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Pregnancy, Childbirth and Midwifery Care among Women with Intellectual Disability in Sweden

Epidemiological and Descriptive Studies

BERIT HÖGLUND



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Abstract

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The overall aim of this thesis was to investigate pregnancy and childbirth in women with intellectual disability (ID), in Sweden, the health of their newborns and midwifery care for these women. Two register studies and two descriptive studies are included. Pregnancy and birth outcomes as well as data on the newborns' health were examined by linking data from the National Patient Register and the Medical Birth Register (I-II). The women's experience of pregnancy and delivery was investigated with repeated interviews (III). Midwives' knowledge of, experience of and attitudes towards pregnancy and childbirth in women with ID were evaluated with questionnaires (IV). Mothers with ID were more often teenagers, smoked more during pregnancy and had more Caesarean Sections. Their children had a higher proportion of pre-term births, were small-for-gestational-age, stillborn or died in the perinatal period. The women with ID struggled to attain motherhood and feared to lose custody of the child. The pregnancy was seen as a happy event, even though relatives did not always approve. Parent education was considered important, but not adequately adapted to their needs. The birth process was overwhelming and difficult to understand, but the child was welcomed with warm feelings, and breastfeeding was natural. Midwives stated it was different to care for women with ID and requested additional knowledge. The majority of midwives affirmed that women with ID could not manage the mother role satisfactorily, and one-third expressed that women with ID should refrain from having children. A majority of the midwives considered that the children should grow up with the parents with support from family and society, but one out of five stated that the children should grow up in foster care.

Conclusion: Women with ID and their children should be considered as risk groups in pregnancy and childbirth. Professionals in maternity services need to elucidate their knowledge and skills for counselling and supporting this particular group of pregnant women in pre-, intra- and post-partum care.

Keywords: pregnancy, delivery, women, intellectual disability, newborn health, experience, knowledge, attitudes, education

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To women with ID who struggle for motherhood

List of Papers

This thesis is based on the following papers, which are referred to in the text by their Roman numerals.

- I Höglund, Berit; Lindgren, Peter; Larsson, Margareta. Pregnancy and birth outcomes of women with intellectual disability in Sweden: a national register study between 1999 and 2007. *Acta Obstet Gynecol Scand.* 2012 Aug 6. doi: 10.1111/j.1600-0412.2012.01509.x. [Epub ahead of print].
- II Höglund, Berit; Lindgren, Peter; Larsson, Margareta. Newborns of mothers with intellectual disability have a higher risk of perinatal death and being small-for-gestational age. *Acta Obstet Gynecol Scand.* 2012 Aug 24. doi: 10.1111/j.1600-0412.2012.01537.x. [Epub ahead of print].
- III Höglund, Berit & Larsson, Margareta. Struggling for motherhood with an intellectual disability - a qualitative study of women's experiences in Sweden. *Midwifery.* <http://dx.doi.org.ezproxy.its.uu.se/10.1016/j.midw.2012.06.014>.
- IV Höglund, Berit; Lindgren, Peter; Larsson, Margareta. (2012). Midwives' knowledge of, attitudes towards and experiences of caring for women with intellectual disability during pregnancy and childbirth – a cross sectional study in Sweden (*submitted*).

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Preface

A relative said to Anna (a woman with intellectual disability) that her weight seemed to increase and suggested she should buy a pregnancy test, which confirmed the pregnancy. Anna felt embarrassed and had ambiguous feelings about her condition. Previously, she had experienced a neonatal death and a miscarriage and now she was anxious and afraid that this could happen again. Anna did not tell her husband or anyone else during the first three months since she knew this time was the most critical period for miscarriage.

After four months Anna disclosed the pregnancy; her husband became happy, patting her stomach and laughing. The parents, especially her mother's reaction was very positive. Anna was registered at the antenatal care services, and during pregnancy she felt tired and was not able to ask or talk so much to the midwife. Anna received some brochures about pregnancy and delivery, but they were too difficult to read and understand, so she threw them away. Anna did not disclose her ID diagnosis to the midwife for fear of losing custody of her child. Once before, this situation had been a reality for her biological mother, when Anna was five-months-old.

The delivery started with painful contractions and Anna was registered at the delivery ward. A few hours earlier she had discovered something strange in the vulva that worried her, and she believed it was the baby's fingers. However, the midwife calmed her with an explanation of haemorrhoids. Her husband and a friend supported Anna during delivery, and when the pains became stronger Anna tried to call for the attention of professional support. However, no midwife appeared; so, the baby was born under the blanket without the help of a midwife. Anna had desired an epidural anaesthesia but the delivery happened too fast. After delivery, Anna felt happy and the baby screamed and rested on her chest. The father refrained from cutting the umbilical cord because he thought it would hurt the baby.

When Anna was discharged from the hospital, she and her baby were placed with her husband at a residential home for an assessment of their skills and ability to take care of their baby. Suddenly, when their baby was one-month-old the parents were told that they would lose the custody of the child. When

the day came to hand over the baby to the foster parents, the baby was sleeping in the pram and Anna did not want to loosen her grip on the pram.

After four years, Anna and her husband still do not understand how this could happen to them...their apprehension is that they did not do anything wrong with their baby...and they repeat *'we are still the parents of our child and we will wait for the possibility to take care of her until she will decide where to grow up.....'*

ABBREVIATIONS

BMI	Body Mass Index, weight in kilograms divided by square of height in metres
CHI ² TEST	The chi square test is used to determine whether there is a significant difference between the expected frequencies and the observed frequencies in one or more categories
CI	Confidence interval, the interval computed from a sample that has a given probability that the unknown parameter, such as the mean or proportion, is contained within the interval (here 95%)
ICD	International Classification of Diseases and Related Problems
ID	Intellectual disability
MBR	Medical Birth Register
NPR	National Patient Register
OR	Odds Ratio shows the strength of association between a predictor and the response of interest and varies from 0 to infinity. There is no association if the odds ratio is 1
PIN	Personal identification number
RR	Relative Risk is a statistical calculation of the ratio of disease in an exposed population to that of an unexposed population
SGA	Small for gestational age is a birth weight less than 2 standard deviations below the mean birth weight for gestational age and gender
UK	United Kingdom
WHO	World Health Organization

INTRODUCTION

My interest in this research field started in my professional work as a midwife at a delivery ward, in the beginning of the 90s. I encountered women with intellectual disability (ID) who delivered babies. I had reflections about my own knowledge, experience and probably insufficiency to care for women with ID in relation to pregnancy and childbirth. This topic had never been focused on during my education to become either a nurse or a midwife. Questions were raised such as: does she understand what it means and is she ready to become a mother? Has she received and/or understood important information about the forthcoming birth such as possible pain relief options during labour and delivery? Does she express her needs in the same way as other women do? Can I understand her needs and interpret her desires? Do I underestimate her needs? Does she trust me? In the literature, I found few articles focused on these questions. After several years with speculations, I decided to become a PhD student to explore this field in a scientific way.

Childbirth in women with ID

Studies describing pregnancy, delivery and newborn health in women with ID and their children are few and the knowledge is limited. Women with ID are a neglected group within the science. Few have asked about their opinions, not even within feministic research (1). In 1990 the Swedish National Board of Health and Welfare stated that individuals with ID, with consideration to themselves as well as to the children, should refrain from parenthood (2). Barron described the women's own opinions towards parenthood with words such as difficulties, negativism and impossibility. The interpretation was like a mirror of a message about parenthood and ID from an environment that disqualified their parental skills (1). Other studies claim that parenthood in women with ID should be avoided (3-5). A Swedish study affirmed the difficulties among young adults with ID, who are living in an intersection between independence and dependence. The parents of individuals with ID and professionals surrounding them decide the structure, to a large extent, and that influences even pregnancy and childbirth (6). An Australian study reported that the health of mothers with ID was significantly worse than other women's health in the Australian population

(7). A later study from Australia showed that more than one-third of the women interviewed reported moderate to severe depression, anxiety and stress during pregnancy (8). Another Australian study determined that children born to women with ID were more often born prematurely (28%) and had a low birth weight (22%) (9). An additional Australian study reported that a low birth weight was more than three times prevalent (OR = 3.08) and the children were more frequently admitted to neonatal intensive care (OR = 2.51) (10).

Children born to women with ID

Several independent sources describe parenthood among parents with intellectual disability, including both those with and without custody of the children (11-21). Women with ID enjoy freedom today, but past generations could hardly imagine forming relationships and starting a family of their own (22). Previous studies have affirmed that service providers do not regard people with ID as fit to be parents (12, 23-25); moreover, many children are also taken into foster-care. Different studies have reported that between 30% and 60% of children were not living with their parents with ID (12, 26-30). A consequence is that many of those parents-to-be live in fear of the service system that they often view as hostile and powerful; consequently, they try to develop strategies to ensure their children are not removed from their care.

Researchers affirmed that women with ID were excluded from decision-making, and rarely received any support to understand what was happening in the care and in the child-protection process (27, 31). Compared to other parents, they have more often terminated their parental responsibility and their children are placed in foster care (12, 13, 17, 29). No systematic relationship between parent characteristics and child removal was found (11-14, 16-20). However, North American studies show that parents who had children in foster care were more likely to report a lack of support from extended families and dissatisfaction with services (11, 20). Many parents with ID, but not all, live in difficult circumstances, including poverty, poor housing in low-income communities, more experiences with abuse and/or neglect in their childhood, violence in adult life and /or poor mental health (7, 20, 30, 32). Icelandic studies have affirmed that many parents with ID will lose custody of their children and these children will be permanently placed in foster care (33, 34). Researchers have affirmed different explanations for the loss of custody, as child protection workers and other professionals presume that parents with ID are incapable of raising their children regardless of whether there is evidence of neglect or maltreatment (12, 25). Other explanations are that parents are regarded as children

themselves; thus, cannot be entrusted with the care for another child (35). Difficult circumstances in life, for example, poverty, poor health, social isolation (36) and ID being a permanent condition leads to the assumption that any perceived parenting problems are also permanent (27, 37). Swedish studies state that parents with ID need support for several years and their children could need efforts until they are adults (38). Mothers with ID and a psycho-social overload may find it difficult to take care of their children (39). Starke acknowledged that parents with ID are a risk group and need regular and continuous support with education and skill training (40); furthermore, having good communication between professionals and parents working together as team is of utmost importance (41). McConnell et al. showed that child protection authorities and child welfare courts usually take into consideration several factors that are important for the child's future, such as: the age of the child or vulnerability, complexity and intensity of the parenting and childcare load, as well as the perceived possibility of improving the child's home situation (42). A recent study described that all mothers whose children had been compulsorily removed remained focused on their children who had been placed into foster care (43).

Intellectual disability

Terminology

Individuals with ID have been called different appellations during periods of time. Some of these have stayed for a longer time and some have come and gone quite fast. In the late 19th century, individuals were called the following in Swedish: 'idioter', 'imbecilla' and later as 'sinnesslö', 'svagsint', 'psykiskt efterbliven', 'psykiskt utvecklingsstörd', 'utvecklingshämmad', 'begåvningshandikappad', 'intellektuellt funktionshindrad' and 'utvecklingsstörd'. In 2007, the National Board of Health and Welfare substituted the appellations with "funktionsnedsättning" and "funktionshinder". The disability then refers to either the individual (funktionsnedsättning) or the individual in relation to the environment (funktionshinder) (44). In this thesis, the concept intellectual disability is translated to 'utvecklingsstörning'.

Diverse definitions of ID

ID is a condition, which touches a multidisciplinary arena and can be looked at from many different perspectives. The *functional definition* focuses on a person's functional limitation. The *relative* or *environmental* definition was introduced in the 1960s. It relates to the individual's environment and

situation; moreover, the Swedish law is built on this approach. Other points of view are the *administrative* definition and the *social* model. The first of these is related to the need for assistance, which the individual with ID requires. The social definition applies to the social system and not to a clinical-medical issue. The social model claims that disability is a property of the environment, not of the human being. Thus, our society is not designed to accommodate for persons with physical disabilities, e.g. access ramps, wide doors and elevators for wheel chairs, or microphones and hearing aids to compensate for hearing defects. A fifth definition describes a *subjective* definition of disability. This subjective definition refers to the individual with ID and his/her own self-image. The definition visualizes the individual's own choice to identify him/herself for understanding the world (45).

