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PARENTS´ EXPERIENCES AND REACTIONS WHEN AN UNEXPECTED FINDING IN THEIR FOETUS IS REVEALED AT A ROUTINE ULTRASOUND EXAMINATION

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2009

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Lund University, Sweden, 2009

PARENT'S EXPERIENCES AND REACTIONS WHEN AN UNEXPECTED FINDING IN THEIR FOETUS IS REVEALED AT A ROUTINE ULTRASOUND EXAMINATION

– A MULTI METHOD STUDY

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ISBN 978-91-86253-77-6
Printed in Sweden by
Wallin & Dahlholm Boktryckeri AB

To my father and mother

CONTENTS

ABSTRACT

The overall aim of this thesis was to describe parent's experiences, reactions and needs in connection with a routine ultrasound examination during pregnancy when the foetus was found to have an abnormal ultrasound finding.

The data collection began in February 2005 and the one year cohort study was completed by March 2006. The participants in this study were each asked if they would also consider participating in the qualitative part of the study. This material was collected from 2005 to 2008. The two studies with a qualitative design comprised of 19 parents (10 mothers and 9 fathers), respectively 16 parents (9 mothers and 7 fathers). The parents were interviewed regarding their experience and how they managed the situation when their foetus was found to have choroid plexus cysts or the foetus was found to have an abnormality at a routine ultrasound examination. The material was collected and analysed using the grounded theory method according to Glaser. The two quantitative questionnaire-based studies have a comparative design with matched controls, respectively the entire cohort abnormal findings compared to normal findings. Also the parents´ psychological well-being was investigated. The questionnaires, one to be filled in before and one after the examination, contained in addition to background variables, the instruments State- Trait Anxiety Inventory (STAI), Sense of Coherence (SOC) and the index in Parents´ Expectations, Experiences, Reactions during Ultrasound (PEER-U State of Mind Index). The STAI-instrument was only used in one of the two quantitative studies.

The interview findings revealed that the parents appreciated the ultrasound examination but they did not consider it as a form for foetal diagnosis even if they knew it is a tool for detecting abnormalities. This could be important to remember when new forms or scanning programmes are introduced in the future. Most parents wanted to know about the findings revealed during the examination immediately, but required the complete information without prolonged delay in order to form a strategy for further management of the situation. They also expressed a need for a meeting with a physician or specialist with competent knowledge of the deviation, in connection with the examination. When a deviation is found the parents need someone to talk to, a midwife, physician and sometimes psychological counselling. Initially it seems as it is the abnormal ultrasound finding in itself that causes the parents anxiety, not the consequences of how severe the finding is. The analysis of the material, according to the ultrasound specific instrument State of Mind Index, showed that the parents who had an ultrasound examination, where abnormal findings were found in their foetus, were more anxious than the parents whose foetus was normal. This was not obvious when using STAI, probably because it is not an ultrasound specific instrument and therefore it seems to be more time-dependent than the State of Mind Index. The instrument State of Mind Index can also give information as to what could have caused the anxiety. The item specific analysis of the State of Mind Index showed that many of the statements in the index could be connected to the routines at the ultrasound department. Therefore, this instrument could be helpful for evaluations or the development of new routines.

ABBREVIATIONS

AF	Abnormal finding in the foetus
CPC	Choroid plexus cyst
CUB	Combined Ultrasound and Biochemistry
FUB	Riksförbundet för barn, unga och vuxna med utvecklingsstörning/ The Swedish National Association for Persons with Intellectual Disability
GT	Grounded Theory
IQR	Interquartile range
N/n	Number
NF	Normal findings in the foetus
NS	Not significant
PEER-U	Parents' Expectations, Experiences and Reactions to Routine Ultrasound examinations
SMER	Statens Medicinsk-etiska Råd/ The Swedish National Council on Medical-Ethics
SOC	Sense of Coherence (scale)
SPSS	Statistical Package for the Social Sciences
STAI	State and Trait Anxiety Inventory (scale)
STAI-S	State Anxiety
STAI-T	Trait Anxiety
WHO	World Health Organisation

ORIGINAL PAPERS

This thesis for the degree of Doctorate is based on the following papers referred to in the text by their Roman numerals:

I Information for better or for worse: interviews with parents when their foetus was found to have choroid plexus cysts at a routine second trimester ultrasound examination. Larsson A-K, Crang-Svalenius E, Dykes AK. J Psychosom Obstet Gynaecol 2009; 30(1):48-57.

II Parental level of anxiety, sense of coherence and state of mind when choroids plexus cysts have been identified at a routine ultrasound examination in the second trimester of pregnancy: a case control study. Larsson A-K, Crang Svalenius E, Maršál K, Dykes A-K. J Psychosom Obstet Gynaecol 2009; 30(2):95-100.

III Parents worried state of mind when fetal ultrasound shows an unexpected finding – a comparative study. Larsson A-K, Crang Svalenius E, Maršál K, Ekelin M, Nyberg P, Dykes A-K. *Journal of Ultrasound in Medicine, in press.*

IV Unexpected diagnosis of fetal abnormality: parents´ experiences. Larsson A-K, Lundqvist A, Crang Svalenius E, Dykes A-K. *Submitted*

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INTRODUCTION

The use of ultrasound techniques in obstetrics and gynaecology began already in the late 50´s and was the beginning of a new paradigm in this field. The technical development and use of ultrasound has developed enormously during the last sixty years and has significantly changed the outlook on pregnancy. In Sweden, ultrasound screening during pregnancy is included in the state recommended health care programme for pregnant women and nowadays few parents can imagine a pregnancy without an ultrasound examination [1, 2]. It has become a natural part of the pregnancy and antenatal care and most parents look forward to their ultrasound examination. Parents today know that ultrasound is a tool for the detection of anomalies, but their real purpose for accepting the offer of an examination is rather to get a confirmation of the pregnancy and to see that their foetus is healthy [3, 4]. A study by Dykes and Stjernkvist [5] showed that the ultrasound examination can enhance prenatal bonding.

Foetal diagnosis is not only about a child's well-being, it is also an ethical issue. The debate on ethical aspects regarding foetal diagnosis is constantly on-going within our society. The question appears to be, is foetal diagnosis a form of discrimination since the examination can lead to selective abortion and, in that case, is the ultrasound examination a tool for culling? Naturally it depends on who one asks but in the debate it is seldom that ultrasound examination is reflected upon as being a form of foetal diagnosis, as is the case in most obstetric units. The method is considered to be harmless to the foetus and, so far, no research has shown any harmful effects caused by ultrasound [6, 7], however, the ultrasound community is continuously monitoring for possible adverse effects. Up to now the introduction of longer scanning sessions with three dimensional (3D) or four dimensional (4D) technique has not been evaluated systematically [8]. Perhaps is it the fact that ultrasound is considered to be safe and offered routinely that makes it much easier to have a positive attitude towards this non-invasive prenatal diagnostic procedure than, for example, to amniocentesis [9]. New and probably also less risk filled methods, for both mother and foetus, than invasive testing may be offered to parents in the future. The question is will it be easier to accept an offer of an ultrasound examination due to its simplicity and non-invasiveness and think less about the consequences of the test – the result, and how to deal with it if it shows a deviation? Regarding foetal diagnosis and its purpose, there is a double context which is the parents´ point of view and that of the health professionals. The parent's purpose appears to be the possibility to get acquainted with their child and to see that it is healthy [10, 11, 4]. Society, with the health care system at the forefront, offers and performs prenatal diagnosis with the aim of monitoring foetal development and to detect foetal anomalies [12]. Recently private clinics offer social scanning type 'meet your foetus'.

Before new methods and screening programmes are offered to parents during pregnancy, the psycho-social part surrounding the situation must also be considered. This point is stated in The Swedish Medical Research Council's consensus declaration [13]. In previous research the focus has been on the pregnant woman. Hopefully this research project including both parents´ experiences in connection with an ultrasound

where their foetus was found to have an abnormal finding will influence the development of care, from this aspect, in new screening programmes.

BACKGROUND

The historical development of an ultrasound screening programme during pregnancy

Half a century ago, the development for the clinical use of ultrasound in pregnancy was started, in Scotland, by Prof. Ian Donald. He wrote a very important paper concerning obstetrical and gynaecological ultrasound [14]. One of his disciples, a pioneer of the use of ultrasound in obstetrics and gynaecology, in Sweden was Bertil Sundén [6]. In 1964, at Lund University in Sweden, he defended the first thesis relating to the diagnostic value of ultrasound in obstetrics and gynaecology. He described the method as being simple, quick and a method that did not involve any discomfort for the patient. Due to this new technology many X-rays examinations, which were potentially harmful to a foetus, were exchanged for an ultrasound examination. Among his many observations Sundén was the first to identify a twin pregnancy using ultrasound [15] and to describe a foetus with anencephaly [6].

