneta Cronqvist, vi träffades först när jag gick min grundutbildning och sedan när jag skrev D-uppsats. Jag är glad att ha haft dig vid min sida under hela den här tiden. Jag uppskattar din rättframhet, humor och vetenskapliga stringens, som jag lärt mig så mycket av. Tack. Eric Asaba, att få möjlighet att genomföra photovoice projektet med dig som metodexpert har varit en fröjd. Tack för att du generöst och genuint delat med dig av din djupgående kunskap om framförallt deltagande och narrativ metod, både i praktik och teori, och även inom andra områden. Tillsammans har ni utgjort ett stabilt fundament i den, stundtals svajiga tillvaro, som doktorandtiden innebär.

Malin Karlberg Traav, förutom att det är helt din förtjänst att jag blev inskriven vid just Örebro Universitet, så har du också haft avgörande betydelse även i andra delar av mitt liv. Jag är tacksam över det och att vi nu, inom samma halvår, avslutar den här resan som vi startade tillsammans. Tack för uppehälle under kurser i Örebro och framför allt för ditt ovärderliga stöd och vänskap längs vägen. Vi har gjort många roliga saker, och jag ser fram mot att fortsätta med det.

Spinaliskliniken på Rehab Station Stockholm (RSS) (numer Aleris Rehab Station) är en unik arbetsplats på många sätt. Inte minst för den tydliga sammanvävningen av klinisk kompetens och forskning. Jag är glad över att ha fått ha en fot kvar i den här verksamheten (inte minst för att göra

övergången till ett ”nytt” jobb lite mindre smärtsam). En person som var viktig i starten av det här arbete är Ninni Westgren, som var chef och den som anställde mig som kordinator och sjuksköterska i mmc-teamet på Spinaliskliniken. Du värdesatte den här gruppen på kliniken och satte tydligt av de resurser som krävdes. Du var också en inspiratör för att påbörja en forskarutbildning. Lena Lindbo, numer operativ chef, kom som efterföljare och förvaldade arbetet som påbörjats. Tack båda för att ni delat er kunskap, stöttat och uppmuntrat längs vägen. Tack tidigare medarbetare, särskilt er som jag genom åren jobbat närmast med på sjuksköterske expeditionen: Gunnel Lif och Britt-Marie Holm, och ni i mmc-teamet, Dorothee Riedel Ratzka, Martina Bendt och Anna Persson. Doro, våra projekt har varit mest roliga och även lite slitsamma. Bland annat har vi släpat med en tung och otymplig utställning till olika platser i Sverige och genom snöstorm till Sunnaas sjukhus utanför Oslo. Du, med din förmåga att se möjligheter i de flesta situationer har varit helt avgörande i photovoice projektet. Martina, vi har spenderat så mycket tid tillsammans för att få kartläggningsprojektet i hamn, också roligt och lite slitsamt. Både arbete och en hel del privatliv har passerat revy under de här åren, och jag uppskattar verkligen att vi kunnat dela så mycket. Stort tack till er båda. Andra viktiga personer på Spinaliskliniken/RSS att tacka är tidigare doktorander som inspirerat, läst texter och stimulerat diskussioner under tidiga forskarfrukostar: Inka Löfvenmark, Emelie Butler Forslund, fougruppen och särskilt Tobias Holmlund och Lisa Holmlund. Tack också till Kerstin Wahman och Anna-Carin Lagerström för uppmuntran och support.

Stort tack till Stiftelsen Spinalis som bidragit med medel i de olika studierna, vilket förutom arbetstid, bland annat bidragit till guldkant och det lilla extra, som exemplevis när vi tillsammans med fyra av deltagarna i photovoice studien besökte Oslo i samband med utställningen. Claes Hultling, som VD för stiftelsen, medförfattare och allmän inspiratör, tack för att du alltid önskar bidra med just den där gulkanten på tillvaron. Tack också till Erika Nilsson som alltid stöttar och uppmuntrar.

Medförfattare som inte redan nämnts, Göran Hagman och Erika Franzén, tack för bra samarbete.

Ersta Sköndal Bräcke Högskola, är ännu en unik arbetsplats som jag har förmånen att arbeta på och som på olika sätt möjliggjort att den här avhandlingen kunde avslutas. Tack Jane Österlind och övrig ledning som

sett till att jag har fått tid för att skriva och möjlighet att jobba bredvid andra doktorander. Det har bidragit till att doktorandtiden blivit mer hanterbar och så mycket roligare. Tack till alla doktorander på Ersta, och särskilt tack till PIL-gruppen: Viktoria Wallin, Elin Hjorth och Lilian Pohlkamp. Ni har verkligen bidragit i både akademiska diskussioner och i andra viktiga domäner i livet, såsom brödbak, matlagning, stil och konst. Tack också till alla härliga arbetskamrater som hejat på och som jag nu ser fram mot att återse på mer regelbunden basis.

Tack till Örebro Universitet som fungerat som facilitator för mig under den här utbildningen. Trots att jag inte varit på plats så ofta, så har känslan av att allt fungerar som det ska infunnit sig, inte minst genom upparbetade välfungerande strukturer och system. Tack till fou-administrationen som gjort att allt flutit på som det ska, både i förhållande till doktorandarbetet och i samband med att utställningen visades på Örebro Universitet. Tack till doktorander och andra jag mött under kurser och seminarier som bidragit genom värdefulla kommentarer på mitt arbete.

Tack till släkt och vänner, vars middagar, bruncher, fika och sms uppmuntrat och bidragit till välbehövlig vila från jobb. Jag ser fram mot att umgås mer framöver. Bernt Lindgren, extra tack till dig för att du engagerade dig och hjälpte till med att få bilderna till utställningen i skick att tryckas. Till de som står mig närmast, min familj, tack mina fina barn VilmaRut och Vera, för att ni är just de ni är. Att få vara mamma till er, är och kommer alltid att vara, det största och finaste av allt. Till mina föräldrar, Göran och Birgitta, tack för att ni alltid stöttar, även i situationer då ni inte har helt klart för er vad det är ni stöttar, så finns ni där. Tack. Min bror Henrik med familj, tack för att du är en aktiv del av vår familj, även fast du bor långt bort så finns ditt stöd alltid nära. Micke, tack för din kärleksfulla närvaro genom det här och för välbehövligen avbrott i form av klättring, resor, middagar och promenader. Det har gjort skillnad.