A variation in human conditions

Intellectual disability (ID) is sometimes called learning difficulties. ID is neither a medical condition, nor, strictly speaking, a medical diagnosis. Although not a psychiatric disorder, it is usually listed in diagnostic manuals of psychiatric disorders. ID is still often referred to as a developmental disorder, but this terminology is not appropriate. Although ID is not a pure medical problem, it has very strong links to medical conditions, and to psychiatric and developmental disorders (46).

History of ID

People with ID have in the past been exploited, estranged and not seen as human beings. They have been regarded as less important and have been neglected and haunted. Not until the 20th century did the respect for people with ID begin, together with a confession of their rights. The opinion of eugenics and a justification of efforts not to pass on a heritage of ID dominated from 1910s and thirty years forward. During that time, the philosophy of ID was mostly that it was a genetic condition and not treatable. The consequences were sterilisation, and locked institutions for individuals with ID (44). For the greater part of the 20th century, women with ID were institutionalised and/or sterilised, to prevent pregnancy. In Sweden, involuntary sterilisations occurred from 1935 to 1975 (47). Currently there is no legislation prohibiting childbearing among these women in Sweden, but negative attitudes towards pregnancies and parenthood persist (48). The health care ideology changed in the 30s from protecting individuals with ID from society to the opposite, namely, protecting society from individuals with ID. During the 1930s and 1940s, the society opened special mental hospitals in different parts of Sweden and individuals with ID were institutionalised. The influence of the medical viewpoint increased and the

right to self-determination was minimal for these people. After the Second World War several laws and conventions, including human rights for people with ID have been developed to reduce differences in living conditions in relation to the general population (44).

Modern age

In 1968, the Law of Care (Omsorgslag) was passed and an elimination of segregated institutions started. The new life for individuals with ID consisted of receiving care at homes, access to activities at a day centre, living as a paying guest in integrated homes and a right to education. The state assigned the responsibility for care to the County councils (44). Since 1994 the Law on Support and Service for people with Certain Functional Impairments (LSS) has been in effect. The principle aims of the LSS (ten rights/interventions) are to guarantee full participation in society and freedom to choose one's own path in life. In practice, this means that young adults can move away from their parents to houses or apartments where the staff assists the residents to get their special needs taken care of or they move into their own living and housing conditions. They have a right to apply for help and support related to their own special needs, e.g. daily care, support and other services. A person with ID has legal majority but a trustee could be nominated when a need is identified. The latter is of assistance e.g. with the economy and the civil rights (49).

Research on care and services

Barron describes two positions when providing care and service to persons with ID: support versus control. Support includes more or less everyday life with professionals. Control proceeds from a view of the staff having a superior position of control, classifying individuals with ID as passive recipients with restricted autonomy and influence. It could be a complex situation with contradictory meanings for individuals with ID; they have to be dependent to become independent (1). A study describes difficulties for young adults with ID, and their approaches to sexuality and sexual activity with subsequent parenthood. The professionals, their own parents and others advocate for and express assumptions and signals that the young adults with ID would have less ability and skills in a prospective parent function (6). An Australian study describes that understanding the experiences of pregnant women with ID from their point of view is an essential key (48). When doctors, midwives and other health professionals are giving support to women with ID, it is important to avoid expressing pejorative beliefs about decisions pregnant women are making for and about their babies. Women with ID are painfully aware of such attitudes. Support workers should be

sensitive to, and respectful of, decisions made in the context of a woman's intimate relationship.

The structure of ICD

During the 15th and 16th century, the first systems of classification for medical diseases were presented. A special interest in Sweden is the "Genera morborum" (the disease groups) by Linné, who in 1763 set up the diseases in classes and categories (50). The International Classification of Diseases (ICD) is the international standard diagnostic classification for all general epidemiological, many health management purposes and clinical use published by the World Health Organization (WHO) in 1948. These include the analysis of the general health situation of population groups and monitoring of the incidence and prevalence of diseases and other health problems in relation to other variables such as the characteristics and circumstances of the individuals affected, reimbursement, resource allocation, quality and guidelines. The current, tenth revision (ICD10) was introduced in 1997 (51).

The diagnosis Mental Retardation in ICD classifications

The disease classification is built on 21 chapters of different diseases and categorical groups. The mental and behavioural disorders have been included in chapter V since 1952 (ICD 6). The diagnosis mental retardation has been classified in different combinations of letters and figures over time. The diagnosis ID is classified in chapter V and to F70-F79 in ICD 10. Degrees of mental retardation are conventionally estimated by standardised intelligence tests. These can be supplemented by scales assessing social adaptation in a given environment. These measures provide an approximate indication of the degree of mental retardation. Intellectual abilities and social adaptation may change over time, and, however poor, may improve as a result of training and rehabilitation. Diagnosis should be based on the current levels of functioning (51).

Other major classifications of ID

The American Psychiatric Association (APA) is a national medical society. The psychiatric members have developed an international classification system called Diagnostic and Statistical Manual of Mental Disorders (DSM). The first version was published in 1952 and all versions are related to roman numerals. DSM-IV is the current edition and was released in 1994. The manual is mainly based on psychiatric conditions and not disorders (52).

Both ICD and DSM have origins from a traditional epidemiological viewpoint; in Sweden they complement each other (53). The AAMR (American Association of Mental Retardation) and the ICF (International Classification of Functioning) have been regarded as related in theoretical models. Both are focused on categories of support needs rather than levels of mild, moderate and severe ID. The ICF conceives functioning as a person related to the environment. The ICF is used as a complement to ICD 10 in health conditions, diseases and disorders. In Sweden, Kylén has described and classified ID using a combination of biological age, intelligence and social ability. This theory is based on cognitive and psychodynamic theories in an integrated A-B-C model according to the WHO classifications (54).

Definition of ID in International Classification of Diseases (ICD)

Internationally, an intellectual disability (ID) is defined as an individual with an intelligence quotient (IQ) below 70, a derogation of adaptive capacity, and a debut before 18 years of age (55). This definition is medical and ID is divided into mild (IQ 50-69), moderate (IQ 35-49), severe (IQ 20-34), profound (IQ <20), other and unspecific. In clinical practice, mental retardation is defined as 'a condition of arrested or incomplete development of the mind, which is especially characterised by impairment of skills manifested during the developmental period, contributing to the overall level of intelligence, e.g. cognitive, language, motor and social abilities'. Generally, a person with ID needs a longer time and more help to learn things (51). People with ID are heterogeneous persons just like others; however, ID is a permanent condition and will stay for life. Approximately one per cent to two per cent of the population has an ID and that differs with the definition used. About half of them have a moderate to severe ID. Sometimes the cause of ID is known but in many cases no explanation can be found. When the cause is known, the child may have suffered some damage during pregnancy or birth, or when he or she was very small (56). The most common is trisomy 21, also called Morbus Down's syndrome, which was described by John Langdon Down in 1866 (57). Mb Down, as well as other more severe manifestations of ID, tends to be recognised earlier in life especially when associated with a syndrome. Mild levels of ID of unknown origin are noticed later in life, most commonly in the early school years (54).

The registers at the Swedish National Board of Health and Welfare

The Swedish National Board of Health and Welfare keeps different registers and two of them are the Swedish Medical Register (MBR) and the National Patient Register (NPR)(58). The Swedish Medical Birth Register was established in 1973 through an act by the Swedish Parliament. Medical information containing data from all hospital births and the few home births in Sweden are kept in the MBR. The purpose of the register is to compile information related to antenatal and perinatal factors that may also be of importance for the health of the infant. The register covers 98-99% of all births in Sweden. One of the most important components in the use of the Swedish health registers is the personal identification number (PIN); a unique ten-digit number assigned to each resident in Sweden, which is used in a wide variety of contexts. The PIN allows for the information between different registers and data between defined cohorts and the registers to be linked (59-61).

The National Patient Register (NPR)

The patient register is one of the world's few individual-based registers, which covers the whole population's consumption of health care in institutional and non-institutional care. The information in NPR can be divided into four different groups: patient data, geographical data, administrative data and medical data from 21 county councils in Sweden. From 1987, NPR also includes all in-patient care in Sweden and contains statistics of diseases and surgical treatment of patients in Sweden (60). Starting in 2001, the register also contains outpatient care from both private and public caregivers. However, primary health care is not yet included in the NPR. A quality control of the NPR is regularly performed. On average, 1% of the main diagnosis (1964-2007) was missing. In the same time period, the psychiatric care with a main diagnosis had the highest dropout rate (3.1%) of all the specialties. The dropouts were generally concentrated in a few county councils. In 1986 and 1990, two validity tests were performed. The findings were: incorrect transmission deficits, wrong codes and wrong diagnosis in descending scale (61). Other previous theses that are built on NPR are studies on e.g. Ljungan virus infection and cot death (62), cerebral palsy in children born after *in vitro* fertilisation (63) and long-term risks of osteoporosis and fractures in Malmö (64).

The Swedish Medical Birth Register (MBR)

All health care providers report to the register from medical records about the prenatal, delivery and neonatal care. The register's quality was evaluated

in 1976, 1988 and 2001. The content of the register was modified in 1990, 1994 and 1998. By linkage to registers provided by Statistics Sweden information on personal identification number for the infant, the parents' nationality and the mother's country of birth is also added to the register. There is reason to assume that the rate of neonatal deaths is slightly underestimated in the MBR. It could be explained by uncorrelated medical records and neonatal care during a long time before a fatal outcome (59, 65, 66). The MBR has previously been used in several other research fields, for example, birth centre care and infant morbidity (67), use of epidural analgesia and its relation to caesarean section and instrumental deliveries (68), preterm and small-for-gestation age (SGA) and the risk of subsequent stillbirth (69), and factors associated with increased frequency of caesarean section (CS) (70). Studies based on standardised collection of data gathered at antenatal care units or hospitals are thus mainly free from recall bias. Further, since maternal care is free of charge and attended by most pregnant women and home deliveries are rare in Sweden, selection bias is unlikely.

Care in pregnancy, delivery and the postnatal period

The model of antenatal health care originated in England. In Sweden, this model of care was developed in the 30s. The overall goal of maternal health care is to provide care and support to enhance medical, psychological and social well-being during pregnancy and childbirth. The aim is to secure the health of the woman and her child during pregnancy and delivery and make the entire process a positive experience for the expectant and new parents. The antenatal care consists of a basic programme with approximately 8-10 appointments with a midwife during pregnancy. Special routines are established for women with special needs e.g. chronic diseases, previous obstetric complications, and psychosocial risks. The basic programme identifies risk factors during pregnancy and delivery for the woman and her child. If the midwife estimates that the pregnancy is abnormal or the pregnant woman herself has a need or a wish to consult other professionals during pregnancy, these consultations will be arranged. The professionals include obstetrician, specialist midwife (often named Aurora), physiotherapist, occupational therapist, psychologist, dietician, welfare officer and welfare worker.

Upon registration at the antenatal care, all women are informed about and offered a screening ultrasound and prenatal diagnosis. The psychosocial interventions consist of parental education for the pregnant woman and her partner during the last trimester and also psychosocial support and care during pregnancy and delivery. Ninety-nine per cent of all deliveries occur at hospitals. A normal delivery is the midwife's responsibility. If needed, the

midwife calls for the obstetrician. During the last two decades the postpartum stay in hospital has become shorter. From 1993 to 2005, an early discharge meant to be discharged within six hours to 72 hours after birth. Subsequently, early discharge means soon after delivery with a minimum of six hours hospital stay. During that era, midwife-teams began to work with home visits. Their commitment was to care for and support mothers and children at home to enhance health. During the last two to three decades it has become increasingly common for the father to stay with the mother and the child at the postnatal ward. The newborns are examined twice by a paediatrician, before discharge and within five days (71). The government finances the health care.