Already in the mid 1970´s, pregnant women in Malmö, Sweden were systematically offered an ultrasound scan in pregnancy week 28. The purpose of this routine scan was to detect multiple pregnancies [16]. Over the following years the time for performing the examination was changed to the second trimester ultrasound examination we have today. The University Hospital in Malmö was the first obstetrical unit in Sweden to have a routine ultrasound programme for pregnant women. A couple of years later Malmö adjusted its programme to include one ultrasound scan in the second trimester and another one in the third trimester (week 32). Later this programme, first used in Malmö, was adopted by other hospitals in Sweden and by 1993 all obstetric units in the country could offer one routine ultrasound examination, predominantly in the second trimester. In the majority of the examinations, the foetal anatomy is scrutinised in order to detect potential anomalies [17].

When the ultrasound screening programme was offered to all pregnant women, the essential part of the examination was to determine gestational age, detect multiple pregnancies, viability and to locate the placenta [18]. Due to improved diagnostic methods and technology prenatal ultrasound examinations have gained acceptance as a powerful screening tool for the detection of foetal structural anomalies, even though this was not initially the main purpose. This was a development not reflected upon in the early days and caused a situation requiring management where experience was lacking. Another important discussion at the time was, is ultrasound a beneficial tool to use and is it something that every pregnant woman should be offered or is it something to be used only when specific indications occur? Different studies investigated the issue “systematic ultrasound examination in pregnancy”. Early in the process, Eik-Nes [19] in his thesis, presented the obvious value of ultrasound in

predicting foetal growth and possible foetal weight deviation at birth. Nielsen [20] found in his review, which was focused on routine ultrasound in early pregnancy before 24 weeks, results that supported previous findings by Waldenström et al [21] about an earlier detection of multiple pregnancies and a reduction of induction of labour for post term pregnancy. Further the review stated that an early scan appeared to enable a better gestational age assessment and earlier detection of anomalies. The consequence and benefit in this case is that it is possible to terminate the pregnancy at this time [19]. However no evidence was found regarding the effectiveness in anomaly scanning in the first trimester [22].

The ultrasound screening programme during pregnancy today

Today the ultrasound examination is a natural part of antenatal care and very few women decline the offer. Approximately 95% of the pregnant women in Sweden undergo the examination [17]. A routine ultrasound examination is usually performed at around 18-20 weeks of gestation since this is the point in time considered to be advantageous for diagnosing prenatal anomalies. The ultrasound scan includes a comprehensive examination of the foetal anatomy and is an essential part of the routine examination. In a few obstetric units in Sweden an ultrasound examination in the third trimester is also offered at around 32-33 weeks of gestation (ibid).

Foetal diagnosis is in a state of continuous development and new screening programmes are now being offered such as fetal nuchal translucency, screening combined with a blood test (free β-hCG and PAPP-A) for maternal serum analysis also called Combined Ultrasound and Biochemistry (CUB) [1.] The main emphasis of this screening programme is to detect the foetus at risk for Down syndrome – Trisomy 21 but also includes Trisomies 18 and 13 and is carried out in the first trimester [23]. This test is not available in all obstetric units, organised within the Swedish National Health Care System. Often it is only pregnant women aged 33 years (Lund University Hospital) or 35 years or more who are offered this examination [24]. Most of the women (95%) who undergo this combined screening test will receive a normal test result [25].According to a consensus statement made by the Swedish Research Council [13], parents who do not want to have an examination of the foetal anatomy i.e. foetal diagnosis, are to be offered an early ultrasound scan in the first trimester at 10-12 weeks of pregnancy, to determine the gestational age, the location of the placenta, viability and for the detection of multiple pregnancies.

Ethics in connection to foetal diagnosis

Today's parents know that the ultrasound examination is optional [4] and not many give it much thought. In a quantitative study from Sweden, 1004 women were offered four different options: i) no examination, ii) an abdominal ultrasound examination in pregnancy week 9-11 without a comprehensive examination of foetal anatomy, iii) routine ultrasound examination in pregnancy week 17-18 or iiii) an amniocentesis in

pregnancy week 13 followed by a routine ultrasound examination as in option number three. All the women chose to do some kind of ultrasound examination They did not think it was a difficult decision to make and 98,8% of the women were content with their choice [26]. However, since the parents have such a positive attitude towards this somewhat well-known examination beforehand and some kind of relationship to ultrasound, it is even more important to be assured that the parents are fully aware of what it is they are accepting. Therefore, the question is, can they give an informed consent to the examination when they often have to decide about it quickly and where often there is not much time to consider what the outcome of such an examination may be? In another study by Crang-Svalenius et al [27], 62% of the women had not understood that the routine ultrasound examination was optional. Although a study from 2004 [4] reported improvement in the parents feeling that they were well informed about the fact that ultrasound is optional. The next question is, does this include their own reflections upon how to deal with the ultrasound findings after a normal or an abnormal result? The World Health Organisation WHO [28] emphasises the importance of parents making an informed choice irrespective of their accepting or declining the examination. Garcia et al [29] states that to be able to make an informed choice, correct information must be available to parents concerning the purpose and limitations of the examination. Another important issue is that in order to make the right decision based on informed choice, parents must be able to understand and interpret the meaning of the test. The question arises, is it possible for the parents to make an informed choice in the culture that exists within the health care services and in a medicalised environment? Nicol [30] came to the conclusion that it is not possible in these circumstances.

Ethics are very much a matter of creating, promoting and maintaining awareness about how to act and perhaps react upon things [30]. Relational ethics are, according to Bergum and Dossetor [32], grounded in different relationships. At every level in a given situation a number of relationships are involved. In this context, that is to say an ultrasound examination, there are, for example specific relations between parents and care providers, parents and their families, practitioners and theorists. All kind of relationships are nurtured and sustained due to their reliance on dialogue and mutual concern which also supports the ethical reflection and decision making.

It is necessary that foetal diagnosis is discussed at different levels in society [33] and this is also the philosophy behind relational ethics which emphasise that society consists of relationships that guide us to participate in different situations and interactions and that this can influence the situation that is in focus [34]. The discussions regarding foetal diagnosis must be conducted with respect, an acceptance that different opinions exist and that there exists a lack of knowledge regarding how it is to live with a handicap [35, 36]. In the debate about foetal diagnosis there is a concern about the human value of people with Down syndrome, especially since the introduction of foetal nuchal translucency screening combined with maternal serum (CUB). This possibility can be looked upon as a culling method in order to avoid children with Down syndrome being born [37]. In practice, the findings of chromosome abnormalities will increase with this CUB-test and there is a risk that parents will choose not to have

these children. On the other hand, most anomalies that are discovered prenatally, are detected by ultrasound, which most parents accept to undergo, and are in fact not chromosome abnormalities [17, 11].

Parents are able to make their own choices regarding foetal diagnosis on the basis of their ability and possibility to take care of the child and what they think is best for the child [38]. However, the parents need to have knowledge and also time for reflection before they can make a proper decision. Parents in general do not have enough knowledge regarding how to interpret the ultrasound examination results, its limitations and what it can reveal [35]. It is a difficult task to give understandable information concerning foetal diagnosis, regarding the examination and the important issues to think about that are related to the results of the test, especially if it is possible to choose between different options. This is exemplified in a study where pregnant women were asked about their choice in connection with foetal diagnosis, unfortunately, mostly their impression was that they had not made any choice at all [39]. How and in what way the information is given has a crucial impact on how the ethical issues such as integrity and human dignity are dealt with [33]. Often foetal diagnosis is discussed as a tool for identifying anomalies and if an anomaly is found, the possibility of an abortion. For most parents foetal diagnosis is not about the possibility to request an abortion, rather a way to prepare themselves for the future [40]. The developments in foetal diagnosis have resulted in development in other fields as well, for example intrauterine operations and improved care and treatment of the newborn child [41].

Irrespective of what the parents decide to do, their decision is made from what they believe is the best for them and the foetus, based on the possibilities available to them at the time [38]. Parents also have the right to regret the loss they have had, be it the aborted child or their longing for a healthy child that was not born.

The midwife's part in foetal diagnosis

In Sweden a midwife is solely responsible for the care of a normal pregnancy. Both prior to and after the ultrasound examination the midwife has a counselling role. The health care programme for pregnant women includes approximately eight appointments between the midwife and the pregnant woman at a maternal care centre. During the first appointment an initial discussion regarding foetal diagnosis is instigated. The parents are given information about the ultrasound examination and if they are interested and accept the offer of the examination, the midwife gives them more detailed verbal and sometimes also written information about the examination. However, some parents have described the offer of a routine ultrasound examination as being handed over like a menu, without any of the pros and cons being attached [42]. An important part of the midwife's counselling in connection with foetal diagnosis is to have an opportunity to ask the parents questions concerning their knowledge on the subject. According to relational ethics, the moral in meeting with others is about asking questions, not in having all the answers [43]. By asking questions the midwife can help the parents to think beyond their normal limits - What will this examination do

for us? What will we do with the results from the examination? What possibilities do we have to take care of a malformed child? It is also the midwife at the maternity care centre who later meets with the parents after an ultrasound examination with abnormal findings and must now offer a supporting role in contact with the parents in crisis. In Sweden the routine ultrasound examinations are most often performed by specially trained midwives and they are entitled to tell the parents that all appears normal at the examination. If deviations from normal are found, the physician in charge must be contacted and it is he/she who presents the diagnosis.