Enskilda studier i den här avhandlingen har kunnat genomföras genom generösa ekonomiska bidrag. Tack till Norrbacka-Eugeniastiftelsen, Linnéa och Josef Carlssons Stiftelse samt Stiftelsen Spinalis.

Svensk sammanfattning

Bakgrund

Gruppen vuxna personer med ryggmärgsbråck ökar eftersom de som lever med denna medfödda skada förväntas leva allt längre. Ryggmärgsbråck räknas dock som ett sällsynt tillstånd, då allt färre föds till följd av fosterdiagnostik och efterföljande abort, samt ökad information om att ta folsyra inför planerad graviditet. Ryggmärgsbråck leder ofta till olika fysiska funktionsnedsättningar och omfattar i många fall även kognitiva nedsättningar av olika grad. Detta gör att det kan vara svårt att beskriva personer med ryggmärgsbråck som en grupp, eftersom utfallet av skadan varierar inom gruppen. Att ha kognitiva nedsättningar av exekutiv karaktär kan påverka möjligheten att vara självständig och att delta i olika aktiviteter. Det har framkommit utmaningar i självständighet och deltagande, och det har spekulerats kring om de förseningar i självständighet som framkommit, kan bero på inre aspekter eller låga förväntningar på självständighet från omgivningen. Eftersom antalet vuxna som lever med ryggmärgsbråck ökar, är det viktigt att utforska hälsa och levnadsvillkor, genom att involvera personerna själva i framtagandet av kunskap.

Syfte

Det övergripande syftet med den här avhandlingen, var att generera kunskap om och tillsammans med vuxna personer som lever med ryggmärgsbråck, genom att kartlägga tillståndet och utforska levnadsvillkor och upplevelser i vardagen.

Metod

För att besvara det övergripande syftet valdes olika metoder. Studie I hade en kvantitativ metod med tvärsnittsdesign. Etthundranittiosex vuxna personer med ryggmärgsbråck deltog. Strukturerade intervjuer, frågeformulär och kliniska bedömningar som inkluderade medicinska, sociala, fysiska och kognitiva aspekter, användes som metoder för datainsamling. Studie II var en kvalitativ studie som genom djupgående intervjuer undersökte upplevelser av vardagen på individnivå. Sex personer deltog. Intervjuerna genomfördes enligt principer för reflekterande livsvärldsforskning. Intervjuerna startade med uppmaningen *Kan du berätta om en bra dag?* Intervjuerna tolkades med hjälp av begrepp som rör

existentiell hälsa, såsom livsrytm, mening och sammanhang, och att finna sin plats i existensen. Studie III använde en deltagande metod som heter photovoice. Fem personer träffades tillsammans med facilitatorer, en gång i veckan under åtta veckor. Vid varje tillfälle bestämde gruppen ett tema som var av betydelse för vardagslivet, utifrån vilket medlemmarna i gruppen fotograferade följande vecka. Fotografierna fungerade sedan som utgångspunkt för dialogen när gruppen träffades igen. Gruppen var även delaktig i första delen av analysen där fotografier och de tillhörande berättelserna användes för att formulera teman. Av dessa bilder och berättelser skapade gruppen tillsammans med facilitatorerna en utställning som har visats på tio separata ställen. Forskarna genomförde en narrativ analys som innefattade hela processen. I Studie IV utforskas metodiken som photovoice vilar på. Genom att utgå från de ideologiska hörnstenarna inom photovoice och spegla dessa mot de empiriska erfarenheterna från Studie III och exempel från litteraturen, utforskas processerna i metoden.

Resultat

Resultaten visar att de deltagare som var över 46 år hade mindre komplexa medicinska tillstånd, bättre fysiska och kognitiva funktioner och hade uppnått en högre utbildningsnivå jämfört med de yngre. Huvudtemat i Studie II presenterades som ”Den motsägelsefulla vägen mot välbefinnande i det dagliga livet”. Det innefattar fem underteman som visade motstridiga känslor inför att umgås med andra som har ryggmärgsbräck, en avsaknad av att ha struktur i tillvaron, svårigheter att tolka signaler från kroppen och att agera, samt en önskan om att vara självständig. I Studie III visade medlemmarnas erfarenheter att många lösningar var ”En anpassning för oss, men den fungerar inte för någon”. En av berättelserna illustrerades av ett foto på en stängd dörr, som representerade otillgängligheten till en arbetsledare. Personen hade ett jobb att gå till, men utan att egentligen kunna delta, eftersom det inte fanns några vettiga arbetsuppgifter. Resultatet redovisades vidare i följande tre teman: ”Tillgänglighet – ett oändligt projekt”, ”Spänningar inom en normativ syn” och ”Makt att påverka”. I delstudie IV lyftes den viktiga funktionen av dialog, handling och interaktion som essentiella aspekter i metoden photovoice. Synergien av dessa aspekter skapar en arena för berättande och skapande av berättelser tillsammans med andra.

Konklusion

Den här avhandlingen bidrar till ökad kunskap om vardagen för vuxna personer med ryggmärgsbråck. Gruppen har komplexa behov som innefattar medicinska, fysiska, kognitiva och sociala aspekter som behöver tillgodoses för att förbättra hälsa och levnadsvillkor. En viktig slutsats är att inte alla vuxna med ryggmärgsbråck får det stöd de behöver i vardagen. Framtida generationer av äldre med ryggmärgsbråck kan komma att behöva mer stöd och uppmärksamhet på olika sätt, då de som är i yngre ålder redan idag har mer komplexa behov som förväntas följa med upp till de äldre åldrarna. Detta påverkar även förutsättningarna för att leva självständigt och delta i samhället.