Experience of pregnancy and childbirth

The word 'experience' is explained as 'having worked in many types of situations and knowing how to cope with different problems', 'things which have happened to someone' and 'to live through a situation' (72). Women's experiences during pregnancy, delivery and postpartum have been explored in Sweden (73-76). Risk factors for negative experiences were related to social life, socio-demographic background and lower education level (75, 77). Experience of pregnancy and childbirth in women with ID has affirmed a social resistance with disbelief and dismay in the encounters with surrounding people (30, 48, 78, 79). In women with ID, their coping strategy was to rely on a trusted environment but refrain from contacts with an unreliable surrounding (48). A thesis describing an overall assessment of women's experiences of intra-partum and post-partum care, two months after the birth, indicated that 10% and 26%, respectively, were dissatisfied (80). Another thesis confirmed a positive overall experience of post-partum care (70%), but an equivalent amount expressed discontent with certain parts of the care and support, such as treatment of the new father (81). Other words that are used in this thesis are 'knowledge' and 'attitude', which could be explained as 'all the facts that someone knows about a particular subject' and 'someone's opinions or feelings about something, especially as shown by their behaviour', respectively (82).

The care of the midwife

The midwifery education in Sweden is at the university level and includes both the nurse's exam (3 years) and the midwifery exam (1.5 years). The midwifery program consists of theory in obstetrics, gynaecology, reproductive health and a master's degree in reproductive health. The clinical education includes taking care of, at the minimum, one hundred women and supporting fifty women during their deliveries. Additional parts are antenatal care, postnatal care, gynaecology care, reproductive care and

care at maternal health services. A description of an ideal midwifery care in childbirth can be summarised as a trusting relationship, balancing between a natural and a medical perspective and having bodily knowledge. In a worthy relationship, mutuality, trust, on-going dialogue, shared responsibility and continuous presence are included. Bodily knowledge includes authenticity and skills from a theoretical, practical, intuitive and reflective nature (83). A Swedish observational study disclosed three basic patterns in consultations between the midwives and the pregnant women during antenatal care. The patterns included ‘the respectful gardener and her developing plants’, ‘the propagandist teacher and her ignorant pupils’, ‘the steering inspector and representatives of the population at her disposal’(84).

Care in pregnancy and childbirth for women with ID

Although the sterilisation law in Sweden terminated in 1975, there has never been an evaluation of the needs and desires of women with ID in relation to childbirth. In the daily Swedish maternal health care, women with ID may be excluded from the basic program and become classified as “pregnant women who should be offered an early contact with the physician or be handled according to a local care program” (71). Pregnancy, delivery and newborn health in women with ID and their children is still an unexplored arena, both from an epidemiological and medical point of view as well as from the women’s own experience.

Theoretical framework

Many theoretical models are developed to describe, explain and predict human behaviour and understand the phenomena of concern (85). Several are used in midwifery theory development. Theories and models are abstract pictures of thinking about one or more individuals relating to some aspect of the world. Theories are developed from understandable related concepts and tested through observation or other forms of research but must be regarded as mental constructs (86). Some have expressed concerns about believing in theories, since they are only tools for organising data, for making sense of and explaining reality, so confusions could be predicted (87). Two of the existing midwifery theories have guided and served as an inspiration for this thesis. Reva Rubin was a nurse-midwife and her model was developed in the 1960s in the US. Her research questions were stimulated by role theory and explored the development of the maternal role. This model describes the attainment of the mother role, and is developed in relation to the key concepts of person, health, environment and midwifery. The process of attaining the mother role strives to attain four goals and spans throughout the whole pregnancy to the postnatal period. It includes taking-on activities,

taking-in activities, and letting-go activities (88-90) see Figure 1. Five means related to those activities were described by Rubin, in how the maternal identity is incorporated into the image of herself as a mother.

- Taking-on activities: mimicry and role play
- Taking-in activities: fantasy and introjection-projection-rejection
- Letting-go activities: grief-work

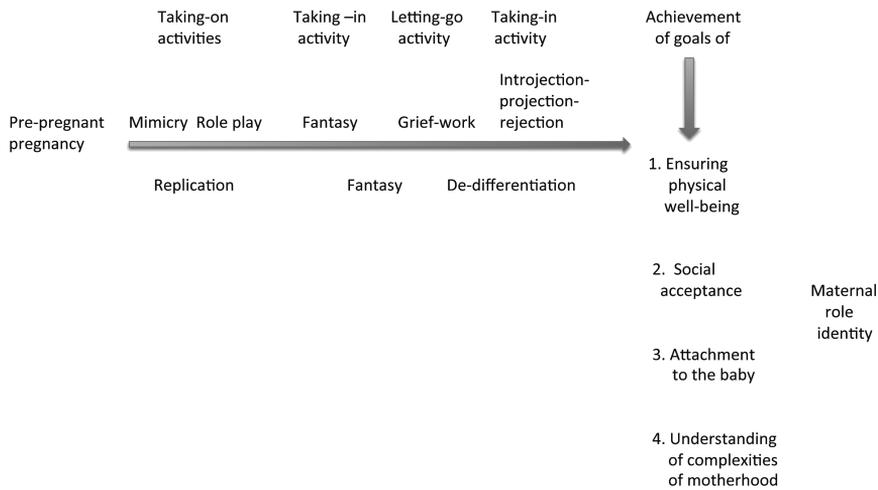


Figure 1. Linear representation of Rubin's model of attainment of the maternal role

Jean Ball, a British midwife, developed another useful theory in the 1980s. She advocates the importance of the social context of the woman's life in her adaptation to motherhood. Jean Ball's deck-chair theory of maternal, emotional well-being identifies the pregnancy and the postnatal period as a time for adaption of a new role. The purpose of all maternity care is to enable a woman to successfully become a mother; in addition, the process involves physiological, psychological and emotional components. The maternal well-being depends partly on the woman's personality, previous experiences and life-crises. These factors relate to the care from midwives and other professional groups at maternity services, attitudes and resources in society and also support from families and peers (86).

AIMS

The overall aim of this thesis was to investigate pregnancy and delivery in women with ID and the health of their newborn children. Another aim was to describe midwives' work with and attitudes towards women with ID in relation to pregnancy and childbirth.

The specific aims were:

1. To investigate the antenatal health and demographic factors as well as pregnancy and delivery outcomes in women with ID.
2. To investigate the mode of birth, preterm birth rates, Apgar scores, small-for-gestation-age, stillbirth and overall perinatal health in children born to mothers with ID.
3. To gain a deeper understanding of the experiences of pregnancy and childbirth in women with ID.
4. To describe the midwives' knowledge and experiences of and attitudes towards women with ID during pregnancy and childbirth.

METHODS

An overview of the studies is presented in Table 1

Design	Data collection method	Participants	Data analysis
I. Comparative cohort study	Register (NPR, MBR)	326 women with ID 340,624 women without ID or any psychiatric diagnosis	Chi ² , Fischer's exact test, t-test, relative risk, logistic regression analysis
II. Comparative cohort study	Register (NPR, MBR)	326 newborns to mothers with ID 340,624 newborns to mothers without ID or any other psychiatric diagnosis	Chi ² , Fischer's exact test, t-test, relative risk, logistic regression analysis
III. Descriptive qualitative study	Individual interviews	10 women with ID	Content analysis
IV. Cross-sectional survey	Postal questionnaire	180 midwives at antenatal health services 195 midwives at delivery wards	Chi ² , Fischer's exact test, Mann Whitney U-test, logistic regression analysis

NPR - National Patient Register; MBR - Medical Birth Register

Population and sample

Paper I and II

The study was carried out on a Swedish population-based cohort of women with ID diagnosis Retardatio mentalis in ICD 8 to ICD 10 (defined here as non-syndromic ID) and a cohort of woman without ID, or any psychiatric diagnoses. The women with ID were identified from the NPR, linked to the MBR at the National Board of Health and Welfare, from which the control group was also selected. The samples were based on diagnoses from hospital and non-institutional care and medical records from pregnancy and delivery and included all births between 1973 and 2007 for the whole of Sweden. A more stringent cohort was chosen for analysis, with only singletons and primiparas between 1999-2007, which resulted in 326 women with ID and their children (n= 326) and 340,624 women without ID or any psychiatric diagnosis and their children (n= 340,624), see Figure 2.

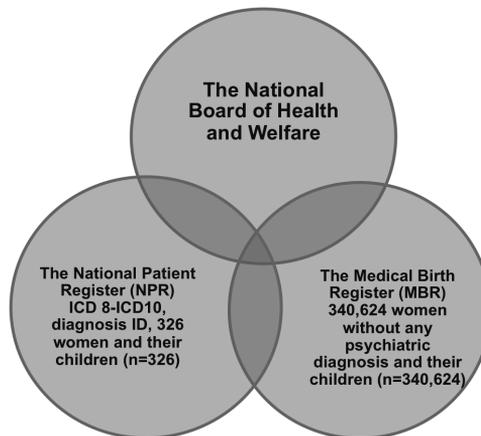


Figure 2. The women with ID were identified at the NPR, linked to the MBR at The National Board of Health and Welfare. The women without ID or any psychiatric diagnosis were identified at the MBR in studies I and II

Paper III

A modified snowball sampling was used. The researcher contacted key persons such as school nurses at compulsory schools, psychologists and social welfare officers at rehabilitation centres, where women with ID could be identified in the whole of Sweden. These key persons informed eligible women with ID, who had given birth between 2004 and 2010, about the

study. The women who were recognised all had the diagnosis of ID that entitled them to formal support services. The selection of the time-period for their childbirth was made in order for them to maintain an optimal memory of their experiences of pregnancy and delivery. If the woman agreed to be contacted by the researcher, she was given both oral and written information. Thereafter, if she wanted to participate in the study she was included. To avoid possible feelings of being exposed, they were advised to inform and discuss the study with their trustees or trusted person with whom they had a close relationship. Furthermore, if she wished, the researcher offered to inform their trustees or/and any trusted person with whom they had a close relationship about the study. Ten women with ID who had given birth from 2004 to 2010 were included. A semi-structured topic guide covering experiences of pregnancy and childbirth was constructed. At first, two midwives, a physician, a psychologist and two social welfare officers developed the guide. Subsequently, a pilot study was performed with two women; thereafter, some adjustments were made. In every interview the initial question was, 'Please, would you like to tell me about your experiences when you were pregnant and gave birth to your child'. The interviews were performed in a relaxed atmosphere adapted to the women's ability to communicate. The interviewer simplified the language if it was needed. Each interview was tape-recorded after obtaining permission from the woman. The interviews took place from May 2010 to January 2011.

Paper IV

A questionnaire and an invitation letter were sent out in August 2011 to a random sample of 600 midwives in Sweden: 300 midwives working at antenatal care services and 300 midwives working at delivery wards. The addresses were obtained from a public register of health professionals, The Health and Care Address Register. The invitation letter explained the purpose of the study. Voluntary participation and confidentiality related to information on personal data was assured. Non-respondents got reminders via SMS (Short Message Service) or postcards after five weeks and then reminded with a new questionnaire after eight weeks. A questionnaire in two similar versions was developed for the study, one for midwives at antenatal care and the other for midwives at delivery care. The questions were based on findings from the previous interview study with women with ID and from personal experiences of the authors, a psychologist and social workers. Furthermore, an obstetrician and two midwives working at antenatal care and at a delivery ward, confirmed face validity of the questionnaire. A test-retest was conducted among 40 midwives who fulfilled the inclusion criteria. Internal reliability of the Likert scale items was tested using Spearman's correlation test. All items with a correlation coefficient ≥ 0.70 were retained; other items were reconstructed or deleted. The questionnaire consisted of

65/60 questions altogether: socio-demographics (6 items), organisation of care (4 items in antenatal care and 2 items at the delivery ward), knowledge (12 items), attitudes (10 items), experiences (17 items in antenatal care and 14 items at the delivery ward), and education and education needs (16 items). All questions were multiple-choice or a 4-point Likert scale ranging from totally agree to totally disagree.