The disclosure of an abnormal ultrasound finding

Pregnancy itself is a period in life when it is natural to feel anxiety. This feeling can, however, be intensified due to unexpected circumstances connected to the pregnancy. An abnormal ultrasound finding in the foetus is a typical example of an unexpected situation. Usually parents have mixed feelings about the ultrasound examination. They wish for it because they want to meet their child but on the other hand they can also feel anxious due to their knowledge that ultrasound is also a tool for detection of structural anomalies in a foetus [9]. It is natural to feel increased anxiety while waiting for a test result or after a negative finding. Previous studies have revealed how women experience their situation in connection with receiving information about an abnormal ultrasound finding and their encounters with different health care professionals [9, 44]. Conclusions drawn from these studies are that anxiety increases if the woman has to wait for information related to the ultrasound finding, whereas the continuity in their contact with health care professionals helps decrease anxiety. There is, however, a lack of studies focusing on both parents´ experiences when a routine foetal ultrasound examination reveals an abnormal finding. Furthermore, previously only generic instruments have been used for measuring parents´ anxiety in connection with an ultrasound examination. A new instrument, specifically focusing on the ultrasound examination, called Parents' Expectations, Experiences and Reactions to Ultrasound examinations, has been developed and used in studies relating to normal findings [45]. Therefore it would be of great value to use this instrument for investigation even when abnormal findings are identified in a foetus.

AIMS

The overall aim of this thesis was to describe parent's experiences, reactions and needs in connection with a routine ultrasound examination during pregnancy when the foetus was found to have an abnormal ultrasound finding.

Specific aims:

- to gain a theoretical understanding of parents´ experiences and handling of the situation when their foetus was found to have choroid plexus cysts at a routine second trimester ultrasound examination (paper I) or found to have a foetal abnormality at a routine second or third trimester ultrasound scan (paper IV)

- to compare parents' experiences of a routine ultrasound examination in the second trimester of pregnancy when choroid plexus cyst/cysts (CPC) were found (study group) or not found (control group). Furthermore, to evaluate how their sense of coherence, level of anxiety and state of mind differed before and after the ultrasound examination among and between the study and control groups respectively (paper II)

- to evaluate and compare parents' anxiety, sense of coherence and specific satisfaction, before and after, a second trimester routine ultrasound that revealed either normal or abnormal findings (paper III)

STUDY POPULATION AND METHODS

Design

The complexity regarding the issue of this thesis requires both a quantitative and a qualitative method. Grounded Theory was used in the two qualitative studies: "Information for better or for worse: Interviews with parent's when their foetus was found to have choroid plexus cysts at a routine second trimester ultrasound" (Paper I) and "Unexpected diagnoses of foetal abnormality: parents´ experiences" (Paper IV). The thesis was supplemented with two questionnaire-based studies with a quantitative approach in Papers II and III. Paper II is a case-control study that evaluates both the parents´ anxiety and further their sense of coherence when choroid plexus cysts were found at a routine ultrasound examination. Paper III focuses on evaluating anxiety and sense of coherence and comparing groups when the ultrasound examination showed normal or abnormal findings in the foetus. Also the PEER-U (Parents' Expectations, Experiences and Reactions to Ultrasound examinations), State of Mind Index was scrutinised. The design and number of participants related to the papers above are shown in Table 1.

Table. 1 Design, Method and participants

	Design, data collection	Participants (n)
Paper I	Qualitative interview study	19 parents (10 mothers and 9 fathers)
Paper II	Quantitative case-control study using questionnaires	22 parents with 66 matched controls
Paper III	Quantitative comparative study using questionnaires	66 parents with an abnormal finding (AF) in their foetus compared to a group of 1983 parents with normal findings (NF) in their foetus
Paper IV	Qualitative interview study	16 parents (9 mothers and 7 fathers)

Study population

Data sources for this thesis were from the main database of the one-year cohort [46] (Figure 1).

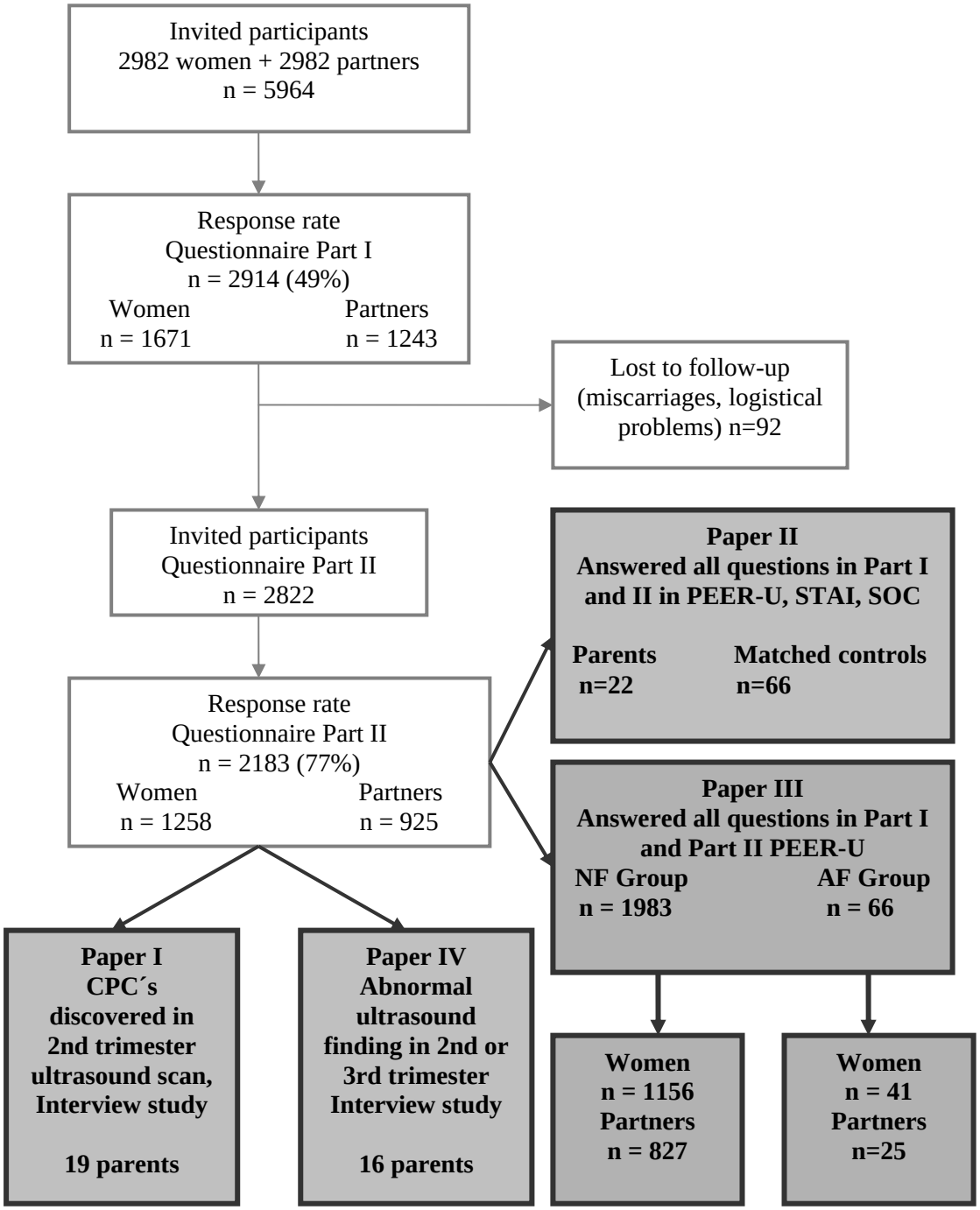

Figure 1. Flow chart participants Papers I, II, III and IV from the one-year cohort

Papers I and IV

The theoretical sampling is important in GT but also suitability and in these studies parents whose foetus was found to have choroid plexus cysts at a second trimester ultrasound examination (Paper I) and parents whose foetus had been diagnosed with an abnormality during an ultrasound examination in the second or third trimester (Paper IV) were included. Further the participants had to understand and converse in Swedish (Figure 1). Theoretical sampling is a process where the sample has been generated from data that prepossessed the sampling, not a sample that matches the whole population. All parents (Papers I and IV) had already given their written consent, through participation and answering a questionnaire that related their total ultrasound experience, as part of a larger ultrasound cohort study [46]. All couples were interviewed together. This began with one open question asking for their experience. All interviews were taped and transcribed verbatim.

Paper II

The first part of the questionnaire was sent to all of the women and their partners (n=2982x2) together with the appointed time for their second trimester ultrasound examination at Lund University Hospital during the period - February 10th, 2005 to March 30th, 2006. The first part of the two part questionnaire was sent out about two weeks before the examination. The envelope was addressed to the mother who was requested to give the enclosed copy to her partner. Those who answered the first part of the questionnaire were sent the second part after the ultrasound examination. Both questionnaires were only available in Swedish. Two reminders for the second part were sent out to the women if necessary. Parents with severe abnormal ultrasound findings in their foetus did not receive the Part II questionnaire by mail. From the cohort study, 22 parents whose foetus was found to have choroid plexus cyst/cysts at a routine second trimester ultrasound examination were included (Study group) and 66 parents, whose foetus examination revealed normal findings, served as matched controls (Figure 1). All items in each section of each instrument in the questionnaires had to be fully completed in order to be included in the study; otherwise the questionnaire was excluded.