Erfarenheterna och berättelserna i den här avhandlingen vittnar om att vuxna personer med ryggmärgsbråck har en historia av att inte bli tillfrågad om sin egen situation. Att ha uteslutits från delaktighet har lett till otillräcklig integration av personernas erfarenheter i samhällelig planering och i de insatser som samhället skapat i en strävan att stötta den här gruppen individer. Det finns visserligen utmaningar som påverkar förutsättningarna för att delta. Trots dessa utmaningar, som exempelvis kognitiva nedsättningar av exekutiv karaktär, var den deltagande metoden photovoice genomförbar med vuxna personer med ryggmärgsbråck. Projektet fungerade som ett tillfälle och möjlighet att vara en del av ett sammanhang, agera och interagera. Det är ett exempel på hur samarbete och samkonstruktion av kunskap kan gå till, vilket kan användas i framtida studier och när man planerar för stödinsatser i vård och samhälle. Photovoice skulle också kunna användas som en pedagogisk intervention inom klinisk verksamhet, för att aktivt träna deltagande beslutsfattande och samarbete, alternativt organisera ett photovoice projekt med fokus på att medvetandegöra aspekter av egenvård.

References

- Alriksson-Schmidt, A., Josenby, A. L., Lindquist, B., & Westbom, L. (2018). Pain and health status in adults with myelomeningocele living in Sweden. *The Journal of Pediatric Rehabilitation Medicine*, 11(4), 255-264. doi:10.3233/PRM-170517
- Alsaker, S., Bongaardt, R., & Josephsson, S. (2009). Studying Narrative-in-Action in Women With Chronic Rheumatic Conditions. *Qualitative Health Research*, 19(8), 1154-1161.
- American Academy of Orthopedic & Surgeons (1988). *Joint motion*. Edinburgh: Churchill Livingstone.
- Armour, B. S., Ouyang, L., Thibadeau, J., Grosse, S. D., Campbell, V. A., & Joseph, D. (2009). Hospitalization for urinary tract infections and the quality of preventive health care received by people with spina bifida. *Disability and Health Journal*, 2(3), 145-152. doi:https://doi.org/10.1016/j.dhjo.2009.02.001
- Aronowitz, R. (2008). Framing disease: An underappreciated mechanism for the social patterning of health. *Social Science & Medicine*, 67(1), 1-9. doi:https://doi.org/10.1016/j.socscimed.2008.02.017
- Arvidsson, P., & Granlund, M. (2018). The Relationship Between Intelligence Quotient and Aspects of Everyday Functioning and Participation for People Who Have Mild and Borderline Intellectual Disabilities. *Journal of Applied Research in Intellectual Disabilities*, 31, e68-e78.
- Asaba, E., Aldrich, R. M., Gabrielsson, H., Ekstam, L., & Farias, L. (2020). Challenging conceptualisations of work: Revisiting contemporary experiences of return to work and unemployment. *Journal of Occupational Science*, 1-14. doi:10.1080/14427591.2020.1820896
- Asaba, E., & Jackson, J. (2011). Social Ideologies Embedded in Everyday Life: A Narrative Analysis about Disability, Identities, and Occupation. *Journal of Occupational Science*, 18(2), 139-152. doi:10.1080/14427591.2011.579234

- Asaba, E., Laliberte Rudman, D., Mondaca, M., & Park, M. (2015). Visual methods: Photovoice in focus. In S. Nayar & M. Stanley (Eds.), *Research methodologies for occupational therapy and occupational science*. London: Routledge.
- Askheim, O-P., Bengtsson, H., & Richter Bjelke, B. (2014). Personal assistance in a Scandinavian context: similarities, differences and developmental traits. *Scandinavian Journal of Disability Research*, 16(S1), 3-18, <http://dx.doi.org/10.1080/15017419.2014.895413>
- Backhaus, B., Kaefer, M., Haberstroh, K., Hile, K., Nagatomi, J., Rink, R., . . . & Bizios, R. (2002). Alteration in the molecular determinants of bladder compliance at hydrostatic pressures less than 40 cm H₂O. *Journal of Urology*, 168, 2600-2604.
- Bakaniene, I., Ziukiene, L., Vasiliauskiene, V., & Prasauskiene, A. (2018). Participation of Children with Spina Bifida: A Scoping Review Using the International Classification of Functioning, Disability and Health for Children and Youth (ICF-CY) as a Reference Framework. *Medicina (Kaunas)*, 54(3). doi:10.3390/medicina54030040
- Bartonek, A., Saraste, H., & Knutson, L. M. (1999). Comparison of different systems to classify the neurological level of lesion in patients with myelomeningocele. *Developmental Medicine & Child Neurology*, 41(12), 796-805.
- Beauchamp, T. L., & Childress, J. F. (2013). *Principles of Biomedical Ethics* (7th ed.). New York: Oxford University Press.
- Bendt, M., & Bartonek, Å. (2016). Walking in adults with spina bifida with respect to muscle function. *European Journal of Physiotherapy*, 18(3), 154-160. doi:10.3109/21679169.2016.1150510
- Betz, C. L., Smith, K. A., Kysh, L., Roland, M., Van Speybroeck, A., Castillo, P., . . . & Mannino, J. E. (2019). Psychosocial outcomes for adults with spina bifida: a scoping review protocol. *JBI Database of Systematic Reviews and Implementation Reports*. doi:10.11124/JBISRIR-D-19-00072
- Bodin, C. R., Rasmussen, M. M., Tabor, A., Westbom, L., Tiblad, E., Ekelund, C. K., . . . & Petersen, O. B. (2018). Ultrasound in Prenatal Diagnostics and Its Impact on the Epidemiology of Spina

Bifida in a National Cohort from Denmark with a Comparison to Sweden. *BioMed Research International*, 2018, 9203985. doi:10.1155/2018/9203985

Bogart, K. R. (2014). The role of disability self-concept in adaptation to congenital or acquired disability. *Rehabilitation Psychology*, 59(1), 107-115. doi:10.1037/a0035800

Bogart, K. R., & Matsumoto, D. (2010). Living with Moebius syndrome: adjustment, social competence, and satisfaction with life. *The Cleft Palate-Craniofacial Journal*, 47(2), 134-142. doi:10.1597/08-257.1
10.1597/08-257_1

Bogart, K. R., Rosa, N. M., & Slepian, M. L. (2018). Born that way or became that way: Stigma toward congenital versus acquired disability. *Group Processes & Intergroup Relations*, 22(4), 594-612. doi:10.1177/1368430218757897

Borell, L., Asaba, E., Rosenberg, L., Schult, M.-L., & Townsend, E. (2006). Exploring experiences of “participation” among individuals living with chronic pain. *Scandinavian Journal of Occupational Therapy*, 13, 76-85.