Data analysis and statistical methods

Statistical analyses were performed with the SPSS 15.0 and 20.0 software program for Windows (IBM Statistical Package for the Social Sciences). Differences between the two groups were tested with Fischer's exact test and Pearson's Chi-square test for two independent samples on nominal-scaled variables (papers I, II, IV), and with the Mann Whitney for the ordinal-scaled variables (paper IV). Differences were considered significant if $p < 0.05$ (91). Interval scaled variables were analysed by comparing means with the independent – samples T-test (papers I, II). Relative risks (RR) were calculated to determine the differences as a ratio between the percentages and a 95% confidence interval by Mantel-Haenszel's method (papers I, II). To examine the main predictors for the dependent variables: *preterm birth*, *CS*, *non-use of nitrous oxide* and *discharge to a place other than home* (paper I) and *preterm birth*, *Apgar <7 points*, *small-for-gestational age (SGA)*, *perinatal death* and *stillbirth* (paper II), multiple logistics regression models were fitted for each of the dependent variables, respectively.

In order to control for potential confounders in Paper IV, a logistic regression model was fitted for each of the four dependent outcome variables: *women with ID should not become pregnant at all*, *women with ID should have the children they want*, *women with ID can manage their maternal role with satisfaction* and *the children of mothers with ID should be/grow up in foster care*.

Content analysis was used to analyse the interview data (92, 93). This analysis was a stepwise process of categorisation based on the expressions of feelings, thoughts and actions described throughout the text. After careful reading of all the interviews, the text was divided into meaning units; a piece of text that related to the aim of the study. All meaning units were then condensed while preserving the core content into codes; a labelling that allowed the data to be understood in relation to the context. The codes were then grouped into sub-categories and thereafter into categories, depending on similarities and differences in content. Finally, there was a search across categories to identify recurring regularities expressed into a theme (Table 2). The first author performed the analysis throughout the whole analytical scheme, and the last author participated in all steps after the coding process (paper III).

The answers to the questions regarding knowledge were added to form an index of knowledge, which could add up to a maximum of 12 points if all answers were entirely correct (paper IV).

Table 2. Example of the analytical scheme

Theme	Struggling for motherhood with an intellectual disability
Meaning unit	<i>'I went to another room nearby when the midwife was talking... it was not just because of her talking... it was about being together, it happens often but I don't suffer from it, it was more likely that the others were thinking that I was sitting alone, it is difficult to explain because it is so many things...when I came home I was totally exhausted...'</i>
Condensed meaning unit	Being with others in a parent group resulted in tiredness and a feeling of exhaustion
Code	Diagnosis intellectual disability and tiredness
Sub-category	Advantages and disadvantages with parental education
Category	The significance of having an intellectual disability

Ethical considerations

Women with ID who were approached for interviews about their experience of pregnancy and childbirth were in a vulnerable situation (paper III). To minimise an intrusion into the world of the woman with ID they decided themselves if, where and when to meet. We informed key persons to tell women with ID, who they assumed were eligible to participate, about the study. Women received both oral and written information from the researcher and we also advised them to inform and discuss the participation of the study with their trustees or a trusted person. Furthermore, the researcher offered to inform their trustees or/and any trusted person about the study if they wanted. The women were informed that they could withdraw from the study at any time without explanation. When the women gave their written consent, an assurance of confidentiality was granted. If any woman experienced any problem after the interviews she was offered referral for further counselling. Since the addresses in study IV were obtained from a public register, no ethical review was needed according to Swedish law, and participation was entirely voluntary; however, the information on personal data was assured to be handled confidentially. The

procedure followed the ethical principles outlined by the Helsinki declaration (WMA Declaration of Helsinki - Ethical Principles for Medical Research Involving Human Subjects). Papers I, II and III were approved by the Regional Research Ethics Committee in Uppsala.

RESULTS

The results are presented as summaries of the papers.

Paper I

The mean age of the women was 24.2 (range 16-46) and the age distribution is shown in Figure 3.

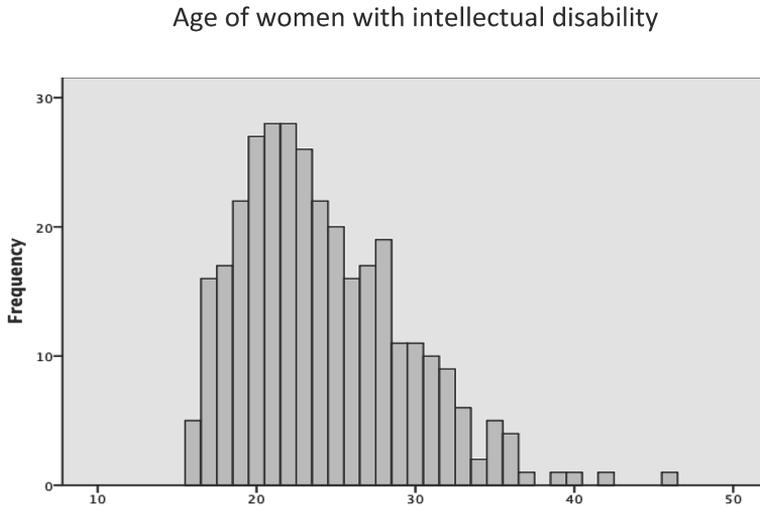


Figure 3. Age distribution of participating women

The women with ID who came to register at the antenatal care were more often: teenagers (11-19 years) (18.4% vs. 3.3%), obese (20.1% vs. 8.6%), daily smokers (27.9% vs. 7.9%) and single (36.6% vs. 6.2%) compared to women without ID or any psychiatric diagnosis. Preterm births were doubled for women with ID (12.2% vs. 6.1%) and deliveries ended more often with CS (24.5% vs. 17.7%). The women used less nitrous oxide as pain relief during delivery (59.5% vs. 75.8%), which remained the same when CS was excluded (71.9% vs. 84.1%). In a logistic regression model (adjusted OR),

women with ID had a higher risk for preterm birth (OR 1.68), CS (OR 1.55), non-use of nitrous oxide (OR 1.89) and discharge from hospital to a place other than home (OR 2.24) (Table 3).

Table 3. Adjusted odds ratios (95% confidence interval) for preterm birth, CS, non-use of nitrous oxide and discharge to a place other than home

	Preterm birth	CS	Non-use of nitrous oxide	Discharge to a place other than home
Variables	OR (95% CI)	OR (95% CI)	OR (95% CI)	OR (95% CI)
Women with ID*	1.68 (1.06-2.67)	1.55 (1.11-2.17)	1.89 (1.43-2.50)	2.24 (1.22-4.17)
Maternal age (year)**	1.01 (1.01-1.01)	1.09 (1.08-1.09)	1.03 (1.03-1.03)	0.99 (0.98-1.00)
BMI***	1.40 (1.33-1.48)	1.89 (1.83-1.95)	1.19 (1.16-1.23)	1.33 (1.23-1.45)
Cohabitation+	0.97 (0.90-1.05)	1.00 (0.95-1.05)	1.16 (1.11-1.20)	0.94 (0.84-1.06)
Working++	0.96 (0.91-1.01)	1.06 (1.02-1.09)	1.24 (1.20-1.27)	0.84 (0.78-0.91)
Smoking+++	1.14 (1.07-1.21)	1.18 (1.13-1.22)	1.16 (1.12-1.20)	1.12 (1.02-1.23)
Epilepsy++++	1.07 (0.84-1.36)	1.25 (1.08-1.45)	1.12 (0.98-1.28)	1.51 (1.10-2.08)

* Dichotomous women without intellectual disability (ref.) vs. women with intellectual disability

** Maternal age is a continuous variable

*** Dichotomous not obese (BMI \leq 29.9) (ref.) vs. obese (BMI \geq 30)

+ Dichotomous cohabitation with the child's father (ref.) vs. no cohabitation with the child's father

++ Dichotomous working (ref.) vs. not working

+++ Dichotomous not smoking (ref.) vs. smoking

++++ Dichotomous not epilepsy (ref.) vs. epilepsy

Paper II

The children born to mothers with ID had a higher proportion of cases of stillbirth (1.2% vs. 0.3%) and perinatal death (1.8% vs. 0.4%) than children born to mothers without ID. These children were more often born via CS (24.5% vs. 17.7%), had preterm birth (12.2% vs. 6.1%) and SGA (8.4% vs. 3.1%) and a lower Apgar score (<7 points) at 5 minutes (3.7% vs. 1.5%), compared to children born to mothers without ID. In a multivariate analysis, an increased risk of SGA (adjusted OR 2.25), stillbirth (adjusted OR 4.53) and perinatal death (adjusted OR 4.25) remained for these children (Table 4).

Table 4. Crude OR (95% confidence interval) and adjusted OR (95% confidence interval) for preterm birth, Apgar <7 points at 5 minutes, SGA, perinatal death and stillbirth in women with ID compared to women without ID

	Women with ID vs. women without ID			
	Crude OR	(95% CI)	Adjusted OR	(95% CI)
Preterm birth	2.15	(1.55-3.00)	1.58	(0.99-2.51)
Apgar <7 points at 5 minutes	2.59	(1.45-4.61)	1.83	(0.81-4.15)
SGA	2.86	(1.93-4.24)	2.25	(1.37-3.69)
Perinatal death	4.37	(1.95-9.19)	4.25	(1.72-10.50)
Stillbirth	3.93	(1.46-10.56)	4.53	(1.65-12.44)

Adjusted for maternal age, obesity, cohabitation with the child's father, working, smoking, epilepsy and CS

Paper III

Table 5. Characteristics of the women with ID, pregnancies, births and current care of their children

Age	Civil status	Parity	Vaginal delivery	CS	Mother as the caregiver	Child in foster care
21	S	1	1	0	0	1
26	C	2	2	0	0	2
29	NP	1	1	0	0	1
31	M	1	1	0	R	0
31	M	2	2	0	0	1
32	M	2	1	1	2	0
36	C	3	2*	1	3	0
38	S	7	7	0	5	2
38	C	1	1	0	1	0
41	M	2	0	2	2	0

M – married to the child/children’s father; S – single; C – cohabited with the child/children’s father; NP – lived with a new partner; R – the parents and their child lived at a treatment home for six months; * included an instrumental delivery at birth (vacuum extraction)

The characteristics for women with ID varied in demographics, the history of pregnancy, mode of delivery and care of their child/children (Table 5). It became clear in paper III that the women with ID struggled for motherhood despite having welcomed the pregnancy as a happy event in their lives, even if some pregnancies were unplanned. They encountered mixed reactions from partners and relatives about the pregnancy, and an induced abortion was suggested to some women. The women with ID experienced the pregnancy as a responsible life event, but avoided disclosing the diagnosis due to fear of losing custody of the child. They desired either parent-education in a parent-group with other people with ID, or in a group led by a midwife having special knowledge or as individual parent education with repeated information. They stated that the birth process was sometimes hard to understand and they used several strategies to handle the pain and strain of labour. The women had different modes of delivery and some suffered from pre-eclampsia or had preterm births. All the women with ID breastfed their children initially after birth. They felt fear and distress about the issue of custody of the expected and newborn child, and not all women currently had custody of their children.

Paper IV

Table 6. Socio-demographic characteristics of the midwives working at antenatal care services and delivery wards in study IV

	Midwives at antenatal care services (n=180)	Midwives at delivery wards (n=192)	All midwives (n=375) %
Age years (mean)	51.2 (range 28-67)	49.7 (range 28-67)	
Working years (mean)	12.6 (range 1-42)	16.9 (range 1-43)	
Working in Stockholm, Gothenburg, Malmö (including suburbs)			29.6
Working in other cities			60.9
Working in municipalities			9.5

In paper IV, a total of 375 midwives at antenatal care services (n=180) and delivery wards (n=195) participated. The mean ages of midwives were quite similar (51.2 years vs. 49.7 years) (Table 6). A majority of all midwives (97.1%) reported that it is different to care for women with ID in pregnancy and childbirth than caring for women without ID, and almost one-half (47.3%) had not received any specialised education about women with ID. Most of the midwives requested evidence-based knowledge of women with ID (95.4%). A high proportion (69.7%) of the midwives affirmed that women with ID could not manage the mother role satisfactorily and more than one out of three (35.7%) of the midwives considered that women with ID should not have children at all. The majority of midwives described that children of women with ID should grow up with parents and receive support from society (87.0%), but almost one-fifth (19.1%) stated that children should grow up in foster care.

In a multivariate analysis there were no associations between the statements: *women with ID should not become pregnant at all, women with ID should*

have the children they want, women with ID can manage their maternal role satisfactorily, the children of mothers with ID should grow up in foster care and demographic characteristics, experience of caring for women with ID or level of knowledge about women with ID.