Paper III

From the larger one-year cohort study, the study group of 2049 parents of whom 1983 parents (1156 women and 827 men) had a foetus with normal ultrasound findings (NF group) and 66 (41 women and 25 men) who had a foetus with abnormal ultrasound findings (AF Group) in the second trimester of pregnancy were included (Figure 1). Only the participants who had answered all the questions in the PEER-U, State of Mind Index, both in Part I and Part II, were included in this study. The second instrument the SOC was taken from the questionnaires as a background factor for indicating how the parents dealt with stressors in general.

Data collection

The data collection began in February 2005 and the one year cohort study was completed by March 2006 [46]. The author of this thesis took part in the organisation and data collection of the cohort study and the data material in Papers II and III was collected from the cohort. Additional material was collected by follow-up interviews regarding Papers I and IV between 2005 and 2008. The Ultrasound Department at Lund University Hospital in Sweden helped in recruiting the participants who were also included in the one-year cohort.

Context

The study took place at an obstetric unit located in a university city with approximately 107 000 inhabitants. Annually, the hospital has approximately 3000-3500 deliveries and about the same number of second trimester routine ultrasound examinations. The unit is a referral centre and the hospital's catchment area is the towns and villages in the surrounding area. In this city the educational level is above the average in Sweden [47] and the women's mean age at the time for the first delivery is approximately 29 years. In most cases, specially trained midwives perform the routine ultrasound examination. Before the examination the parents are informed, verbally and sometimes in writing about the ultrasound examination by the midwife at the maternity care centre. The information continues during the ultrasound examination and the parents can also follow the examination on a separate screen. In cases where the ultrasound examination shows abnormal findings in the foetus a physician is contacted to confirm or state the diagnosis and to inform the parents about the findings and future management.

Methods

Valid and used instruments from the questionnaire

The questionnaire PEER-U including the State of Mind index, developed by Ekelin et al [45] was used together with the State- Trait Anxiety Inventory STAI [46] with the full form of the trait scale (20 items) and a six-item version of the state-scale [49] together with the short form of the Sense of Coherence scale, SOC (13 items) [50].

The specific instrument PEER-U, Parents´ Expectations, Experiences and Reactions during Ultrasound with the specific State of Mind Index developed by Ekelin et al [45] were used for measuring the parents´ state of mind. The parents were asked to mark how well they agreed with the statements on a four point Likert scale [51] regarding statements of their views about their ultrasound examination. The choices were: *not at all, to some extent, quite a lot, absolutely*. Part I consists of 30 items measuring seven dimensions of expectation prior to the ultrasound examination. In Part II, 23 items measure five dimensions of experiences and reactions in connection with the ultrasound examination. The ultrasound specific index, State of Mind Index was analysed and included sixteen questions (statements) asked prior to the examination

(Part I) and thirteen after (Part II). The Cronbach's alpha for the State of Mind Index was 0.86 for Part I and 0.74 for Part II. The scores for the State of Mind Index ranged from 16 to 64 in Part I and from 13 to 52 in Part II. High scores indicate a higher degree of anxiety [45]. In order to make the index in Part I comparable to Part II, the State of Mind Index was linearly transformed to a range of 0-100 (Paper II and III).

STAI – State-Trait Anxiety Inventory Scale [48] which measures the state of mood at this moment (state) incorporates anxiety and nervousness. The trait measures average mood i.e. personality, and is relatively stable. Twenty items were used to evaluate the trait from responses on a 4-point scale: *almost never, sometimes, often, almost always.* The 6-item short version developed by Marteau and Bekker [49] was used to evaluate state and the choices were: *not at all, somewhat, moderately so, very much so.* This generic instrument is considered to have high stability and validity. The Cronbach's alpha is 0.90 for the trait part and 0.82 for the short version of state. The STAI-scores can vary from 20-80 and a low score stands for a low anxiety level. Spielberger [48] classifies the STAI scores as; low anxiety = <39, moderate anxiety = 40-59, severe anxiety = >60) (Paper II).

The instrument SOC, Sense of Coherence with the 13-item version [50] is a generic instrument as well. SOC is considered to remain stable during a lifetime, with little or no permanent change for important life events. The instrument can give values from 13 to 91. A high score expresses a strong SOC. The higher SOC score the better the capacity to cope with difficult situations. Langius et al [52] divided the SOC scores into; low = 35-60, moderate = 61-75, high = 76-91. According to Antonovsky [49] the 13-item version has a Cronbach's alpha of 0.82.

DATA ANALYSIS

Qualitative analysis, Grounded Theory

In papers I and IV the Grounded Theory (GT) analysis method was used. This method was created and used by two American social scientists, Barney Glaser and Anselm Strauss of the University of California, San Francisco [53]. Later they diverged and developed the method in two different directions and in this thesis Glaser´s version is used. In GT almost all data can be incorperated, both quantitative and qualitative, although it has a qualitative approach. The method is suitable for behavioural research. The objective of the method is to generate a theory or a model based on an inductive interactive process with a constant comparative analysis in order to systematically approach the data that explains the main concern and how it can be solved. In GT, theoretical sampling is central. The purpose is to make the discovery of categories and their properties, and their interrelationship, more systematic i.e. to make the process of the sampling concentrated towards the emerging theory [54]. In grounded theory the concept "validity" is not an issue that is discussed. The four concepts that are in focus for judging the validity of the material are; fit, relevance, workability, and modifiability [53, 55]. It is about how well the concepts represent the incidents, relevance

is about how interesting the readers find it, workability is about how well the results explain the participants' problem and how they solve it and if it is modifiable so that new data can alter the model. A grounded theory study can never be right or wrong it can only have more or less fit, relevance, workability and modifiability (ibid).

The analysis of the data began directly after the first interview and the purpose of this was to conceptualize the material. Memos were made after each interview and the information used later in the analysis. In the initial stage the open coding, line by line analysis was performed and each interview generated many substantive codes. These codes were closely compared for similarities and differences, some were grouped together and some new ones were generated and categories explaining the content emerged. When new interesting subjects were found, questions about this were asked in the subsequent interviews to enable better understanding of the essential in the narratives. From the analysis of the material an integrated and meaningful structure of the data was obtained and out of these categories the core category was selected. This category has according to Glaser [56] [24 p.75] "*several important functions for generating grounded theory: integration, density, saturation, completeness and delimiting focus*". When the core category was found, the selective coding begun, i.e. only the codes that were relevant for the core category were analysed. The deductive phase in GT is called "theoretical coding". The purpose of this phase is to conceptualize how the substantive codes can be interrelated as hypothesis in order to be integrated into a theory. The information in the memos was used even in this part of the analysis process when looking for the theoretical codes that conceptualized how the categories were related to each other. Glaser suggests, in his version of GT, eighteen theoretical code families which are examples of interrelations that can exist between the substantive codes.

Statistical analysis

In Paper II, from the cohort a sub-group with choroid plexus cysts found during the ultrasound scan was selected and matched with three controls, with normal ultrasound findings. The instruments PEER-U, STAI and SOC were analysed. For presentation of the results, median and interquartile ranges were used and the statistical evaluation was done with the non-parametric method Mann-Whitney U-test using asymptotic inference and p-values < 0.05 were considered statistically significant.

The comparative study (Paper III) includes 66 parents to whom an abnormal finding in their foetus was detected at routine ultrasound in the second trimester. These were compared with 1983 parents with normal ultrasound results from the same one-year cohort. The parents who had answered all the questions in the PEER-U, State of Mind Index, both in Part I and Part II, were included. Also a gender specific analysis was made separately. In this paper the different items in the specific index, State of Mind Index in Part II (after the examination) were analysed. As all the variables analysed were on an ordinal level, non-parametrical tests were applied throughout. For the within group analyses of differences before and after the ultrasound examination the

Wilcoxon's signed rank test was used and for analyses of differences between parents with abnormal and parents with normal ultrasound findings in their foetus the Mann–Whitney U-test was applied. Due to multiple comparisons a Bonferroni correction was applied. Consequently, a p-value < 0.0125 was considered statistically significant (four individual hypotheses were tested) and when analysing the specific items in the State of Mind Index a p-value of < 0.0038 was regarded as significant (thirteen individual hypothesis were tested).

The Statistical Package for the Social Sciences (SPSS) was used for statistical analyses (Papers II and III).

Pre understanding

The author of this thesis does not have a professional pre-understanding relating to performing ultrasound examinations. This could be considered as a limitation but it could also allow for a more neutral attitude. The author's clinical affiliation is as a midwife at a maternal care centre where parents are offered the option of a routine ultrasound examination. Visits to the ultrasound department have occurred during personal pregnancies but also as a midwife for educational purposes. Anna-Karin Dykes, the main supervisor, is also a midwife and has long experience of performing routine ultrasound examinations. She is also experienced in instrument development and qualitative research, especially grounded theory according to Glaser. Elizabeth Crang-Svalenius, the second supervisor, also a midwife and due to her extensive knowledge on the subject and experience of performing ultrasound examinations, she has cooperated in two reports on foetal diagnosis for the Swedish Council for Technology Assessment in Health Care (Hagenfelt et al 1998, Nilsson et al 2006). Anita Lundqvist, the third supervisor, is a paediatric nurse with long experience of children born with an anomaly and she is well experienced methodologically with qualitative research.