Boudos, R. M., & Mukherjee, S. (2008). Barriers to community participation: Teens and young adults with spina bifida. *Journal of Pediatric Rehabilitation Medicine: An Interdisciplinary Approach*, 1.

Bowman, R. M., McLone, D. G., Grant, J. A., Tomita, T., & Ito, J. A. (2001). Spina Bifida Outcome: A 25-Year Prospective. *Pediatric Neurosurgery*, 34, 114-120. doi:10.1159/000056005

Brennan, C., Traustadóttir, R., Anderberg, P., & Rice, J. (2016). Are Cutbacks to Personal Assistance Violating Sweden’s Obligations under the UN Convention on the Rights of Persons with Disabilities? *Laws*, 5(2)(23). doi:10.3390/laws5020023

Bruegel, K. (2017). *The relationship between self-determination and well-being in adults with spina bifida*. (Doctoral dissertation, Palo Alto University, pacific graduate school of psychology).

- Buffart, L. M., van den Berg-Emons, R. J., Burdorf, A., Janssen, W. G., Stam, H. J., & Roebroek, M. E. (2008). Cardiovascular disease risk factors and the relationships with physical activity, aerobic fitness, and body fat in adolescents and young adults with myelomeningocele. *Archives of Physical Medicine and Rehabilitation*, 89(11), 2167-2173. doi:10.1016/j.apmr.2008.04.015
- Cardillo, L. W. (2010). Empowering Narratives: Making Sense of the Experience of Growing Up with Chronic Illness or Disability. *Western Journal of Communication*, 74(5), 525-546. doi:<https://doi.org/10.1080/10570314.2010.512280>
- Charlton, J.I. (1997) *Nothing about us without us: disability oppression and empowerment*. Berkeley: University of California Press.
- Clandinin, D. J. (2013). *Engaging in narrative inquiry*. Walnut Creek, CA: Left Coast Press.
- Connelly, F. M., & Clandinin, D. J. (2006). *Narrative Inquiry* (3rd ed.). Mahwah, NJ: Lawrence Erlbaum.
- Courbier, S., & Berjonneau, E. (2017). *Juggling care and daily life: the balancing act of the rare disease community A rare barometer survey*. France: Paris. Retrieved from: http://download2.eurordis.org.s3.amazonaws.com/rbv/2017_05_09_Social%20survey%20leaflet%20final.pdf
- Creswell, J. W., & Poth, C. N. (2017). *Qualitative inquiry and research design: Choosing among five approaches* (4th ed.). Thousand Oaks: Sage Publications.
- Cromby, J., Harper, D., & Reavey, P. (2013). *Psychology, Mental Health and Distress*. Basingstoke: Palgrave Macmillan.
- Daalen-Smith, C. V. (2014). 'My mom was my left arm': The lived experience of ableism for girls with Spina Bifida. *Contemporary Nurse*, 23(2), 262-273. doi:10.5172/conu.2006.23.2.262
- Dahlberg, K., Dahlberg, H., & Nyström, M. (2008). *Reflective Lifeworld Research* (2nd Edition). Lund: Studentlitteratur.

- Dahlberg, K., & Segesten, K. (2006). 'The Individual in the World - The World in the Individual': Towards a Human Science Phenomenology that Includes the Social World. *The indo-pacific journal of phenomenology*, 6(special edition), 1-9.
- Dahlberg, K., & Segesten, K. (2010). *Hälsa och vårdande i teori och praxis (Health and Caring in Theory and Practice)*. Stockholm: Natur & Kultur.
- De Chesney, M. (Ed.) (2015). *Nursing research using participatory action research*. New York: Springer Publishing Company, LLC.
- Degener, T. (2016). Disability in a Human Rights Context. *Laws*, 5(4), 35. doi:10.3390/laws5030035
- Dennis, M., Cirino, P. T., Simic, N., Juranek, J., Taylor, W. P., & Fletcher, J. M. (2016). White and grey matter relations to simple, choice, and cognitive reaction time in spina bifida. *Brain Imaging Behavior*, 10(1), 238-251. doi:10.1007/s11682-015-9388-2
- Dennis, M., Landry, S. H., Barnes, M., & Fletcher, J. M. (2006). A model of neurocognitive function in spina bifida over the life span. *The Journal of the International Neuropsychological Society*, 12(2), 285-296. doi:10.1017/S1355617706060371
- Dennis, M., Nelson, R., Jewell, D., & Fletcher, J. M. (2010). Prospective memory in adults with spina bifida. *Childs Nervous System*, 26(12), 1749-1755. doi:10.1007/s00381-010-1140-z
- Dicianno, B. E. (2014). 21st Century Challenges to the Provision of Health Care to Adults with Spina Bifida: A Rehabilitation Approach. *Archives of Physical Medicine and Rehabilitation*, 95, 1601-1602. doi:http://dx.doi.org/10.1016/j.apmr.2014.01.011
- Dicianno, B. E., Karmarkar, A., & Houtrow, A. (2015). *American Journal of Physical Medicine & Rehabilitation*, 94(null), 1015.
- Dicianno, B. E., Kurowski, B. G., Yang, J. M. J., Chancellor, M. B., Bejjani, G. K., Fairman, A. D., . . . & Sotirake, J. (2008). Rehabilitation and medical management of the adult with spina bifida. *American journal of physical medicine & rehabilitation / Association of Academic Physiatrists*, 87(12), 1027-1050.