An index based on questions assessing knowledge of ID was calculated, with a maximum score of 12 points. The mean value for the participating midwives (n=251) was 8.93, and more than one-third (36.3%) had good knowledge (range 9–12 out of maximum 12 points). The distribution is shown in Figure 4.

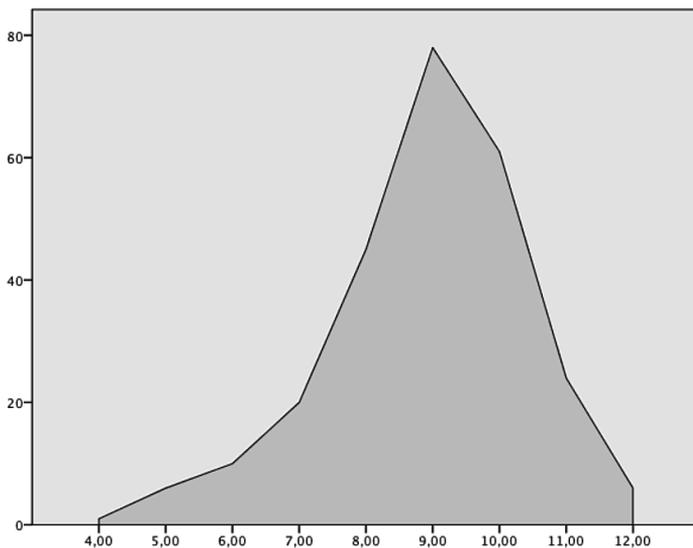


Figure 4. Midwives' knowledge of women with ID in an index ranged 0 -12 points

DISCUSSION

Methodological considerations

Paper I and II

Papers I and II are probably the only studies that have investigated pregnancy and birth outcomes in a national sample of women with ID. It is a unique possibility to perform such a study in Sweden since there is uniform antenatal care and almost entirely hospital births. Both the registers (MBR and NPR) have a high degree of coverage and the unique PIN-number allows for the linking of these two registers.

The nationwide coverage of care in the Patient Register emanates since 1987 for institutional care and from 2001 for non-institutional care. Three percent of the patients, who had an entry indicating a contact with psychiatric care from 1971 to 2007, lacked a psychiatric diagnosis. The Swedish Medical Register covers 98-99% of all pregnancies and deliveries since 1973. The register is internationally unique. In the register there are shortcomings with regard to important variables such as education levels and living conditions, which could be indicators of socio-economic conditions. There are sources of error e.g. incorrect documentations, missing medical records, wrong diagnosis and deficient transmission to the register (66). In the first two papers we had no data on the underlying causes of ID or its degree of severity. In the comparative analysis we used a more stringent sample with only single pregnancies and primiparas, for analysis of pregnancy and birth variables of interest to conduct the study during the above timeframe. The cohort constitutes a well-defined sample of women with ID and their children during pregnancy and childbirth. The sample of women with ID and the access to the cohort of women without ID or any psychiatric diagnosis, who were pregnant and gave birth in Sweden from 1999 to 2007, allows for a generalisation to populations with a similar socio-demographic structure as in Sweden.

We revealed several differences between women with ID and their children and women without ID or any psychiatric diagnosis and their children. The proportion of missing values in all variables was similar in the two groups

except for “working at registration” where data was missing for 24.5% of women with ID and 14.2% of women without ID. Missing values were excluded from the analysis. Interpretations of “working at registration”, however, must be interpreted with caution, since in many cases the occupation for women with ID is not paid employment but rather day-care activities. In paper II we selected variables with good quality for comparison (i.e. few missing cases) and those that previous studies found important (i.e. preterm birth, birth weight). All psychiatric diagnoses were excluded to eliminate incorrect adjacent classifications in the group of women without ID, which could mean that this group was healthier than the general population.

Paper III

In paper III we used a modified snowball sampling method with key persons to identify eligible women. We therefore relied on the judgment of these key persons in the selection of eligible women. However, all the women clearly met the inclusion criteria and it was a strength that they were recruited from different parts of the country, since support services may differ across counties. A semi-structured topic guide was used, covering experiences of pregnancy and childbirth. Important themes that should be included were: ‘how did you experience your pregnancy’, ‘how did you experience your delivery’ and ‘how did you experience your first time with the newborn child at the delivery ward and/or the postpartum ward’. Two interviews with each woman ensured comfortable situations with the opportunity to expand or to change the story.

The interviews were conducted in a relaxed and reflective atmosphere with simple language, and with a tape-recorder if the woman permitted. The researcher (BH) conducted all the interviews. The women with ID could have found it difficult to deny participation to those people whom they trusted and depended on despite the offer to inform/discuss with a trustee or a trusted person and the two-day interview set up. This may have been balanced with a researcher who was familiar with the context of pregnancy and childbirth and could facilitate for women to articulate their experiences. Barron described that unpredictable events could occur when interviewing women with ID, but also that the women were happy and grateful to share the story about their lives (1). The interviewer has to be prepared for that and also be flexible and sensitive.

The judgement of trustworthiness in qualitative studies is based on credibility, dependability, transferability and confirmability (94). To ensure credibility, we described the entire process in detail and inserted quotations to make it possible to judge the credibility of our findings. Dependability

was created by recruiting women from different parts of Sweden and by having two interviews during two consecutive days with the same women, using the same topic guide. To establish confirmability, the researchers created an open dialogue. The authors discussed the interpretation of the data, until consensus was reached (92). Lastly, transferability refers to the extent to which findings can be transferred to other non-Swedish women. The purpose with qualitative studies is to gain a deeper understanding of people's lived experiences, but striving for generalisations are neither desirable nor possible (95).

Paper IV

In study IV we used a public register of health professionals (The Health and Care Address Register) and randomly selected six hundred midwives working at various antenatal care clinics and delivery wards. There is a weakness in paper IV since we could not find any previously validated instrument. Therefore, we developed a questionnaire and validated it through face validity and test-retest with a two-week interval among 40 midwives who met the inclusion criteria. The questionnaires were sent by mail to six hundred midwives and two reminders were sent. The response rate was 62.5% (antenatal care 60.0% and delivery wards 65.0%) and the internal response loss varied between 0 -15%. This could mean that the midwives were reluctant to disclose sensitive opinions or they found the survey too long or uninteresting. Another explanation for non-response could be that midwives who lacked experience of caring for women with ID felt less inclined to participate. On the other hand, response rates for academic studies with similar designs are usually between 50-60%, and a response rate of 62.5% must be considered acceptable (96, 97).

In papers I, II and IV the authors consider the reliability to be high and that results could be generalised to countries with a similar socio-demographic structure, conditions and an organisation of care as in Sweden. There was no intention to generalise the findings from the qualitative study (paper III); however, we believe that in theory, similar findings would occur in settings with a context as in Sweden.

Reflections on results

Several important differences were observed in pregnancy, delivery and newborn health outcomes between woman with ID and women without ID and their children, respectively. Women with ID were more often teenagers, obese and single at the time of registration at the antenatal care than women without ID. A previous Swedish study affirmed that early childbirth (<20

years of age) was related to being single (98). Previous studies affirmed that a high BMI is related to persons with ID (99, 100). A high proportion of women with ID smoked at the time of registration at the antenatal care. Possible reasons could be their younger age and that they had not received any information about smoking in relation to pregnancy, or that this information was not understood. Also, the ability or the wish to stop smoking may be different than that of women without ID. Raatikainen et al. affirmed that smoking in early gestation or through pregnancy was related to a young age and being a primipara (101). In Sweden, there is a long tradition of antenatal care services. Midwives are the predominant profession who encounter most of the pregnant women with ID; thus, they could play an important role with counselling and providing support to make healthy choices.

More frequently, the women with ID had epilepsy compared to women without ID. Investigators have indicated that epilepsy is associated with an ID diagnosis (102-104). In women with ID, there was less spontaneous onset of labour and more CS. There are several reasons for induction of labour e.g. pre-eclampsia, which was found to be overrepresented in women with ID (10). However, data on pre-eclampsia could not be obtained from our dataset. The high CS rate could be explained by less spontaneous onset of labour, less instrumental delivery with vacuum extraction, more preterm births and more children being SGA.

Women with ID used less nitrous oxide as pain relief during delivery even when CS was excluded. The authors concluded that less use of nitrous oxide may depend on the women's knowledge, her ability to express her needs and requests, on the midwives' knowledge, prejudice, and sensitivity towards these needs and also on the higher frequency of CS. Women with ID had a two-fold increase of preterm births compared to women without ID. The preterm birth could be either spontaneous or induced; however, this could not be determined in this study. Other studies indicate similar data on preterm births (9, 101). Women with ID were more often discharged to a place other than home compared to women without ID. It could mean their newborn children needed care at a neonatal intensive care unit or a referral for assessing parental skills at a residential/treatment home. An Australian study reported that it was more common for children born to mothers with mild to moderate ID to be admitted to a specialised care unit (105).

Stillbirths were four-fold, and perinatal deaths were more than four times more prevalent in children born to women with ID compared to children born to women without ID. Perinatal death in these children was associated with maternal age, obesity, unemployment and smoking. Other studies confirmed that smoking (106-108) and obesity in pregnancy (107, 109-114)

had an association to stillbirth. The children born to women with ID were more frequently born preterm, more often SGA and had a lower Apgar score (<7 points) during the first five minutes. An Australian study described that Apgar score at 1 minute was not different in children born to women with ID than children born to women without ID (10). Being born preterm to women with ID was associated with maternal age, obesity and smoking. This is described in other previous studies (115, 116).

Even though the sterilisation law disappeared in 1975, the women with ID still perceived many barriers in relation to pregnancy and childbirth, and they struggled to attain motherhood. Several of the activities in Revin's theory of attainment of the mother role were described by the women, such as yearning for the child during pregnancy (taking-on activity), participating in parental education (taking-in activity) and disclosing her diagnosis and giving up smoking (letting-go activity). Nevertheless, it was clear that the women had difficulties in reaching the four goals in that theory: physical well-being, social acceptance, attachment to the baby and understanding the complexities of motherhood.

Authorities, professionals and relatives restricted and sometimes even violated their own autonomy in decision-making about themselves and their unborn and newborn child. Other studies have described similar negative reactions to pregnant women with ID (30, 48, 78, 79, 117, 118). According to Jean Ball's theory, the support from family and relatives is an important prerequisite for the attainment of maternal well-being. If this support is lacking, this could undermine and hinder the transition to motherhood.

Some of the children of the women in paper III were cared for at a neonatal care unit, which is in line with other studies (10, 119). One of the women described her experience of the neonatal care unit as a scary place with all the technical equipment. Other studies have described mothers' experiences and feelings of being separated from their premature or sick children. They perceived themselves as being an excluded outsider with lack of control, and anxiety and stress were predominant feelings (120-122). Women with ID may even be more bewildered and confused in this particular situation and would need extra care and support to be able to participate in the care of their newborns.

Women with ID were anxious and distressed about the custody of their expected/newborn child and not all women had the custody of their children. Other studies have confirmed emotional distress in parents with ID when children were removed from their care (31, 123-125). Losing custody of a child born to a woman with ID is not uncommon, nevertheless, a painful deprivation for these mothers. Professionals need to identify and support

these women, who may not always disclose their diagnosis, for fear of losing custody of their children.

Some midwives evaluated women with ID as unable to satisfactorily care for their children. Researchers have previously recognised this also in other services (12, 23-25, 48). A high proportion of midwives stated that women with ID should refrain from having children and some of the midwives were of the opinion that children should grow up in foster care. They felt uncertainty about how to adapt and provide care and requested evidence-based knowledge. One of the most important concepts in Jean Ball's theory for the attainment of maternal well-being is the support from health services. If this support is lacking or is deficient, the deck chair will collapse and the woman will not be able to obtain maternal well-being. A Swedish study described that midwives related to expectant mothers both in a distanced and in a caring way. The predominant topics in the consultations were physical aspects of pregnancy, childbirth and parenthood, and the transition to motherhood was described as a risky project for their bodies. The midwives steered the consultations and the expectant parents complied with this pattern. The meanings of pregnancy shifted between understanding the transition to motherhood as trustworthy or as unreliable. It is important that midwives reflect on their own understanding of pregnancy, childbirth and parenthood. This is developed and also displayed in the encounters with pregnant women (84, 126, 127).