ETHICAL CONSIDERATIONS

The work in this thesis was performed according to the four ethical principles; *autonomy non-maleficence beneficence and justice* [57]. The parents could tell their story which was then de-identified in the presentation. Care was taken to listen to them and they were given the possibility to ask questions about the project. Furthermore their narratives can hopefully be helpful in the development of future routines and be beneficial to either themselves, later on, or to other future parents. An advantage of participating in research studies is the opportunity to be heard [58].

All studies (Papers I-IV) included participants with abnormal findings in their foetus at a routine scan in the second or third trimester. Each participant had received written and oral information about the study that explained the purpose and emphasised that they could withdraw from the study or terminate an interview at any time without consequence for their future care. The parents (Papers I and IV) had already agreed to fill out the questionnaires and were later contacted by phone and asked if they were

willing to be interviewed. All the parents who were asked to participate accepted, except one (Paper IV) who could not attend. The parents could choose the time and place for the interviews.

Some of the parents in study IV received part II of the questionnaire in connection with an interview. This was based on an ethical perspective as their foetus was shown to have severe abnormalities. On such an occasion there also was the possibility to ask questions about the study.

In both interview studies (Papers I and IV) the material was coded and treated confidentially. Since the interviews concerned a very sensitive topic this was taken into consideration during the interviews and extra time was reserved, before and after the interview, in order to give the parents all the time necessary for them. Furthermore the parents were interviewed together. In Papers II and III all the data was coded and a code list was kept separately and only available to the research group.

None of the members in the research-team were working at the selected ultrasound department at the time when the studies took place. Ethical approval and permission to undertake the four studies was obtained from the Research Ethics Committee of the Medical Faculty of the Lund University No. LU 453-00.

RESULTS

Paper I conceptualized and gave a deeper understanding of how parents could experience and manage the situation when their foetus was found to have choroid plexus cysts at a routine second trimester ultrasound examination. In Paper II the psychological aspect of this group of parents was investigated and compared with matched controls whose foetus had normal ultrasound findings, with regard to anxiety, state of mind and sense of coherence. The comparison between the groups continued in Paper III and parents to a foetus with abnormal ultrasound findings were compared with parents to a foetus that had normal ultrasound findings. The two instruments of interest were PEER-U State of Mind Index and SOC. Further the State of Mind Index was examined, item by item to clarify if and how the two groups differed regarding the statements about the ultrasound examination. A deeper understanding was obtained in Paper IV about management and parents´ experiences and handling of the situation when their foetuses were found to have a foetal abnormality at a routine second or third trimester ultrasound scan.

Need for knowledge and Pendulating between the abnormal and the normal

Integration of Papers I and IV

The participating parents knew about ultrasound as a tool for identifying prenatal abnormalities even if they saw the examination as a confirmation of the pregnancy.

They also had a positive attitude towards foetal diagnosis. Parents with an abnormal ultrasound examination wanted to know about the finding and wanted information about it and how it should be managed, they had ***a need for knowledge***. The information they needed most was about the diagnosis itself but also information about the importance of further and more extensive investigations and diagnostic methods with different kinds of risks. The pros and cons of the decisions they had to make must be judged in both a short and long term view. Information from specialists such as obstetricians, paediatricians regarding diagnoses, labour and planned care after delivery was also needed and appreciated. Before their meeting with the professionals the parents did not know what to believe and often felt that they had lost control. Those parents with a foetus found to have choroid plexus cysts were given a new appointment and an extra examination, and at the same time they were told that there was nothing to be worried about. The parents were of the opinion that the staff had the required skill with the ultrasound technique and managed the examination in a way that inspired trust. Many experienced extensive anxiety, but only during a relatively short time period. Their feelings fluctuated between *hope and despair* (Figure 1).

The parents already with a diagnosed foetal abnormality (Paper IV) had similar feelings. Mostly these parents were not in a state of uncertainty which is in contrast with the parents in Paper I. The parents in Paper IV received information about the finding and initially they experienced a sense of crushed expectations. Their wish for a perfect child was exchanged for broken expectations. These parents were ***Pendulating between the abnormal and the normal.*** The ultrasound result showed an abnormality but after receiving the information that most children could be treated or operated on without any severe sequel, if any, their experience was that maybe everything could be normal anyway. The parents´ sometimes were of the impression that the staff saw the finding differently and simplified it. Guidance and counselling were important issues for the parents in connection with foetal diagnosis. It was important to have someone to talk to besides just receiving facts about the findings. At this juncture being cared about embraced both the needs of the parents and the foetus as well and was preferable to just being cared for. For the parents there was a kind of strength in knowing that the expected child had an abnormality and to be aware of this was considered by them as beneficial. This made it possible for them to prepare and to understand what and how the diagnosis would affect the child and the family. Those parents who received bad news from the ultrasound examination entered a new path in their lives; they adapted to the new knowledge and gradually regained control over their daily life (Figure 2). The two theoretical code families that were used in these studies were; chain process (Paper I) and the six C´s that was applicable for “cause and effect” (Paper IV).

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Figure 1

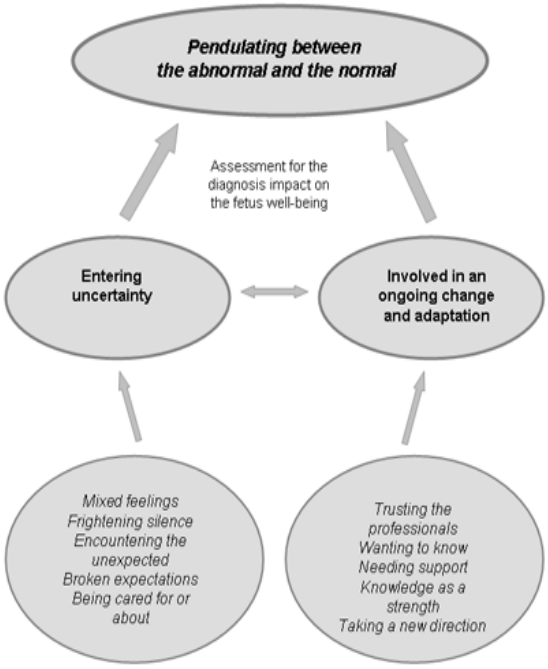

Figure 2

Figure 1. The theoretical model of parents experience and handling of their situation when the soft marker, choroid plexus cyst, is found at a routine ultrasound examination

Figure 2. Generated theoretical model of parents experiences of the ultrasound situation when their foetus was found to have an abnormality.

Parents anxiety when an abnormal finding in the foetus was evident at an ultrasound examination

Paper II

Results from the three instruments used, (PEER-U State of Mind Index, STAI and SOC) were compared between 22 parents to foetuses with choroid plexus cysts (CPC´s) and matched controls, three to one, n=66 parents to foetuses with normal findings. Men and women were analysed together, these details are given in Table 2, Paper II. The STAI-state/trait showed no significant difference between the cases and controls or within the respective group before and after the ultrasound examination. Neither was there any significant difference between the two groups concerning SOC when comparing before and after ultrasound and within their own group. The SOC scores were practically equal between the study group and the control group and they showed a similar pattern when comparing before and after ultrasound within the respective group. The controls had a significant better level of State of Mind Index (less anxious), compared to the cases where CPC´s had been found ($P<0.001$). The control group had also a significantly better level of State of Mind Index (less anxious) when comparing their results before and after as their scores had decreased after the ultrasound examination ($P<0.001$). This did not occur for the cases with CPC´s. There

was a trend towards a higher STAI-state/trait score (more anxious) after the ultrasound examination for the cases with cysts as compared to the control cases however, this was not significant. A gender analysis was not performed due to the lack of power with such small numbers.

Paper III

After the ultrasound examination, the parents with normal findings on their foetus (NF group) showed a significantly improved level of State of Mind Index (less anxious), whereas no significant difference between the two occasions (before and after) was seen in those with abnormal ultrasound findings (AF group) (Table 2 in Paper III). In the NF group, an improved level of State of Mind Index (less anxious) was found for the women after their ultrasound examination (Median 22.92, [IQR] 18.75 (before) vs Median 8.33, [IQR] 10.26 (after); $P<0.001$) but also a significance ($P<0.001$) when comparing the women in both groups, NF and AF, after the ultrasound examination. The women with normal findings were less anxious according to the State of Mind Index. The partners with a normal ultrasound finding also had a higher level of State of Mind Index when comparing their scores before and after the ultrasound scan (Median 16.67, [IQR] 14.58 vs Median 10.26, [IQR] 12.82; $P<0.001$). However, there was no significant difference when comparing the two groups of partners (NF and AF). There was also no significant difference between the NF and AF groups regarding SOC scores, neither before nor after the examination, within each group (Table 2 in Paper III).