- Dodds, T. A., Martin, D. P., Stolov, & Deyo. (1993). A validation of the functional independence measurement and its performance among rehabilitation inpatients. *Archives of Physical Medicine and Rehabilitation*, 74, 531-536.
- Edwards, T. C., Patrick, D. L., & Topolski, T. D. (2003). Quality of life of adolescents with perceived disabilities. *Journal of Pediatric Psychology*, 28(4), 233-241.
- Eslinger, P. J. (1996). *Conceptualizing, describing, and measuring components of executive function: A summary*. Baltimore, MD: Brookes.
- Fletcher, J. M., & Brei, T. J. (2010). Introduction: Spina bifida—A multidisciplinary perspective. *Developmental Disabilities Research Reviews*, 16(1), 1-5. doi:10.1002/ddrr.101
- Frank, A. (1995). *The wounded storyteller: Body, illness and ethics*. Chicago: University of Chicago Press.
- Freire, P. (1993). *Pedagogy of the Opressed*: Penguin Classics.
- Fridström Montoya, T. (2015). *Leva som andra genom ställföreträdare – en rättslig och faktisk paradox (Live like others through deputies – a legal and factual paradox)*. Uppsala: Iustus Förlag AB.
- Gabrielsson, H., Traav, M. K., & Cronqvist, A. (2015). Reflections on Health of Young Adults with Spina Bifida: The Contradictory Path towards Well-Being in Daily Life. *Open Journal of Nursing*, 05(04), 303-312. doi:10.4236/ojn.2015.54033
- Galvin, K., & Todres, L. (2013). *Caring and well-being: A lifeworld approach*: Oxfordshire: Routledge.
- Grue, J. (2011). Discourse analysis and disability: Some topics and issues. *Discourse & Society*, 22(5), 532-546.
- Hallberg, U. (2017). *Vård och omsorg om personer med funktionsnedsättning (Care and nursing for people with disabilities)* (Vol. 1st). Stockholm: Liber.
- Hammel J, Magasi S, Heinemann A, Whiteneck G, Bogner J, & Rodriguez, E, (2008). What does participation mean? An insider perspective

from people with disabilities. *Disability and Rehabilitation*, 30(19), 1445-1460. doi:10.1080/09638280701625534

Hampton, L. E., Fletcher, J. M., Cirino, P. T., Blaser, S., Kramer, L. A., Drake, J., & Dennis, M. (2011). Hydrocephalus status in spina bifida: an evaluation of variations in neuropsychological outcomes. *Journal of Neurosurgery-Pediatrics*, 8(3), 289-298. doi:10.3171/2011.6.Peds10584

Hemmingsson, H., & Jonsson, H. (2005). An Occupational Perspective on the Concept of Participation in the International Classification of Functioning, Disability and Health-Some Critical Remarks. *American Journal of Occupational Therapy*, 59, 569-576. doi:10.5014/ajot.59.5.569

Hilberink, S. R., van der Slot, W. M. A., & Klem, M. (2017). Health and participation problems in older adults with long-term disability. *Disability and Health Journal*, 10(2), 361-366. doi:10.1016/j.dhjo.2016.12.004

Hislop, H. J. (1995). *Daniel's and Worthingham's muscle testing. Techniques of manual examination*. Philadelphia, PA: Saunders Company.

Hoffer, M. M., Feiwell, E., Perry, R., Perry, J., & Bonnett, C. (1973). Functional ambulation in patients with myelomeningocele. *The Journal of Bone & Joint Surgery*, 55(1), 137-148.

Holland, D., Lachicotte, W., Skinner, D., & Cain, C. (1998). *Identity and agency in cultural worlds*. Cambridge, MA: Harvard University Press.

Holmbeck, G. N., & Devine, K. A. (2010). Psychosocial and family functioning in spina bifida. *Developmental Disability Research Reviews*, 16(1), 40-46. doi:10.1002/ddrr.90

Hughes, R. (2010). The social model of disability. *British Journal of Healthcare Assistants*, 04(10), 508-511.

Hultman, L. (2018). *Live life! Young peoples' experience of living with personal assistance and social workers' experiences of handling LSS assessments from a Child perspective*. (Doctoral dissertation, Karolinska Institutet, Stockholm).

- Hurst, R. (2003) The International Disability Rights Movement and the ICF. *Disability and Rehabilitation*, 25 (11-12): 572-6.
- Husserl, E. (1984). *The crisis of European sciences and transcendental phenomenology: An introduction to phenomenological philosophy* (6th ed.). Evanston, IL: Northwestern University Press.
- Iddon, J. L., Morgan, D. J., Loveday, C., Sahakian, B. J., & Pickard, J. D. (2004). Neuropsychological profile of young adults with spina bifida with or without hydrocephalus. *Journal of Neurology, Neurosurgery and Psychiatry*, 75(8), 1112-1118. doi:10.1136/jnnp.2003.029058
- Judkins-Cohn, T. M., Kielwasser-Withrow, K., Owen, M., & Ward, J. (2014). Ethical principles of informed consent: exploring nurses' dual role of care provider and researcher. *The Journal of Continuing Education in Nursing*, 45(1), 35-42. doi:10.3928/00220124-20131223-03
- Kinavey, C. (2007). Adolescents born with spina bifida: experiential worlds and biopsychosocial developmental challenges. *Issues in Comprehensive Pediatric Nursing*, 30(4), 147-164. doi:10.1080/01460860701728352
- Kirshblum, S. C., Burns, S. P., Biering-Sorensen, F., Donovan, W., Graves, D. E., Jha, A., . . . & Waring, W. (2011). International standards for neurological classification of spinal cord injury (revised 2011). *Journal of Spinal Cord Medicine*, 34(6), 535-546. doi:10.1179/204577211x13207446293695
- Lapides, J., Diokno, A., Silber, S.J, & Lowe, B.S., (1972). Clean, intermittent self-catheterization in the treatment of urinary tract disease. *The Journal of Urology*, 107, 458-461.
- Lennie, J. (2006). Increasing the rigour and trustworthiness of participatory evaluations: learning from the field. *Evaluation Journal of Australasia*, 1, 27-35.
- Levi, R., & Ertzgaard, P. (1998). Quality indicators in spinal cord injury care: a Swedish collaborative project. The Swedish Spinal Cord Injury Council 1998. *Scandinavian Journal of Rehabilitation Medicine. Supplement*, 38, 1-80.