A high proportion of the midwives in our study considered that it was different to care for women with ID than for women without ID. Moreover, they had limited knowledge about women with ID and doubted the parental ability of a woman with ID. The midwives' lack of knowledge and their hesitant attitudes may influence the quality of care that they provide to the woman and thereby, have an impact on her transition to parenthood. It is therefore possible that women with ID will not receive the optimal support they need from the midwife during pregnancy and childbirth. Based on these results we, therefore, recommend that health professionals in general and midwives in particular need improved knowledge in relation to childbearing in women with ID. Basic midwifery education as well as continuous in-service education should focus on how best to provide medical and psychosocial care and support to women with ID during pregnancy, childbirth and the postpartum period.

CONCLUSIONS AND IMPLICATIONS

The results in this thesis describe pregnancy and childbirth in women with ID in Sweden, the health of their newborn children and also midwives' knowledge of, experiences in and attitudes towards childbearing in women with ID.

Women with ID compared to women without ID or any psychiatric diagnosis:

- Were more often teenagers, obese and single
- Had a higher incidence of smoking at the time of registration at antenatal care services,
- Had more preterm births and CS
- Used less nitrous oxide as pain relief during labour
- Were more often discharged from hospital to a place other than home
- They struggled to attain motherhood and displayed a vulnerable situation
- They avoided disclosing the diagnosis due to fear of losing custody of the child
- The birth process was sometimes difficult to understand but the women expressed initial warm feeling towards their children
- The clinical procedures and the hospital environment were not always supportive for the women
- Their children had a higher proportion of preterm birth, SGA, CS, lower Apgar score (<7 points) during the first five minutes
- Their children died more often perinatally
- A majority of midwives affirmed that women with ID could not satisfactorily manage the mother role
- One out of three midwives considered that women with ID should not have children at all
- The majority of midwives stated that children should grow up with parents and with support from society, but one-fifth considered that the children should grow up in foster care

Implications based on these findings are that women with ID and their children are vulnerable and should be considered as risk groups in pregnancy and childbirth. Professionals in maternity services need to elucidate the

knowledge and skills for counselling this particular group of pregnant women in pre-, intra- and postpartum care. Professionals need to identify women with ID and support them in their transition to motherhood. Midwives need evidence-based knowledge about women with ID in pregnancy and childbirth since they may underestimate the capacity of women with ID with regard to the mother role. Pregnant women with ID should be individually supported to help them cope with feelings of anxiety and fear of losing custody of the child. A potential way to improve the health of their newborns may be to care for women with ID through an extended individual-based care with more accessible, interactive and better-tailored information and support. Professionals in the health care system must be facilitated and ensured cooperation in a multidisciplinary system during their care for women with ID.

The following recommendations suggest a strengthening of the psychosocial work in maternity services.

- National guidelines for maternity care services are needed to establish clear evidence-based instructions, which ensure women with ID individualised and equal care and support similar to other women.
- Evidence-based parent education programs must be tailored to reduce distress in parenting and increase confidence and skills.
- Obstacles within the health care system and the society should be removed and professional cooperation in a multidisciplinary arena must be facilitated.
- Routines for continuous education and supervision, to establish and maintain competency and quality over time should be implemented.
- Ongoing development of methods and research regarding women with ID in pregnancy and childbirth should be encouraged.

Summary in Swedish – svensk sammanfattning

Det övergripande syftet med delstudierna i denna avhandling var att undersöka graviditets- och förlossningsutfall hos kvinnor med utvecklingsstörning, samt hälsa hos deras nyfödda barn. Ett annat syfte var att ta del av kvinnornas egna upplevelser av att vara gravid och att föda sitt barn. Ett ytterligare syfte var att beskriva barnmorskors arbete med och uppfattning om kvinnor med utvecklingsstörning under graviditet och förlossning. Utvecklingsstörning definieras enligt Världshälsoorganisationen (WHO) som en intelligenskvot <70, (medelvärde 100) en nedsättning av den adaptiva förmågan och diagnosen ställd före 18 års ålder.

I de två första delstudierna länkades Patientregistret (PAR) vid Socialstyrelsen till det svenska Medicinska Födelseregistret (MFR) (Socialstyrelsen). PAR syftar till att nationellt registrera den svenska sjukvårdskonsumtionen (dock ej primärvård ännu) och MFR registrerar årligen olika variabler och utfall i samband med graviditet och förlossning i Sverige. Ett flertal utvärderingar av dessa register har gjorts fortlöpande och de bedöms ha en hög validitet och täckningsgrad.

En grupp kvinnor med utvecklingsstörning samt en kontrollgrupp med kvinnor utan utvecklingsstörning eller annan psykiatrisk diagnos och deras respektive barn identifierades i Patientregistret länkat till MFR där datamaterialet hämtades ifrån.

Den tredje delstudien var en nationell intervjustudie med 10 kvinnor med utvecklingsstörning. Inklusionskriterier var att ha diagnosen utvecklingsstörning, ha fött barn mellan 2004-2010, ha ett verbalt tal och bo i Sverige. Den fjärde delstudien bestod av enkäter till 600 slumpvist utvalda barnmorskor vid barnmorskemottagningar och förlossningsavdelningar i Sverige. Totalt deltog 375 barnmorskor som besvarade frågor om sitt arbete med och uppfattning om kvinnor med utvecklingsstörning under graviditet och förlossning.

Resultatet från delstudierna I och II visade ett antal skillnader mellan kvinnorna med utvecklingsstörning och deras barn jämfört med kvinnorna utan utvecklingsstörning och deras barn i motsvarande variabler. Kvinnor med utvecklingsstörning var yngre och rökte i större utsträckning vid

inskrivning vid barnmorskemottagning samt hade oftare ett BMI >30. De födde oftare i förtid och med kejsarsnitt men använde mindre lustgas som smärtlindringsmetod under förlossningen.

Barnen födda av kvinnor med utvecklingsstörning föddes för tidigt, oftare med kejsarsnitt, var lätta för tiden (small for gestational age) och mådde sämre under de 5 första minuterna (Apgar <7 poäng) jämfört med barn till kvinnor utan utvecklingsstörning. Det var också vanligare att barn till kvinnor med utvecklingsstörning dog perinatalt (från graviditetsvecka 28 - 1vecka efter födelsen).

I intervjustudien framkom att kvinnorna kämpade för att etablera ett moderskap. Kvinnorna välkomnade oftast initialt sin graviditet men möttes av olika reaktioner från omgivningen såsom misstroende och råd om att göra abort. Flera av kvinnorna förstod att de var gravida när de kände illamående och inte fick sin menstruation. Några av kvinnorna uttryckte att det var ett stort ansvar att vara gravid och slutade därmed att röka. Kvinnorna upplevde ibland att det var svårt att förstå när en förlossning startade men detta blev alltmer uppenbart när ett starkt värkarbete kom i gång. Att föda barn upplevde några av kvinnorna som ett hårt arbete och det var svårt att förstå födelseprocessen både om den hade ett snabbt eller ett långsamt förlopp. Några av kvinnorna fick havandeskapsförgiftning och födde för tidigt eller med kejsarsnitt, och några av barnen vårdades vid en intensivvårdsavdelning för nyfödda. Kvinnorna kände ängslan och oro över att barnet kunde bli omhändertaget och avslöjade endast sin diagnos för barnmorskan under graviditeten om de ansåg att detta gynnade dem i deras situation. Hälften av kvinnorna hade inte vårdnaden om alla sina barn.

I enkätstudien angav de flesta av barnmorskorna att de saknade kunskap om kvinnor med utvecklingsstörning och deras graviditet och barnafödande och de efterfrågade evidensbaserad kunskap. En majoritet av barnmorskorna ansåg att kvinnor med utvecklingsstörning inte kunde hantera modersrollen på ett tillfredsställande sätt och mer än en tredjedel av barnmorskorna tyckte att kvinnor med utvecklingsstörning inte skulle bli gravida och föda barn överhuvudtaget. Flertalet av barnmorskorna svarade att barnen skulle växa upp med föräldrar och hjälp från samhället men en femtedel av barnmorskorna ansåg att barnen skulle växa upp i fosterhem.

Avhandlingsarbetet visar att kvinnor med utvecklingsstörning och deras ofödda/nyfödda barn har mer medicinska komplikationer under graviditet och förlossning och därför bör betraktas som riskgrupper. Kvinnor med utvecklingsstörning känner oro och rädsla att inte få vårdnaden om sina barn. Barnmorskorna anser att de har lite tidigare kunskap om kvinnor med utvecklingsstörning under graviditet och barnafödande och efterfrågar

evidensbaserad kunskap. Barnmorskor behöver evidensbaserade vårdprogram för gravida och födande kvinnor med utvecklingsstörning för att säkerställa att de får en individuell och likvärdig vård/stöd i likhet med vad andra kvinnor får. Utbildare och arbetsgivare har ett övergripande ansvar för barnmorskors möjligheter till utbildning, fortbildning, handledning och tvärprofessionellt arbete.

I framtida forskning skulle det vara intressant att följa upp barnen till kvinnor med utvecklingsstörning med avseende på deras hälsa under uppväxttiden. Det vore även intressant att intervjua ungdomar på boenden och deras föräldrar samt personal på dessa boenden beträffande attityder till och kunskap om sexualitet, reproduktion och fertilitetskontroll.

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REFERENCES

1. Barron K. Women with learning difficulties (re)constructing their self-identity. *Scandinavian Journal of Disability Research*. 2002;1:58-79.
2. Socialstyrelsen. Föräldrar med nedsatt begåvning och deras barn. Rapport 1990;24. Stockholm: Socialstyrelsen. Report No.: ISBN 91-38-11175-6. ISSN 1100-2808.
3. Hilier L, Johnson, K, Harrison, L. Sex, secrets and desire: people with intellectual disabilities talk about their lives. *Intellectual Disability Australasia*. 2002;23:6-8.
4. Llewellyn G, McConnell, D, Ferronato, L. You have to prove yourself all the time. People with learning disabilities as parents. Open University Press; 2005. p. 441-67.
5. Traustadóttir R, Johnson, K. Women with intellectual disabilities: finding a place in the world. London: Jessica Kingsley 2002.
6. Löfgren-Mårtenson L. Får jag lov? Om sexualitet och kärlek i den nya generationen unga med utvecklingsstörning. Gothenburg: Gothenburg University; 2003.
7. Llewellyn G, McConnell, D, Mayes R. Health of mothers with intellectual limitations. *Australian and New Zealand Journal of Public Health*. 2003;27:17-9.
8. McConnell D, Mayes, R, Llewellyn G. Pre-partum distress in women with intellectual disabilities. *Journal of Intellectual and Developmental Disability*. 2008;33:177-83.
9. McConnell D. Developmental profiles of children born to mothers with intellectual disability. *Journal of Intellectual and Developmental Disability*. 2003;2:122-34.
10. McConnell D, Mayes R, Llewellyn G. Women with intellectual disability at risk of adverse pregnancy and birth outcomes. *Journal of Intellectual Disability Research*. 2008;52:529-35.
11. Aunos M, Goupil G, Feldman M.A. Mothers with an intellectual disability who do and do not have custody of their children. *Journal of Developmental Disabilities*. 2004;10:65-79.
12. Booth T, Booth W, McConnell D. Care proceedings and parents with learning difficulties: Comparative prevalence and outcomes in an English and Australian court sample. *Child and Family Social Work*. 2005;10:353-60.
13. Llewellyn G, McConnell D, Ferronato L. Prevalence and outcomes for parents with disabilities and their children in an Australian court sample. *Child Abuse Neglect*. 2003;27:235-51.
14. McConnell D, Feldman M, Aunos M. Child welfare investigation outcomes for parents with cognitive impairment and their children in Canada. *Journal of Intellectual Disability Research*. 2008;52:722.
15. McConnell D, Feldman M, Aunos M, Prasad N. Parental cognitive impairment and child maltreatment in Canada. *Child Abuse Neglect*. 2011;35:621-32.
16. Sigurjonsdóttir HB, Traustadóttir R. Motherhood, family and community life. London: Jessica Kingsley (ed). 2000.