The specific items in Part II of the State of Mind Index were analysed. In the thirteen statements made after the examination, there was a significant difference between the two groups, NF and AF, regarding seven of the statements ($P<=0.001$). The statements were also examined from a gender perspective and four out of these thirteen statements showed a significant difference ($P<0.001$) between the two groups, NF and AF, when separating men and women (Table 4 in Paper III).

The parents to a foetus with an abnormal ultrasound finding (AF) felt, to a lesser extent, that they were given information, during the ultrasound scan, about what could be seen, and they also felt that they were informed to a lesser extent during the ultrasound scan; this was also notable when separating men from women. The whole AF group felt, to a lesser extent, a sense of security during the ultrasound scan. This last statement was also significant when separating men from women. The parents to a foetus with an abnormal ultrasound finding (AF) felt to a lesser extent that the attitude of the midwife doing the ultrasound scan was personal and that it was alright to ask questions during the scan. These parents also felt anxious, to a greater extent, despite the fact that they could understand the ultrasound image, even this was significant when separating by gender, however they also felt, to a greater extent, more anxious as they could not understand the ultrasound image but this last statement was not significant when separating men and women (Table 4 in Paper III).

DISCUSSION

Methodological considerations

The reason for the choice to use two different designs – a qualitative design and a quantitative, questionnaire-based, comparative design in this thesis was that they complement each other. The research area is multifaceted. Hence, in order to gain knowledge about the parents' lived experience of ultrasound examinations, a qualitative approach was required. For the sake of achieving results that are general for the whole population a quantitative method was used.

Participants

In Sweden there has been an effort to involve partners during the pregnancy and the care of the child since the 70s. There are a number of studies relating to women's experience of an ultrasound examination with both normal and abnormal findings with regard to anxiety and information however, there are not so many studies where the partners are included. Therefore it was natural to include both parents in all our studies.

At first the participants were included consecutively but selected on the basis of diagnosis i.e. choroid plexus cysts in one group and malformations in the other group. The effort was always to systematically try to expand and include as many different situations and diagnoses as possible until saturation was reached during analysis (Papers I and IV). It was also important to have as broad a variation regarding primiparaes and multiparaes, marital status and education level as possible. Unfortunately, this was not so easy to achieve. Many of the participants have a high education level, above the average for Sweden and only one among those interviewed was single and this could be considered as a limitation. Another limitation was that only Swedish-speaking parents were interviewed.

All those who were asked to participate in the interview studies agreed to do so (Papers I and IV). This was very positive, but also surprising, since the interviews were about a sensitive topic. All the participating parents had both positive and negative experiences in their narratives which can suggest that they were not people with extreme experiences who wanted to participate in the study. All the couples were interviewed together (Papers I and IV) The advantages in interviewing them together was the time saving for the parents and that they could also help one another in recalling educational courses and specific things that happened around the event which otherwise might have been excluded due to having been forgotten. It cannot be excluded that the interviews would have been different if the parents had been interviewed separately however, the interviews had not necessarily been more comprehensive.

Trustworthiness

Lincoln and Gubas [59] use the concept trustworthiness for studies with qualitative design which include credibility, transferability, dependability and confirmability. *Credibility* refers to the truthfulness of the data [60]. It also comprises the importance of how the data collection has been done, the interview situation and that the data are covered in the categories that emerge and that any non relevant data is sorted out. By letting parents choose the time and interview location increased the prospects that they would experience the interview as pleasant. The interpretation of the material was made visible for the reader through the use of quotations from the transcribed text mass to verify the selected categories. With regard to the above, the results (Papers I and IV) could be viewed as credible. The *transferability* is a question of whether the results can be relevant in any other social environments [59]. Regarding the study of choroid plexus cysts (Paper I), the result could apply to other soft markers as well, not only CPC´s. Parents who are informed about an abnormality after another type of foetal diagnosis, not only routine ultrasound examinations, could perhaps initially experience similar reactions as the parents did in Paper IV when they received information concerning the abnormal ultrasound findings. Based on this, the result could perhaps be considered as transferable, at least in paper I. The *confirmability* refers to objectivity in a quantitative study [59] and is difficult but in an effort to achieve this two of the interviews in each study (Papers I and IV) were performed by two different interviewers. It gave the possibility to establish that the first author, who conducted most interviews, did not, by her attitude, influence the interviews and the outcome. Some of the authors analysed a number of interviews separately; the analysis and interpretations were then compared and discussed. The question of pre-understanding was taken into consideration. Within the team there was expertise in ultrasound examinations with abnormal findings. Although the main purpose was to examine parents' experience of the event it was an advantage to have access to this knowledge Thus, the result could be viewed as both objective and truthful. The *dependability* refers to the reliability [59] and together with transferability probably the most difficult parts in terms of qualitative research as it often deals with various social phenomena. The difficulty about repeating a qualitative study is that social environments can not be “frozen” over time.

These concepts are not used in Grounded Theory. Instead it is very much up to the reader to decide if the theory has validity since the cornerstones regarding validity in GT is about how well it fits, its relevance, how well it works and if it is modifiable [61]. Both in Papers I and IV the authors discussed and agreed upon the *fit* of the categories. The clinical *relevance* was also an important issue and two of the authors have extensive experience of performing ultrasound examinations and of parents’ reactions when an abnormal ultrasound finding is discovered and their experiences matched well with the results (Papers I and IV). The *workability* is shown in the implications (Papers I and IV) and the *modifiability* i.e. the possibility to alter the theory when new relevant data is included is part of an ongoing process in our research.

Sample

In paper II matched controls were used. The cases (n=22) were matched against each three controls (n=66). The matching succeeded almost totally with only a few minor exceptions, such as the year of birth and country of origin, as an American born woman was matched against a woman born in Sweden. In study III all parents who had a deviation were included (n=66) in the study no matter how slight or severe the deviation was. These parents were compared to the parents with normal findings (n=1983).

Validity

Internal validity refers to the extent to which the ultrasound examination can account for the results rather than any other factors [62]. Examples of such additional factors which could influence the result is selection bias i.e. group differences and that more women than men participated (Paper II and III). In an effort to strengthen the internal validity the participants in Paper II was matched by sex, age, marital status, education, country of origin and parity. Another threat to internal validity is history. During the time between the two questionnaires the parents may have been affected by events not related to the ultrasound examination. This is of particular concern regarding the generic instruments STAI and SOC. The PEER-U, State of Mind Index seems to be more time independent since it is an ultrasound specific instrument.

External validity refers to whether the results can be generalized to persons, settings, time, measures and characteristics [62]. In Sweden there is a recommendation, this is a programme for pregnant women including ultrasound examinations. In settings similar to the one in these studies the results will be valid and useful. However, routines can diverge but also possibilities to offer multiple ultrasound examinations during various trimesters and using different technical equipment. This could affect the external validity of studies II and III. Furthermore with regard to the external validity, it could be considered as a limitation that the questionnaires were only available in Swedish and maybe parents with Swedish language difficulties were involuntarily excluded for this reason. An external drop-out analysis of the cohort [46] showed that the most common reasons for partners not to participate were that they had not received the questionnaire. It was addressed to the woman and she had not handed it over to her partner or. If they had received it, the reason for not completing it was oblivion, lack of time or not having been present at the ultrasound examination or disapproval of questionnaires. The most common reason for the women not answering the questionnaire was that they had forgotten to fill it out or they returned the questionnaire uncompleted or that it was just too extensive. The less successful recruitment of men needs to be addressed for future studies.

Statistical validity

This refers to whether the statistical methods used are able to show causations or differences between groups and time points [62]. In these studies non-parametric methods were used for analysis of continuous variables. Non-parametric methods are

most appropriate when the variables are on an ordinal level and sample sizes are small. In this thesis non-parametric tests were used as the variables either were not normally distributed or measured on an ordinal level [63, 64]. For comparisons between groups the Mann-Whitney U-test was chosen. For the comparison of variables measured on two occasions in the same sample the Wilcoxon signed-ranks test was used. In study II no gender analyses were performed due to the lack of power. In study III no comparisons were made within the AF group between groups with different deviation severity or with particular anomalies as the material was considered too small to allow for subgroup analyses. Such sub-analyses would result in a series of case reports with limited scientific value.

General discussion of the results

Generally the parents in Papers I and IV expressed how they had wished for their first routine ultrasound examination. During pregnancy there is often a mix of emotions, joy and expectation but also uncertainty and fear [65]. Parents also recognised that ultrasound is a tool for detecting foetal abnormalities. Baillie et al [66] found the same situation among their respondents in 2000 as well as Williams et al [67] in 2005 and Lalor et al [9] in 2007. Therefore it is easy to imagine that prenatal diagnosis can cause anxiety independent of the method used.

The information about an abnormal ultrasound finding comes unexpected to most parents [68, 44] and creates anxiety. This was also shown in our studies and initially it involved shattered expectations as the parents expected the ultrasound examination to show a normal result. Usually, during the ultrasound examination, the parents could sense that something was wrong even before they received confirmation of a problem. This fact was found in both studies I and IV. The parents anxiety was intensified by the immediate environmental factors such as the dark room, the droning sound from the ultrasound machine and of course the silence. How sensitive parents are to the surrounding environment is also noted by Lalor et al [9] and Van der Zalm [44]. The parents in Papers I and IV noticed and assumed that the midwife who performed the ultrasound examination was limited regarding what she was permitted to discuss. The analyses of the items in the State of Mind Index (Paper III) showed that the parents with an abnormal finding in their foetus felt to a lesser extent that they were given information during the examination about what could be seen and they also felt to a lesser extent well informed during the scan, also noted when separating men and women. This significant difference between the two groups, NF and AF could perhaps be explained by the limitation in what the examiner is permitted to discuss with the parents due to how the routines are organised.