- Lidal, I. B., Lundberg Larsen, K., & Hoff, M. (2019). 50 Years and older – born with spina bifida: participation, health issues and physical function. *Disability and Rehabilitation*, 1-10.
doi:10.1080/09638288.2019.1621953
- Lincoln, Y. S., Lynham, S. A., & Guba, E. G. (2011). Paradigmatic controversies, contradictions, and emerging confluences. In K. Denzin & Y. S. Lincoln (Eds.), *The SAGE handbook on of qualitative research* (4th ed., pp. 97-128). Thousand Oaks, CA: Sage.
- Lindberg, L. (2016). *Svensk forskning om funktionsbinder, forskningen som haltar (Swedish disability research the research that is limping)*. Stockholm: De hörselskadades riksförbund.
- Lindehall, B. (2007). *Teenagers and young adults with myelomeningocele and clean intermittent catheterisation - urological and psychosocial aspects*. (Doctoral dissertation, Göteborg University).
- Lindqvist, R. (2012). *Funktionshindrade i välfärdssamhället (Disabled people in the welfare society)*. Malmö: Gleerups Utbildning AB.
- Lindsay, S. (2014). A qualitative synthesis of adolescents' experiences of living with spina bifida. *Qualitative Health Research*, 24(9), 1298-1309. doi:10.1177/1049732314546558
- Liptak, G. S., & El Samra, A. (2010). Optimizing health care for children with spina bifida. *Developmental Disability Research Reviews*, 16(1), 66-75. doi:10.1002/ddrr.91
- Liptak, G. S., Robinson, L. M., Davidson, P. W., Dziorny, A., Lavalley, R., Flaherty, M. G., & Dosa, N. P. (2016). Life course health and healthcare utilization among adults with spina bifida. *Developmental Medicine & Child Neurology*, 58(7), 714-720. doi:10.1111/dmcn.12952
- Mathiowetz, V., Weber, K., Kashman, N., & Volland, G. (1985). Adult Norms for the Nine Hole Peg Test of Finger Dexterity. *The Occupational Therapy Journal of Research*, 5(1), 24-38. doi:10.1177/153944928500500102

- Mattingly, C. (1998). *Healing dramas and clinical plots: The narrative structure of experience*. Cambridge: University Press.
- McComb, J. G. (2015). A practical clinical classification of spinal neural tube defects. *Childs Nervous System*, 31(10), 1641-1657. doi:10.1007/s00381-015-2845-9
- Mitchell, L. E., Scott Adzick, N., Melchionne, J., Pasquariello, P. S., Sutton, L. N., & Whitehead, A. S. (2004). Spina Bifida. *Lancet*, 364.
- MMCUP. (2017). MMC Uppföljningsprogram för spinal dysrafism och hydrocephalus (*MMC Follow-up program for Spinal Dysraphism and hydrocephalus*) Årsrapport 2016 MMCUP Annual report 2016 . Retrieved from: <http://mmcup.se/wp-content/uploads/2017/09/170925-Årsrapport-MMCUP-2016-2017.pdf>
- MMCUP. (2020). MMC Uppföljningsprogram för spinal dysrafism och hydrocephalus (*MMC Follow-up program for Spinal Dysraphism and hydrocephalus*) Årsrapport 2019-juni 2020 Annual report 2019-june 2020. Retrieved from: <http://mmcup.se/wp-content/uploads/2020/09/200907-MMCUP-Arsrapport-2020-om-2019-till-SKR.pdf>
- Morley, C. P., Struwe, S., Pratte, M. A., Clayton, G. H., Wilson, P. E., Dicianno, B. E., . . . & Turk, M. A. (2020). Survey of U.S. adults with spina bifida. *Disability Health Journal*, 13(2), 100833. doi:10.1016/j.dhjo.2019.100833
- Nelson, K. (2000). Narrative, time and the emergence of the encultured self. *Culture and psychology*, 6(2), 183-196.
- Nilsen, T., Hermann, M., Eriksen, C. S., Dagfinrud, H., Mowinckel, P., & Kjekken, I. (2012). Grip force and pinch grip in an adult population: reference values and factors associated with grip force. *Scandinavian Journal of Occupational Therapy*, 19(3), 288-296. doi:10.3109/11038128.2011.553687
- Nisbett, R. E., Aronson, J., Blair, C., Dickens, W., Flynn, J., Halpern, D., F., & Turkheimer, E. (2012). Intelligence: new findings and theoretical developments. *American Psychologist*, 67, 130-159.

- Nordenfelt, L. (1995). *On the nature of health: an action-theoretic approach* (2nd rev ed.). Dordrecht: Kluwer.
- Nordenfelt, L. (2009). On health, ability and activity: Comments on some basic notions in the ICF. *Disability and Rehabilitation*, 28(23), 1461-1465. doi:10.1080/09638280600925886
- Oakeshott, P., & Hunt, G. M. (2003). Long-term outcome in open Spina Bifida. *British Journal of General Practice*, 53, 632-636.
- Oakeshott, P., Hunt, G. M., Poulton, A., & Reid, F. (2010). Expectation of life and unexpected death in open spina bifida: a 40-year complete, non-selective, longitudinal cohort study. *Developmental Medicine & Child Neurology*, 52(8), 749-753. doi:10.1111/j.1469-8749.2009.03543.x
- Ochs, E., & Capps, L. (2001). *Living narrative: Creating lives in everyday storytelling*. Cambridge, MA: Harvard University Press.
- Omerov, P., Kneck, A., Karlsson, L., Cronqvist, A., & Bullington, J. (2020). To Identify and Support Youths Who Struggle with Living-Nurses' Suicide Prevention in Psychiatric Outpatient Care. *Issues in Mental Health Nursing*, 41(7), 574-583. doi:10.1080/01612840.2019.1705946
- Partinen, M., & Gislason, T. (1995). Basic Nordic Sleep Questionnaire (BNSQ): a quantitated measure of subjective sleep complaints. *Journal of Sleep Research*, 4(1), 150-155.
- Patton, M. Q. (2015). *Qualitative Research & evaluation Methods: Integrating Theory and Practice* (4th edition ed.). California: SAGE Publications.
- Piaget, J. (1951). *Play, dreams and imitation in childhood*. New York: W.W. Norton.
- Plunkett, R., Leipert, B. D., & Ray, S. L. (2013). Unspoken phenomena: using the photovoice method to enrich phenomenological inquiry. *Nursing Inquiry*, 20(2), 156-164. doi:https://doi.org/10.1111/j.1440-1800.2012.00594.x