17. Taylor CG, Norman D. K, Murphy J. M, Jellinek M, Quinn D, Poitras F. G, Goshko M. Diagnosed intellectual and emotional impairment among parents who seriously mistreat their children: prevalence, type, and outcome in a court sample. *Child Abuse Neglect*. 1991;15:389-401.
18. Taylor SJ. Children's division is coming to take pictures? Family life and parenting in a family with disabilities. In SJ Taylor, R Bogdan, Z M Lutfiyya (eds), *The variety of community experience: Qualitative studies of family and community life*. Baltimore: Paul H. Brookes (ed)1995. p. 23-45.
19. Traustadóttir R, Sigurónsdóttir H. B. The mother behind the mother: Three generations of mothers with intellectual disabilities and their family support networks. *Journal of Applied Research in Intellectual Disabilities*. 2008;21:331-40.
20. Tymchuk AJ, Andron L. Mothers with mental retardation who do or do not abuse or neglect their children. *Child Abuse Neglect*. 1990;14:313-23.
21. Westad C, McConnell D. Child welfare involvement of mothers with mental health issues. *Community of Mental Health Journal*. 2012;48:29-37.
22. Mayes R, Llewellyn G, McConnell D. Active negotiation: Mothers with intellectual disabilities creating their social support networks. *Journal of Applied Research in Intellectual Disabilities*. 2008;21:341-50.
23. Edgerton RB. Foreword. *Journal of Intellectual and Developmental Disability*. 1999;24:1-2.
24. Espe-Sherwindt M, Crable S. Moving beyond the myths. *Topics in Early Childhood Special Education*. 1993;13:154-73.
25. McConnell D, Llewellyn G. Stereotypes, parents with intellectual disability and child protection. *Journal of Social Welfare and Family Law*. 2002;24:297-317.
26. Mirfin-Veitch B, Bray A, Williams S, Clarkson J, Belton A. Supporting parents with intellectual disabilities. *New Zealand Journal of Disabilities Studies*. 1999;6:60-74.
27. McConnell D, Llewellyn G, Ferronato L. Disability and decision-making in Australian care proceedings. *International Journal of Law, Policy and the Family*, 2002;16:270-99.
28. Mayes R, Llewellyn G. What happens to parents with intellectual disability following removal of their child in child protection proceedings? *Journal of Intellectual and Developmental Disability*. 2009 Mar;34:92-5.
29. Booth T, Booth W. Findings from a court study of care proceedings involving parents with intellectual disabilities. *Journal of Practical Intellectual Disability* 2004a;1:179-81.
30. Booth T, Booth W. *Parenting under pressure: Mothers and fathers with learning difficulties*: Buckingham: Open University Press; 1994.
31. Booth T, Booth W. Parents with learning difficulties in the child protection system: Experiences and perspectives. *Journal of Intellectual Disability Research*. 2005;9:109-29.
32. Ehlers-Flint ML. Parenting perceptions and social supports of mothers with cognitive disabilities. *Sexuality and Disability*. 2002;20:29-51.
33. Sigurjónsdóttir HB. *Family support services and parents with learning difficulties*. Sheffield: University of Sheffield; 2005.
34. Sigurjónsdóttir HB, Traustadóttir R. *Invisible families: Mothers with learning disabilities and their children* 2001.
35. Hayman R. Presumptions of justice: Law, politics and the mentally retarded parent. *Harvard Law Review*. 1990;103:1201-71.
36. McConnell D, Llewellyn G. Social inequality, the deviant parent and child protection practice. *Australian Journal of Social Issues*. 2005;40:553-66.

37. Booth T, McConnell D, Booth W. Temporal discrimination and parents with learning difficulties in the child protection system. *British Journal of Social Work*. 2006;36:997-1015.
38. Kollberg E. Omstridda mödrar, en studie av mödrar som förtecknats som förståndshandikappade. Göteborg: Nordiska Hälsovårdshögskolan; 1989.
39. Gillberg C, Geijer-Karlsson M, Rasmussen P. Children born to mentally retarded women: A 1-21 year follow-up study of 41 cases. *Socialmedicinsk Tidskrift*. 1983;60:260-5.
40. Starke M. Föräldrar med utvecklingsstörning - vad finns det för kunskap? Socialstyrelsen; 2005.
41. Starke M. Barn som har föräldrar med utvecklingsstörning. Socialstyrelsen; 2007; Available from: http://www.socialstyrelsen.se/Lists/.../9308/2007-131-8_20071319.pdf.
42. McConnell D, Llewellyn G, Ferronato L. Context-contingent decision-making in child protection practice. *International Journal of Social Welfare*. 2006;15:230-9.
43. Mayes R, Llewellyn G. Mothering differently: narratives of mothers with intellectual disability whose children have been compulsory removed. *Journal of Intellectual and Developmental Disability*. 2012;37:121-30.
44. Grunewald K. Från idiot till medborgare: Gothia Förlag AB; 2008.
45. Grönvik L. Definitions of disability in social science. Uppsala: Uppsala University; 2007.
46. Gillberg C, Soderstrom H. Learning disability. *Lancet*. 2003;362(9386):811-21.
47. Swedish Government Official Reports. Recommendations regarding legislation of the interruption of pregnancy, Pub. L. No. 1935:15.
48. Mayes R, Llewellyn G, McConnell D. Misconception: The experience of pregnancy for woman with intellectual disability. *Scandinavian Journal of Disability Research*. 2006;2-3:120-31.
49. Lag (1993:387) om stöd och service till vissa funktionshindrade.
50. Kock W. Medicinhistorisk exposé. Södertälje: Fingraf Tryckeri; 1989. p. 88.
51. Socialstyrelsen. International Classifications of Diseases and Related Health Problems, Tenth Revision (ICD 10): National Board of Health 1996.
52. <http://www.psych.org/MainMenu/Research/DSMIV.aspx>. [cited 2010 0325].
53. Cullberg J. Dynamisk psykiatri. Stockholm: Stockholm: Natur och Kultur; 2003.
54. Gustafsson C. Intellectual disability and mental health problems. Uppsala: Uppsala University; 2003.
55. <http://www.who.int/classifications/icd/en/>. [cited 2010 0325].
56. Andersson K. What are intellectual disabilities? Landstinget i Uppsala Län; [cited 2010 0515].
57. Langdon Down J. 1866. Available from: <http://www.habilitering.nu> (cited 2010 0325).
58. <http://www.nationalboardofhealthandwelfare/register>. [cited 2010 0325].
59. Cnattingius S, Ericson A, Gunnarskog J, Kallen B. A quality study of a medical birth registry. *Scandinavian Journal of Social Medicine*. 1990;18:143-8.
60. <http://www.nationalboardofhealthandwelfare/patientregister>. [cited 2010 0325].
61. <http://www.socialstyrelsen.se/publikationer2009/2009-125-15>. [cited 2010 0325].

62. Niklasson B, Almqvist P. R., Hornfeldt B, Klitz W. Sudden infant death syndrome and Ljungan virus. *Forensic Science, Medicine and Pathology*. 2009;5:274-9.
63. Källén AJ, Finnstrom O. O, Lindam A. P, Nilsson E. M, Nygren K. G, Olausson P. M. Cerebral palsy in children born after in vitro fertilization. Is the risk decreasing? *European Journal of Paediatric Neurology*. 2010;14:526-30.
64. Kanis JA, Johnell O, Oden A, Sembo I, Redlund-Johnell I, Dawson A, De Laet C, Jonsson B. Long-term risk of osteoporotic fracture in Malmo. *Osteoporosis International*. 2000;11:669-74.
65. Socialstyrelsen. *Statistics Health and Diseases. Pregnancies, deliveries and newborn infants*, 2008. [cited 2010 0318]; Available from: <http://www.socialstyrelsen.se/epc>.
66. Socialstyrelsen. *The Swedish Medical Birth Registry - a Summary of Contents and Quality*, 2003. [cited 2010 0325]; Available from: <http://www.sos.se/eoc/epceng.html>.
67. Gottvall K, Winblad B, Cnattingius S, Waldenstrom U. Birth centre care over a 10-year period: infant morbidity during the first month after birth. *Acta Paediatrica*. 2005;94:1253-60.
68. Eriksson SL, Olausson P. O, Olofsson C. Use of epidural analgesia and its relation to caesarean and instrumental deliveries-a population--based study of 94,217 primiparae. *European Journal of Obstetrics & Gynecology and Reproductive Biology*. 2006;128:270-5.
69. Surkan PJ, Stephansson O, Dickman P. W, Cnattingius S. Previous preterm and small-for-gestational-age births and the subsequent risk of stillbirth. *New England Journal of Medicine*. 2004 Feb 19;350:777-85.
70. Florica M, Stephansson O, Nordstrom L. Indications associated with increased cesarean section rates in a Swedish hospital. *International Journal of Gynaecology & Obstetrics*. 2006 Feb;92:181-5.
71. <http://www.sfog.se/ARG+intressegrupper/arg-forord/Info>. [cited 2010 0325].
72. Norstedts. *Medicine engelsk-svensk-engelsk*: PH Collin och Norstedts förlag; 1992.
73. Berg M, Lundgren I, Hermansson E, Wahlberg V. Women's experience of the encounter with the midwife during childbirth. *Midwifery*. 1996;12:11-5.
74. Callister LC, Vehvilainen-Julkunen K, Lauri S. Giving birth. Perceptions of Finnish childbearing women. *MCN: The American Journal of Maternal/Child Nursing*. 2001;26:28-32.
75. Waldenstrom U, Rudman A., Hildingsson I. Intrapartum and postpartum care in Sweden: women's opinions and risk factors for not being satisfied. *Acta Obstetrica et Gynecologica Scandinavica*. 2006;85:551-60.
76. Zasloff E, Schytt E, Waldenstrom U. First time mothers' pregnancy and birth experiences varying by age. *Acta Obstetrica et Gynecologica Scandinavica*. 2007;86:1328-36.
77. Waldenstrom U, Hildingsson I, Rubertsson C, Radestad I. A negative birth experience: prevalence and risk factors in a national sample. *Birth*. 2004;31:17-27.
78. Aunos M, Feldman M.A. Attitudes towards sexuality, sterilisation and parenting rights of persons with intellectual disabilities. *Journal of Applied Research in Intellectual Disabilities*. 2002;15:285-96.
79. Llewellyn G. *Intellectual disability and parenting: A shared experience*. Sydney: Sydney University; 1994.
80. Rudman Ingemarsdotter A. *Women's Evaluations of Intrapartum and Postpartum care*. Stockholm: Karolinska Institute; 2007.