It is not obvious for parents to assess the severity of an abnormality and even if it is operable, the situation will cause them anxiety [40]. This could be the reason why some parents in Paper IV experienced that the staff gave the impression that they considered that the parents had overreacted concerning the abnormality. The parents felt confused and were fluctuating between hope and despair, wondering whether their child would

be normal or abnormal. Their child was in one way unhealthy but on the other hand not so very unhealthy, so perhaps their child was healthy after all. Their concern about the foetus well-being was constantly on their minds. The parents wanted to receive information about the finding and they felt that they could also handle the situation, but that is not the same as saying that they were not anxious. Their anxiousness was shown in both studies II and III with reference to the State of Mind Index. However, correct information about the finding and a meeting with the responsible specialist together with information about further assessment, counselling and management decreased the parent's anxiety and improved their situation. Etchegary et al [42] found that one part of the women's decision in accepting the ultrasound screening was the possibility to be prepared if they were carrying a child with a disability. This part, about preparing, was also mentioned in study IV. It seems to be important for the parents to have someone to talk to, not only about the findings but perhaps even the sense of guilt they can feel that is common in these situations. It is not unusual for women to speculate about why their child has an abnormality and question if it could be a consequence of her own behaviour [65, 69]. It is not necessarily psychological counselling that is needed, perhaps it is sufficient with just a person who represents the health care services and who is prepared to listen and who can be a contact person if other professionals are needed. Parents should at least not have to cope with bad news in the absence of planned support.

It is likely that the parents´ ultrasound experience may affect an eventual following pregnancy and ultrasound examination. It is important that the staff working with these matters can recognise when complex skills are needed, because it is not only a matter of knowledge it is also a matter of awareness of values and ethics. Health-care professionals, who perform ultrasound examinations, should be trained in how to inform and what to explain to expectant parents when abnormal ultrasound findings occur. Furthermore, the parents are in a vulnerable situation and should not receive information concerning an abnormal ultrasound finding while the woman is lying down, half-naked, in a dark room. In such a situation the parents are most likely to feel that they are at a disadvantage. An interview study pointed out that parents are sensitive towards how bad news is given [9].

Also worth remembering is that there may be a need for both general and individual care planning. The instrument PEER-U, State of Mind Index [44] could be a helpful tool for improvement of such routines. In paper III, when analysing the index, many of the items that differed between the two groups AF and NF could be connected to the routines. Even if our results to some extent confirm previous knowledge of how women experience the routine ultrasound examination when an abnormal finding is discovered in their foetus, our studies have added an important dimension by analysing the partners´ (fathers') experiences as well. In Paper III a gender analysis with reference to State of Mind Index was carried out and the women with a normal ultrasound finding had significantly better scores (less anxious) after the examination and they had also significantly better scores after the examination when compared to the women with an abnormal finding. Almost the same pattern was found for the men except when comparing the two groups NF and AF after the examination, where no

significant difference was found. The result is interesting from a gender perspective. Lundqvist and Jakobsson [70] and in Lundqvist et al [71] found that when they examined men's´ experience of becoming fathers to preterm infants, the fathers' initial concern was about the woman's condition and well-being. The equivalent may already exist during pregnancy. The man is focused on the woman, while the woman is focused on the foetus. This could explain why there was no difference between the groups NF and AF when comparing the men in our study III. On the other hand, Ekelin et al [46] noted that fathers' anxiety did not decrease as much as the mothers' anxiety did after a normal ultrasound examination. The reason given for this was that since the examination the father was more clearly aware that he would soon be a parent. Therefore, his anxiety did not decrease in the same way as for the woman. Now the father had an image of the child which perhaps gave rise to feelings that did not exist before the examination. The impact of the ultrasound image regarding the mothers´ bonding to the child and pregnancy was noted in an article by Dykes & Stjernkvist [5] from 2001.

Prenatal diagnosis is a very complex and sensitive topic. It includes much ethical dilemma and much of what foetal diagnosis is about is opposites. It can be beneficial or do harm, create or destroy, be useful in its ability to cure or to kill.

An integration of the categories from studies I and IV give an argument which theoretically could be able to explain how parents, with any form of deviation in their foetus from the normal, experience their situation. The unexpected information about the abnormal ultrasound finding creates a feeling of uncertainty and the ***need for knowledge*** increases as the parents´ fluctuate between *hope and despair* while at the same time ***pendulating between the abnormal and the normal*** is ongoing. While the information sinks in, the parents are still *entering uncertainty* but they are also creating strategies. The *frightening and confusing* information about the findings occupies their minds – they ask what is wrong and different from usual. Information is sought but also given. Choices are to be considered and the parents have to *judge the risk and make a choice* that is beneficial for all those involved, the family and the foetus. Finally the parents find some sort of consolation which means that they are capable of going on with their lives the feeling of comfort arises. *They are involved in an ongoing change and adaptation* but the difference from earlier is that the situation is now manageable.

CONCLUSIONS AND CLINICAL IMPLICATIONS

The fact that ultrasound is a tool for detecting foetal abnormalities is known among the parents of today, even though their reasons for having the examination is primarily to have confirmation that their child is healthy and an opportunity to meet the new member of their family. Parents to a foetus with an abnormal ultrasound finding are more anxious after the ultrasound examination than parents to a foetus with a normal ultrasound finding. However, they still have a positive attitude towards ultrasound examinations but they do not think of it as foetal diagnosis. Most parents want to know

about the finding and can handle the information they receive. They are also in need of sufficient information regarding a detected deviation or abnormality in connection with the ultrasound examination and a strategy for further management. For example, the soft marker should be given the correct term and spelling independent of the examiner. A meeting with a physician or a specialist in the field of the deviation is also desirable. As part of a routine in a situation like this all parents should be offered a professional contact person to talk to, for example a midwife, physician or psychological counselling if required. Initially it seems as it is the finding of a deviation or an abnormality in the foetus itself that causes anxiety, not how severe the consequences will be. The parents do not compare the finding in terms of a more or less severe deviation, just the fact that it was not a normal ultrasound examination. The new instrument, PEER-U State of Mind Index, does not only measure the parents state of mind (anxiety) in connection with an ultrasound examination, it can also give some information as to what could have caused this anxiety and further the instrument does not appear to be time dependent. The results also show that many of the statements in the index could be connected to the routines at ultrasound departments. This might be useful in quality development concerning how to care for the parents. Anxiety can never be totally excluded in a situation like this but it could be reduced with the use of correct information before and after the examination while the parents are properly seated. It is also important that the family is cared about which embraces both the foetus and each parent. Further the ultrasound community ought to think carefully before introducing actions based on new markers. Also it is of outmost importance that the sonographers, irrespective of profession, are trained in what to explain to the expectant parents when such markers are seen. Finally the examiner should be aware of how anxiety can be intensified by environmental factors.

FUTURE PERSPECTIVES

Today's technology has developed and many parents appreciate scanning that incorporates 3-D or 4-D technology. The assumption that ultrasound examinations are risk-free is of course encouraged by the fact that scanning is considered safe enough to be offered as a routine examination to every pregnant woman in Sweden, as in other countries [67]. On the other hand this new technology has not yet been systematically evaluated, and therefore it is not self-evident to say that it is a totally benign procedure [8]. This could be important to remember since parents are more likely to accept non-invasive diagnostic methods with the respect to their foetus. In the future probably more advanced screening programmes and methods will be offered. It also seems as there is a need for social ultrasound scanning, besides the strict medical one. This could possibly lead to more ultrasound examinations being performed per woman and thereby each foetus would be scanned more often. Another risk with this new trend is that it could give a false sense of security. In Sweden today, the obstetric units try to incorporate the social part of the ultrasound examination into their medical examinations such as physicians do elsewhere [8.] The limitation as to what can be discovered by an ultrasound scan is an important issue to discuss with parents. A recently Greek study showed that 86.9% of the participating women (n=300) believed

that foetal karotype could be determined by an ultrasound examination [72]. This in turn could lead to parents accepting to have an ultrasound examination based on the wrong grounds.

It might be of interest to investigate statistically if the severity of the deviation affects how the parents estimate their well-being with respect to STAI, SOC and PEER-U State of Mind Index. The interview studies (Papers I and IV) indicated that initially it is the abnormal finding in the foetus itself that causes anxiety but this is likely to change during the pregnancy period. Therefore, it would be of interest to make two measurements, one shortly after the examination and one around pregnancy week 30, or after the routine ultrasound examination in the third trimester.

It might also be of interest to interview parents with regard to abortion in the context of malformations or chromosome abnormalities and how they make their decision regarding abortion or keeping the child and if the medical staff has any impact on their decision and if so in what way.