- Polit, D. F., & Beck, C. T. (2010). Generalization in quantitative and qualitative research: Myths and strategies. *International Journal of Nursing Studies*, 47(11), 1451-1458. doi:10.1016/j.ijnurstu.2010.06.004
- Polkinghorne, D. E. (1995). Narrative configuration in qualitative analysis. *International journal of qualitative studies in education*, 8(1), 5-23. doi:https://doi.org/10.1080/0951839950080103
- Quinn, G. (2009). Disability and Human Rights: A New Field in the United Nations. In C. Krause & M. Scheinin (Eds.) *International Protection of Human Rights: A textbook*. Turku: Åbo Akademi.
- Ricoeur, P. (1984). *Time and narrative* (K. McLaughlin & D. Pellauer, Trans. Vol. 1). Chicago, IL: The University of Chicago Press.
- Ricoeur, P. (1985). *Time and narrative* (K. McLaughlin & D. Pellauer, Trans. Vol. 2). Chicago, IL: The University of Chicago Press.
- Ridosh, M., Braun, P., Roux, G., Bellin, M., & Sawin, K. (2011). Transition in young adults with spina bifida: a qualitative study. *Child: Care, Health and Development*, 37(6), 866-874. doi:10.1111/j.1365-2214.2011.01329.x
- Rose, B. M., & Holmbeck, G. N. (2007). Attention and executive functions in adolescents with spina bifida. *Journal of Pediatric Psychology*, 32(8), 983-994. doi:10.1093/jpepsy/jsm042
- SFS 1993:387, Lag om stöd och service till vissa funktionshindrade (LSS) (Act concerning support and service for persons with certain functional impairments). Stockholm: Department of Justice.
- SFS 2001:453, Socialtjänstlag (SoL) (Social services act). Stockholm: Department of Justice.
- Socialstyrelsen (National board of health) (2011). *Personer med funktionsnedsättning – insatser enligt LSS år 2010 (People with disabilities - contributions according to LSS in 2010)*. Retrieved from: <http://www.socialstyrelsen.se/Lists/Artikelkatalog/Attachments/18319/2011-4-18.pdf>: Sveriges officiella statistik. Stockholm.

- Socialstyrelsen (National board of health) (2014) *Birth defects 2014*. Retrieved from:
<http://www.socialstyrelsen.se/publikationer2016/2016-3-4>
- Spina Bifida Association (2018). Guidelines for the care of people with spina bifida. (4th Ed). Retrieved from:
<https://www.spinabifidaassociation.org/wp-content/uploads/Guidelines-for-the-Care-of-People-with-Spina-Bifida-2018.pdf>
- Strömfors, L., Wilhelmsson, S., Falk, L., & Höst, G. E. (2016). Experiences among children and adolescents of living with spina bifida and their visions of the future. *Disability and Rehabilitation*, 39(3), 261-271. doi:10.3109/09638288.2016.1146355
- Talamonti, G., D'Aliberti, G., & Collice, M. (2007). Myelomeningocele: long-term neurosurgical treatment and follow-up in 202 patients. *Journal of Neurosurgery: Pediatrics*, 107(5), 368-386. doi:10.3171/ped-07/11/368
- Tallberg, I.M., Ivachova, E., Jones Tingheg, K., & Ostberg, P. (2008). Swedish norms for word fluency tests: FAS, animals and verbs. *Scandinavian Journal of Psychology*, 49, 479-485. doi: <https://org/101111/j.1467-9450.2008.00653.x>
- Tengland, P. A. (2020). Health and capabilities: a conceptual clarification. *Medicine, Health Care and Philosophy*, 23(1), 25-33. doi:10.1007/s11019-019-09902-w
- Tew, B., & Laurence, K. (1976). The effects of admission to hospital and surgery on children with spina bifida. *Developmental Medicine & Child Neurology Supplement*, 119-125.
- Thibadeau, J. K., Ward, E. A., Soe, M. M., Liu, T., Swanson, M., Sawin, K. J., . . . & Schechter, M. S. (2013). Testing the feasibility of a National Spina Bifida Patient Registry. *Birth Defects Research (Part A)*, 97(1), 36-41. doi:10.1002/bdra.23094
- Todres, L., Galvin, K., & Dahlberg, K. (2007). Lifeworld-led healthcare: revisiting a humanising philosophy that integrates emerging trends. *Medicine, Health Care and Philosophy*, 10, 53-63. doi:10.1007/s11019-006-9012-8