81. Ellberg L. Outcomes of various care options in Sweden. Umeå: Umeå University; 2008.
82. <http://www.macmillandictionary.com>. [cited 2012 0630].
83. Berg M. A Midwifery Model of Care for Childbearing Women at High Risk: Genuine Caring in Caring for the Genuine. *The Journal of Perinatal Education*. 2005;14:9-21.
84. Olsson P, Jansson L. Patterns in midwives' and expectant/new parents' ways of relating to each other in ante- and postnatal consultations. *Scandinavian Journal of Caring Sciences*. 2001;15:113-22.
85. Moody LE. *Advancing Nursing Science through Research* 1990.
86. Bryar R, Sinclair M. *Theory for Midwifery Practice*. Second Edition ed: PALGRAVE MACMILLAN; 2011.
87. Morse JM. Editorial: If You Believe in Theories. *Qualitative Health Research*. 1992;2:259-61.
88. Rubin R. Attainment of a Maternal Role. Part I. Processes. *Nursing Research*. 1967a;16:237-45.
89. Rubin R. Attainment of the maternal role. 2. Models and referrants. *Nurs Res*. 1967b;16:342-6.
90. Rubin R. *Maternal Identity and the Maternal Experience* 1984.
91. Polit D, Beck C. *Nursing Research Principles and Methods*: Philadelphia: Lippincott Williams & Wilkens 2004.
92. Graneheim UH, Lundman B. Qualitative content analysis in nursing research: concepts, procedures and measures to achieve trustworthiness. *Nurse Education Today*. 2004;24:105-12.
93. Sandelowski M. Focus on research methods. Whatever happened to qualitative description? *Research in Nursing & Health*. 2000;23:334-40.
94. Lincoln YS, Guba E.G. *Naturalistic inquiry*: Sage Publications Ltd., Beverly Hills, CA; 1985.
95. Kvale S, Brinkmann, S. *Den kvalitativa forskningsintervjun*. Lund: Studentlitteratur AB, Lund; 2009.
96. Baruch Y. Response Rates in Academic Studies – A Comparative Analysis. *Human Relations*. 1999;52:421-38.
97. Malaney GD. You Still Need High Response Rates with Web-based Surveys 2002.
98. Olausson PO, Haglund B, Weitoft G.R, Cnattingius S. Teenage childbearing and long-term socioeconomic consequences: a case study in Sweden. *Family Planning Perspectives*. 2001;33:70-4.
99. Bhaumik S, Watson J. M, Thorp C.F, Tyrer F, McGrother C.W. Body mass index in adults with intellectual disability: distribution, associations and service implications: a population-based prevalence study. *Journal of Intellectual Disability Research*. 2007;52:287-98.
100. Walsh PN, Kerr M, van Schroyen Lantman-de Valk H. M. Health indicators for people with intellectual disabilities: a European perspective. *European Journal of Public Health*. 2003;13:47-50.
101. Raatikainen K, Huurinainen, P., Heinonen, S. Smoking in early gestation or through pregnancy: a decision crucial to pregnancy outcome. *Preventive Medicine*. 2007;44:59-63.
102. Espie CA, Watkins J, Curtice L, Espie A, Duncan R, Ryan J. A, Brodie M. J, Mantala K, Sterrick M. Psychopathology in people with epilepsy and intellectual disability; an investigation of potential explanatory variables. *Journal of Neurology, Neurosurgery & Psychiatry*. 2003;74:1485-92.

103. Forsgren L, Edvinsson S. O, Blomquist H. K, Heijbel J, Sidenvall R. Epilepsy in a population of mentally retarded children and adults. *Epilepsy Research*. 1990;6:234-48.
104. Matthews T, Weston N, Baxter H, Felce D, Kerr M. A general practice-based prevalence study of epilepsy among adults with intellectual disabilities and of its association with psychiatric disorder, behaviour disturbance and carer stress. *Journal of Intellectual Disability Research*. 2008;52:163-73.
105. Calderon-Margalit R, Qiu C, Ornoy A, Siscovick D. S, Williams M. A. Risk of preterm delivery and other adverse perinatal outcomes in relation to maternal use of psychotropic medications during pregnancy. *American Journal of Obstetrics & Gynecology*. 2009;201:579 e1-8.
106. Cnattingius S. The epidemiology of smoking during pregnancy: smoking prevalence, maternal characteristics, and pregnancy outcomes. *Nicotine Tobacco Research*. 2004;6 Suppl 2:S125-40.
107. Cnattingius S, Stephansson O. The epidemiology of stillbirth. *Semin Perinatology*. 2002;26:25-30.
108. Hogberg L, Cnattingius S. The influence of maternal smoking habits on the risk of subsequent stillbirth: is there a causal relation? *British Journal of Obstetrics and Gynaecology*. 2007;114:699-704.
109. Astolfi P, De Pasquale A, Zonta L. Late childbearing and its impact on adverse pregnancy outcome: stillbirth, preterm delivery and low birth weight. *Revue Epidemiology Sante Publique*. 2005;53 Spec No 2:2S97-105.
110. Brynhildsen J, Sydsjo A, Norinder E, Selling K. E, Sydsjo, G. Trends in body mass index during early pregnancy in Swedish women 1978-2001. *Public Health*. 2006;120:393-9.
111. Cnattingius S, Bergstrom R, Lipworth L, Kramer M. S. Prepregnancy weight and the risk of adverse pregnancy outcomes. *The New England Journal of Medicine*. 1998;338:147-52.
112. Naeye RL. Maternal body weight and pregnancy outcome. *The American Journal of Clinical Nutrition*. 1990;52:273-9.
113. Stephansson O, Dickman P. W, Johansson A, Cnattingius S. Maternal weight, pregnancy weight gain, and the risk of antepartum stillbirth. *American Journal of Obstetrics & Gynecol*. 2001;184:463-9.
114. Villamor E, Cnattingius S. Interpregnancy weight change and risk of adverse pregnancy outcomes: a population-based study. *Lancet*. 2006;368:1164-70.
115. Baba S, Wikstrom A. K, Stephansson O, Cnattingius S. Influence of smoking and snuff cessation on risk of preterm birth. *European Journal of Epidemiology*. 2012;27:297-304.
116. Kyrklund-Blomberg NB, Granath F, Cnattingius S. Maternal smoking and causes of very preterm birth. *Acta Obstetrica et Gynecologica Scandinavica*. 2005;84:572-7.
117. Booth T, Booth W. Unto us a child is born: the trials and rewards of parenthood for people with learning difficulties. *Australia and New Zealand Journal of Developmental Disabilities*. 1995;20:25-39.
118. Llewellyn G, Brigden D. First hand experience: parents with learning difficulties. *Disability, Pregnancy and Parenthood International*. 1995;11:10-2.
119. Sussmann J, McIntosh A, Lawrie S, Johnstone E. Obstetric complications and mild to moderate intellectual disability. *British Journal of Psychiatry*. 2009;194:224-8.
120. Lindberg B, Öhring, K. Experiences of having a prematurely born infant from the perspective of mothers in Northern Sweden. *International Journal of Circumpolar Health* 2008;67:461-71.

121. Nyström K, Axelsson K. Mothers' experiences of being separated from their newborns. *Journal of Obstetric, Gynecologic, and Neonatal Nursing*. 2002;31:275-82.
122. Wigert H, Johansson R, Berg M, Hellström A-L. Mothers' experiences of having their newborn child in a neonatal intensive care unit. *Scandinavian Journal Caring Sciences*. 2006;20:35-41.
123. Baum S, Burns J. Mothers with learning disabilities: experiences and meanings of losing custody of their children. *Learning Disability Research*. 2007;12:3-14.
124. Booth T, Booth W. A family at risk: multiple perspectives on parenting and child protection. *British Journal of Learning Disabilities*. 2004b;32:9-15.
125. Booth T, Booth W. The uncelebrated parent: stories of mothers with learning difficulties caught in the child protection net. *British Journal of Learning Disabilities*. 2006;34:94-102.
126. Olsson P, Jansson L, Norberg A. Parenthood as talked about in Swedish ante- and postnatal midwifery consultations. A qualitative study of 58 video-recorded consultations. *Scandinavian Journal of Caring Sciences*. 1998;12:205-14.
127. Olsson P, Jansson L, Norberg A. A qualitative study of childbirth as spoken about in midwives' ante- and postnatal consultations. 2000;16:123-34.

Appendix I

Interview guide Paper III

Introduction

Please, would you like to tell me about your experiences when you were pregnant and gave birth to your child?

Important themes that should be included

- How did you experience your pregnancy?
- How did you experience your delivery?
- How did you experience your first time with the newborn child at the delivery ward and/or the postpartum ward?

Appendix II

Questionnaire Paper IV

* Midwives at antenatal care

** Midwives at delivery ward

1. Are you female/male? (*/**)
2. How old are you? (*/**)
3. When was your midwifery exam? (*/**)
4. How many years have you worked in antenatal care? (*)
5. How many years have you worked at the delivery ward? (**)
6. Are you working in a large city, other city or a municipality? (*/**)
7. In what region in Sweden are you living? (*/**)
8. Is the organisation of antenatal care different for women with ID compared for women without ID? (*)
9. Is the organisation of childbirth different for women with ID compared to women without ID? (**)
10. When do you advice women with ID to register at antenatal care? (*)
11. Do you advice women with ID to come to the delivery ward at a special time and with some special person/persons? (**)
12. Do you advice women with ID to be accompanied at antenatal care by special person/persons during pregnancy? (*)
13. Have you ever received any knowledge about women with ID in pregnancy and delivery? (*/**)
14. Do you have you experience of caring for women with ID in pregnancy? (*)
15. Have you experience of caring for women with ID in childbirth? (**)
16. Is it different to care for women with ID compared to women without ID in pregnancy? (*)
17. Is it different to care for women with ID compared to women without ID in delivery? (**)

18. Do women with ID often miss booked antenatal care visits? (*)
19. Do women with ID understand the importance of avoiding smoking during pregnancy? (*)
20. Do women with ID understand the importance of avoiding smoking during delivery? (**)
21. Do women with ID understand the importance of avoiding alcohol? (*)
22. Do women with ID understand the importance of avoiding other drugs? (*)
23. Do women with ID understand the importance of a healthy diet? (*)
24. Do women with ID understand the importance of physical exercise? (*)
25. Do women with ID understand the importance of good habits (food and sleep)? (*)
26. Do women with ID understand what it means to become a mother? (*/**)
27. Are the needs of women with ID met by parental education in ordinary groups/ individually? (*)
28. Do women with ID feel fear and worry about losing custody of the child? (*/**)
29. Do women with ID feel fear and worry about being involved with Social Services regarding the custody of the child? (*)
30. Do women with ID feel fear and worry about being referred to residential care facility? (*)
31. Do women with ID feel fear and worry about being referred to treatment care facility? (*)
32. Do women with ID feel fear and worry about needing support from their parents and relatives? (*)
33. Do women with ID feel fear and worry about needing support from the society? (*)
34. Are women with ID prepared for the labour pain? (**)
35. Do women with ID have difficulty to cope with the labour pain? (**)
36. Do women with ID want early pain relief in labour? (**)
37. Do women with ID understand the different stages of the delivery process? (**)
38. Do women with ID understand that labour/delivery could last for more than one day and night? (**)
39. Do women with ID have difficulties in interpreting the signs and signals from the newborn child? (**)

40. Do women with ID need more support with breastfeeding than women without ID? (**)
41. Do women with ID need more support to care for the child? (**)
42. Do women with ID need more support to interpret the needs of the child? (**)
43. Should women with ID be pregnant and give birth at all? (***)
44. Should women with ID not have more than one child? (***)
45. Should women with ID have the number of children they want? (***)
46. Can women with ID manage the mother role satisfactorily? (***)
47. Should the child grow up in foster care? (***)
48. Should the child grow up with their parents with support from society? (***)
49. Should the child grow up with parents and support from own family and relatives? (***)
50. Is it uncertain how to adapt advice to women with ID during pregnancy? (***)
51. Is it difficult to give advice and support to women with ID during pregnancy? (***)
52. Is your education in line with the needs of birthgiving women with ID? (***)
53. Do you need new knowledge about birthgiving in women with ID? (***)
54. Do you need evidence-based knowledge about birthgiving in women with ID? (***)
55. Do you need knowledge about methods of care (investigations, surveillance) for birthgiving women with ID? (***)
56. Do you need knowledge about how to handle educational situations in birthgiving women with ID? (***)
57. Do you need knowledge about collaboration partners in care and support for birthgiving women with ID? (***)
58. Do you need knowledge about support from society for birthgiving women with ID? (***)
59. Do you need knowledge about supportive interventions at home for birthgiving women with ID? (***)
60. Do you need knowledge about residential care facility for birthgiving women with ID? (***)

61. Do you need knowledge about treatment care facility for birthing women with ID? (**/**)
62. Do you need knowledge of whether and when the child of a woman with ID is placed into custody? (**/**)
63. Does ID depend on hereditary causes? (**/**)
64. Does ID depend on biological causes? (**/**)
65. Does ID depend on any special causes? (**/**)
66. Do people with ID more often have epilepsy? (**/**)
67. Is ID an intellectual disability with an intelligence quotient less than 70? (**/**)
68. Is ID reducing the adaptive capacity? (**/**)
69. Do 1-2% of the population have an ID? (**/**)
70. Do people with ID have a higher pain threshold? (**/**)
71. Do people with ID have a lower pain threshold? (**/**)
72. Do women with ID have more children than women without ID? (**/**)
73. Do 25% of pregnant women with ID refrain from antenatal care? (**/**)
74. Are 10-15% of women with ID not having custody of their children? (**/**)

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