SUMMARY IN SWEDISH

Svensk sammanfattning

Ultraljudsteknik började användas inom området obstetrik och gynekologi redan i slutet av 1950-talet, och både tekniken och tillämpningen har utvecklats enormt under dessa sextio år. I Sverige är ultraljudsundersökning under graviditet inkluderad i det rekommenderade vårdprogram som finns för gravida kvinnor [17], och få föräldrar kan i dag tänka sig en graviditet utan dessa undersökningar. Fosterdiagnostik handlar inte bara om barnets välmående utan också om etik. En omdebatterad fråga är huruvida fosterdiagnostik kan betraktas som en form av diskriminering och gallringsmetod, mot bakgrund av att undersökningen kan leda till selektiv abort. Dock är det sällan i diskussionerna som själva ultraljudsundersökningen ses som fosterdiagnostik, vilket den är på många obstetriska enheter.

Ultraljudsundersökningen anses vara riskfri för barnet [6, 7] men de i dagsläget längre undersökningarna med tredimensionell (3D) och fyrdimensionell (4D) teknik har ännu inte blivit systematiskt utvärderade med avseende på risker [8]. Kanske är det med hänsyn till rådande kunskap lättare att som förälder ha en mycket positiv inställning till just icke-invasiv fosterdiagnostik än t.ex. invasiva metoder [65]. Nya och förmodligen också mindre riskfyllda metoder, för både mor och barn, kommer att erbjudas föräldrar i framtiden. Frågan är om det då kommer att bli ännu lättare för föräldrar att acceptera ett sådant erbjudande och i de sammanhangen tänka mindre på vad undersökningen kan komma att visa, dvs. resultatet av undersökningen och hur det ska hanteras. Innan nya metoder och screening program erbjuds föräldrarna, måste också hänsyn tas till psykosociala aspekter på fosterdiagnostik [65], något som även påtalas i konsensus-uttalandet från Vetenskapsrådet [13].

Avhandlingens övergripande syfte är att beskriva föräldrars erfarenheter, reaktioner och behov i anslutning till rutinultraljudsundersökningar där fostret visar sig ha ett avvikande ultraljudsfynd. Avhandlingen består av fyra delstudier som alla omfattar båda föräldrarna. Samtliga delstudier har en prospektiv design, och i delstudie II och III har också en jämförande design använts. För att nå en djupare förståelse för föräldrarna och deras välbefinnande i den aktuella situationen, har både kvalitativa och kvantitativa forskningsmetoder använts.

Syftet i delstudie I och IV var att få en teoretisk förståelse för föräldrarnas erfarenhet och handlande i situationen när fostret under rutinultraljudsundersökning befunnits ha plexus choroideus cystor (delstudie I) eller en fetal missbildning (delstudie IV). Metoden för datainsamling och analys var Grounded Theory [53]. Resultatet för delstudie I gav kärnkategorin ”Behov av kunskap” med underkategorierna *skrämmande och oroande; bedöma risken och fatta beslut; lugnad.* Kategorierna speglar den process som föräldrarna gick igenom, en period med mycket oro men ett relativ snabbt händelseförlopp. I delstudie IV utvecklades kärnkategorin ”Pendlande mellan det onormala och det normala”, med underkategorierna *inträdet i osäkerheten* samt *involverad i en ständigt pågående förändring och anpassning*. En orsak-och-

verkan-process var det som följde föräldrarna från att de hade fått beskedet om missbildningen fram till dess att de kunde ta den till sig och bearbeta de nya tankar och handlingar som det innebar för familjen.

Slutsatsen för studierna är att föräldrar vill ha information om ultraljudsfyndet. Det som efterfrågas är en korrekt information i anslutning till undersökningen där också förslag till vidare behandling och undersökningar diskuteras. Föräldrarna önskade också någon att tala med om sin upplevelse. Därmed är det inte så att det i samtliga fall är en psykolog som ska vara denna part, det skulle också kunna vara en barnmorska eller en läkare. Initialt vid beskedet om ett avvikande ultraljudsfynd blir föräldrar mycket oroliga oavsett hur svår avvikelsen är. Inledningsvis är det troligen inte de konsekvenser avvikelsen för med sig som utgör fokus för deras funderingar, inte heller att någon annan kan ha fått en mer allvarlig diagnos, utan det är den normala ultraljudsundersökningen som de relaterar till.

Syftet i delstudie II och III var att undersöka föräldrarnas psykologiska välbefinnande då fostret under rutinultraljudsundersökning visat sig ha plexus choroideus cystor (delstudie II) eller en fetal missbildning (delstudie III). Följande instrument användes: State Trait Anxiety Inventory (STAI) [48], Känsla av sammanhang (KASAM) [50] och sinnesstämning enligt det specifika indexet i frågeformuläret Parents' Expectations, Experiences and Reactions to routine Ultrasound examination during pregnancy (PEER-U, State of Mind) [45]. Båda delstudierna är jämförande enkätstudier där föräldrarna fick självskatta sitt psykologiska välbefinnande.

I delstudie II jämfördes föräldrar till foster med plexus choroideus-cystor (n=22) med matchade kontroller med normala ultraljudsfynd (n=66) där respondenterna hade svarat på alla frågor som handlade om psykologiskt välmående. I delstudie III jämfördes de normala ultraljudsfynden från den stora kohorten (n=1983) med de i kohorten som hade avvikande fynd (n=66) och som hade svarat på alla frågorna i PEER-U, State of Mind Index, både före och efter ultraljudsundersökningen.

I delstudie III användes inte STAI och varje fråga i indexet State of Mind analyserades för att få en uppfattning kring vad som kan skapa oro i ultraljudssituationen då ett avvikande fynd görs. Resultatet visade att föräldrar med avvikande ultraljudsfynd (delstudie II och III) är mer oroliga än föräldrar med normala fynd, enligt State of Mind Index. De föräldrar som fått besked om normala ultraljudsfynd kände signifikant mindre oro efter undersökningen jämfört med före undersökningen (delstudie II och III). STAI visade ingen signifikant skillnad i oro mellan grupperna (delstudie II), vilket skulle kunna bero på att instrumentet är generellt och inte anpassat speciellt till en ultraljudsundersökning. Dessutom är det troligen mer tidsberoende än PEER-U State of Mind Index som är ett ultraljudsspecifikt instrument. Föräldrarna fyllde inte i enkäten direkt efter undersökningen, utan det kunde dröja ett par veckor. Därmed kan det inte uteslutas att skattningen med STAI kunde spegla någon annan upplevelse. KASAM-värdet var stabilt över tid och påvisade ingen skillnad mellan grupperna normala/onormala fynd (delstudie II och III). Delstudie III visade också att föräldrarna hade god förmåga att hantera svåra situationer enligt KASAM. Analysen av frågorna i

indexet State of Mind indikerade att frågorna som signifikant skilde grupperna normala/onormala åt kunde kopplas till rutiner kring ultraljudsundersökningen.

ACKNOWLEDGEMENTS

This thesis for the degree of Doctorate was carried out at the Department of Health Sciences, Division of Nursing, Lund University. I wish to express my sincere gratitude to:

- All the parents who kindly participated in this thesis, especially those who were interviewed (Papers I and IV) and willing to tell and share their experiences.
- Associate Professor Anna-Karin Dykes, my main supervisor– thank you for believing in me by giving me this chance and for a great education and in between, lots of fun, but also for your critical and fantastic supervision - could not have made this without you.
- Associate Professor Elizabeth Crang Svalenius and Senior Lecturer Anita Lundqvist, my assisting supervisors for their invaluable support and encouragement. It has been good and reassuring to have had you on this journey and fun as well.
- My co-authors Maria Ekelin, Professor Karel Maršál and Senior Lecture Per Nyberg for all their help and vast knowledge which has been extremely favourable to my thesis. Maria, my dear colleague and friend for sharing both setbacks and successes and for always being supportive and of course for lots of fun.
- The staff at the ultrasound department for your invaluable help.
- The members of the research group Health and Care for Women and Children. Thank you for your support and helpful criticism during the seminars.
- Magnus Hovde and Karin Samuelsson, my first room mates at the university, for good support and lots of fun. Magnus, I would also like to thank you for helping me with different computer problems or assignments - could not have done it without you.
- Geoff Dykes for being there for me when needed and for revising the English in this thesis.
- My dear friend Eva Hansson for always supporting me and who was the first to give good advice when I entered this new path in life. Also my dear friend since nursing school, Marie Lindell who has followed my work over the years and has always been there for me.
- Colleagues at USiL, especially MVC-Arlöv – Gunnela, Lotta and Cecilia for your interest in my work and Bodil Knutsson, my chief at USiL for your encouragement.
- Those friends who have been with me for a long time, especially Elisabeth and Eddie and her family who have always supported me.

- My mother Ingrid and my dear sister Marie and her family, Anders, Christian and Tove, for always believing in me.
- Finally my dear husband Micke for supporting and helping me with various technical problems particularly when my patience ran out. Our much loved children Kim, Nikolai and Lotta for your patience towards me, especially near the end of the work with the thesis. I am very proud of you all.
- The study was supported by grants from The Swedish Medical Research Council and the Faculty of Medicine, Lund University, Sweden.

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