- Todres, L., Galvin, K. T., & Dahlberg. (2014). "Caring for insiders": phenomenologically informed insights that can guide practice. *International Journal of Qualitative Studies on Health and Well-being*, 9, 21421. doi:10.3402/qhw.v9.21421
- Törnbom, M., Jonsson, U., & Sunnerhagen, K. S. (2014). Work participation among middle-aged persons with cerebral palsy or spina bifida – a longitudinal study. *Disability and Health Journal*, 7(2), 251-255. doi:10.1016/j.dhjo.2013.06.005
- Törnbom, M., Lundälv, J., Jespersen, A., Sunnerhagen, K., Stibrant, K., & Grimby, G. (2011). Occupations and means of living in adults with cerebral palsy or meningomyelocele during two decades in Sweden. *Scandinavian Journal of Disability Research*, 13(1), 21-35. doi:10.1080/15017411003711791
- United Nations. (2008). CRPD Conventions on the Rights of Persons with Disabilities. *United Nations Treaties Series Vol 2515*.
- Utrikesdepartementet (2008). *Sveriges internationella överenskommelser, Konventionen om rättigheter för personer med funktionsnedsättning (Sweden's international agreements, the Convention on the Rights of Persons with Disabilities)* (2008:26). Utrikesdepartementet (Ministry of Foreign Affairs).
- Van Oudenhove, L., & Cuypers, S. (2014). The relevance of the philosophical "Mind-Body Problem" for the status of Psychosomatic Medicine: A Conceptual Analysis of the Biopsychosocial Model. *Medical Health Care and Philosophy*, 17, 201-213.
- Veenboer, P. W., Procee, A. I., Verheijden, J. M. A., Bosch, J. L., Ruud, H., van Asbeck, . . . & Laetitia, M.O. (2014). Medical and psychosocial problems in middle-aged spina bifida patients: Survey among members of the Dutch Patients' Association. *Disability and Rehabilitation: An International, Multidisciplinary Journal*, 36(7), 539-545. doi:10.3109/09638288.2013.801522
- Verhoef, M., Barf, H. A., Post, M. W., van Asbeck, F. W., Grooskens, R. H., & Prevo, A. J. (2004). Secondary Impairment in Young Adults with Spina Bifida. *Developmental Medicine Child Neurology*, 46, 420-427.

- Vu Minh Arnell, M., & Abrahamsson, K. (2019). Urinary continence appears to enhance social participation and intimate relations in adolescents with myelomeningocele. *Journal of Pediatric Urology*, 15(1), 33 e31-33 e36. doi:10.1016/j.jpuro.2018.08.008
- Wagner, R., Linroth, R., Gangl, C., Mitchell, N., Hall, M., Cady, R., & Christenson, M. (2015a). Perception of secondary conditions in adults with spina bifida and impact on daily life. *Disability and Health Journal*, 8(8), 492-498. doi:http://dx.doi.org/10.1016/j.dhjo.2015.03.012
- Wagner, R., Linroth, R., Gangl, C., Mitchell, N., Hall, M., Cady, R., & Christenson, M. (2015b). Perception of secondary conditions in adults with spina bifida and impact on daily life. *Disability and Health Journal*, 8(4), 492-498. doi:10.1016/j.dhjo.2015.03.012
- Wallerstein, N., & Duran, B. (2008). The theoretical, historical, and practice roots of CBPR. In M. Minkler & N. Wallerstein (Eds.), *Community-Based Participatory Research for Health, From Process to Outcomes* (2nd ed.). San Francisco, CA: Jossey-Bass.
- Wang, C., & Burris, M. A. (1994). Empowerment through photo novella: Portraits of participation. *Health Education Quarterly*, 21(2), 171-186.
- Wang, C., & Burris, M. A. (1997). Photovoice: Concept, methodology and use for participatory needs assessment. *Health Education & Behaviour*, 24, 369-387.
- Ward, K., & Trigler, J. S. (2001). Reflections on Participatory Action Research With People Who Have Developmental Disabilities. *Mental Retardation*, 39(1), 57-59. doi:10.1352/0047-6765(2001)039<0057:ROPARW>2.0.CO;2
- Webb, T. S. (2010). Optimizing health care for adults with spina bifida. *Developmental Disability Research Reviews*, 16(1), 76-81. doi:10.1002/ddrr.99
- Wechsler, D. (2002). *WAIS-III/WMSIII technical manual: Updated*. San Antonio: Psychological Corporation.

- Werhagen, L., Gabriellson, H., Westgren, N., & Borg, K. (2013). Medical Complication in Adults with Spina Bifida. *Clinical Neurology and Neurosurgery*, *115*, 1226-1229.
- Wetzel, J. S., Heaner, D. P., Gabel, B. C., Tubbs, R. S., & Chern, J. J. (2018). Clinical evaluation and surveillance imaging of children with myelomeningocele and shunted hydrocephalus: a follow-up study. *Journal of Neurosurgery Pediatric*, *23*(2), 153-158. doi:10.3171/2018.7.Peds1826
- Williams, E. N., & Morrow, S. L. (2009). Achieving trustworthiness in qualitative research: a pan-paradigmatic perspective. *Psychotherapy Research*, *19*(4-5), 576-582. doi:10.1080/10503300802702113
- Witt, W. P., Kasper, J. D., & Riley, A. W. (2003). Mental health services use among school-aged children with disabilities: The role of sociodemographics, functional limitations, family burdens, and care coordination. *Health Services Research*, *38*(6) (Part 1), 1441-1466.
- World Health Organisation. (2011). Summary world report on disability. Retrieved from: http://apps.who.int/iris/bitstream/10665/70670/1/WHO_NMH_VIP_11.01_eng.pdf
- World Medical Association (2018). Declaration of Helsinki: ethical principles for medical research involving human subjects. Retrieved from: <http://www.wma.net/policies-post/wma-declaration-of-helsinki-ethical-principles-for-medical-research-involving-human-subjects/>
- Young, N. L., Anselmo, L. A., Burke, T. A., McCormick, A., & Mukherjee, S. (2014). Youth and Young Adults With Spina Bifida: Their Utilization of Physician and Hospital Services. *Archives of Physical Medicine and Rehabilitation*, *95*(3), 466-471.
- Zabel, T. A., Jacobson, L. A., Zachik, C., Levey, E., Kinsman, S., & Mahone, E. M. (2011). Parent- and Self-Ratings of Executive Functions in Adolescents and Young Adults With Spina Bifida. *The Clinical Neuropsychologist*, *25*(6), 926-941. doi:10.1080/13854046.2011.586002

Zukerman, J. M., Devine, K. A., & Holmbeck, G. N. (2011). Adolescent predictors of emerging adulthood milestones in youth with spina bifida. *Journal of Pediatric Psychology*, 36(3), 265-276. doi:10.1093/jpepsy/jsq075

