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**Daily life experiences,
symptoms and well being in
women with coeliac disease
A patient education intervention**

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"Where we stand is not as important as the direction in which we are moving."

Oliver Wendell Holmes, Jr
(1841 – 1935)

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ABSTRACT

Daily life experiences, symptoms and well being in women with coeliac disease A patient education intervention

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Background and aims: Despite living with a gluten-free diet (GFD) Swedish women with coeliac disease (CD) report a lower level of well-being than women without the disease and than men with the disease. The aims of this thesis were to describe the life experience of being a woman living with CD (I) and to assess the effects of patient education (PE) with problem based learning (PBL) on psychological well-being (II) and gastrointestinal symptoms (III).

Subjects and methods: In total 106 women, ≥ 20 years, with confirmed CD, who had been treated with a GFD for a minimum of five years were randomized (II&III) to one of two groups: either to the intervention group (n=54) that underwent a ten-session educational program, called 'Coeliac School', with PBL, or to a control group (n=52) that received information regarding CD, which was sent to their home on a regular basis.

Assessments: Well-being (II & III) (the Psychological General Well-Being index and the Gastrointestinal Symptom Rating Scale) was assessed at baseline and after 10 weeks in all patients and after 6 months in the intervention group. Individual interviews (I) were carried out with purposefully selected women (n=15) before the start of the 'Coeliac School'.

Result: The qualitative study showed that CD can influence women's lives in different ways. A desire for a normalised life-world was described. Three conditions necessary to achieve a normalised life-world were described as: being secure, being in control and being seen and included. After ten weeks of education, participants in the "Coeliac school" reported a significant improvement in psychological well-being ($p=0.001$) and gastrointestinal symptoms ($p=0.013$). The controls did not improve significantly. However, at baseline the controls, for reasons which were not apparent, and despite randomization, expressed significantly better psychological well-being and fewer gastrointestinal symptoms than women in the intervention group. Six months after completion of the PE program some of its positive effect had decreased.

Conclusions: A PE with PBL can help women with CD to benefit from a greater degree of well-being. It is important to offer PE to those women who fail to achieve a normalised life-world. However, intervention methods need to be refined in order to provide a more pronounced long-term effect.

Key words: Chronic disease, illness experience, phenomenology, well-being, Patient education, PBL

LIST OF PAPERS

This licentiate thesis is based on the following papers, which will be referred to in the text by Roman numerals.

- I. **Coeliac disease- Women's experiences in everyday life.**
Ring Jacobsson L, Hallert C, Milberg A, Friedrichsen M.
Submitted 2011

- II. **Does a Coeliac School increase psychological well-being in women suffering from coeliac disease, living on a gluten-free diet?**
Ring Jacobsson L, Friedrichsen M, Göransson A, Hallert C.
Accepted for publication in Journal of Clinical Nursing (2011)

- III. **Impact of an Active Patient Education programme on Gastrointestinal Symptoms in Women with Celiac Disease Following a Gluten-free Diet: Randomized Controlled Trial.**
Ring Jacobsson L, Friedrichsen M, Göransson A, Hallert C.
Accepted for publication in Gastroenterology Nursing (2011)

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ABBREVIATIONS

CD	Coeliac disease
GFD	Gluten-free diet
GI	Gastrointestinal
GSRS	Gastrointestinal Symptom Rating Scale
PBL	Problem Based Learning
PE	Patient Education
PGWB	Psychological General Well-Being Index
QoL	Quality of life

INTRODUCTION

Coeliac disease (CD), with a prevalence of almost 1%, is the most common food intolerance in the general Western population (Dubé et al. 2005). CD is a chronic inflammation, with damage in the small intestine due to gluten presented in wheat, barley and rye. The disease leads to malabsorption of different nutrients and, through that, a wide spectrum of symptoms, of which the most common in adults are abdominal pain, chronic diarrhoea and iron-deficiency anaemia (Abdulkarim and Murray. 2003). People suffering from CD who keep strictly to a gluten free diet (GFD) will, in general, experience a significant symptomatic improvement in a short period of time (Garcia-Manzanares and Lucendo. 2011). However, those with treated CD, particularly women, frequently experience a negative impact to their well-being (Midhagen and Hallert. 2003), which is suggested to be closely associated with the perceived disease burden (Hallert et al. 2002). Women suffering from CD consistently report more concerns related to the disorder and its management and have also stated a need to gain more knowledge (Hallert et al. 2002; Hallert et al. 2003). Patient education (PE) is widely recognized as a core component of nursing (Booker et al. 2008), and a positive relationship between received knowledge and health-related quality of life has been found (Leino-Kilpi et al. 2005). With these perspectives in mind, I found it interesting to gain a deeper understanding of how women with CD in Sweden experience their daily life, and to investigate whether PE with problem based learning (PBL) is an effective way to satisfy their knowledge needs in order to support and encourage possible facilitating changes in their lifestyles, and through that experience increased well-being.

BACKGROUND

Living with a chronic disease – gender aspects

In order to understand the context for the women participating in the studies in this thesis it is important to identify some of the central concepts. The first concept is “chronic condition”, which is defined as disability or disease conditions which people live with for a long time (i.e. more than six months). “Chronic disease”, on the other hand, is a concept included as a subset of chronic conditions and refers to a specific medical diagnosis (Lawn and Schoo. 2010). Dramatic increases in people suffering from chronic conditions in the world represent the healthcare challenge of this century (World Health Organization. 2002), and many people, after diagnosis and any necessary treatment, are often told that they should be able to live a normal life if they follow medical advice (Gregory. 2005).

The concept of “gender” involves more than the purely biological differences that exist between men and women. According to WHO’s definition, women and men differ in terms of biological make-up, but also regarding power, status, norms and roles in society (World Health Organization. 2005). As stated by Smirthwaite (2007), previous studies have observed that although women live longer than men, they are more ill, report higher rates of morbidity and disability and also use more health care facilities than men. These findings are often used as a description of the prevailing gender disparities in health in developed countries and she means that possible reasons for the greater proportion of sick leave by women may include the fact that knowledge about women’s health and diseases is inadequate (Smirthwaite and Sveriges kommuner och landsting. 2007). Symptoms are important signals for individuals, and when suffering from a chronic condition the monitoring of symptoms becomes a routine of everyday life. There are studies showing that women with chronic conditions report more symptoms and a lower level of mental health than men (O’Neill and Morrow. 2001), whereas others find no evidence that women are more likely to report conditions of a “trivial” or mental nature (Macintyre et al. 1999). One part of the explanation of gender differences with regard to symptoms may consist of

the fact that women are more likely than men to carry out their daily responsibilities despite illness, and thus they become dependent on being attentive to bodily signals (O'Neill and Morrow. 2001). Symptoms of ambiguous chronic illness in women are often feminized, trivialized and also rejected by health care providers as being psychosomatic in nature (Davis and Magilvy. 2000).

CD is a chronic disease in which scientists (Hallert et al. 1998; Hallert et al. 2002; Hallert et al. 2003; Midhagen and Hallert. 2003; Zarkadas et al. 2006) repeatedly claim to have observed gender differences regarding symptoms and perceived disease burden. Johnson & Johnson (2006) take the view that women in particular are afflicted by ambiguous chronic illnesses, wherein they also include CD. They believe this results in women with this type of disease having to learn how to manage within the context of a health care system and a social culture that are sometimes hostile to the legitimacy of their illness experiences (Johnson and Johnson. 2006).

Coeliac disease

CD, also called coeliac sprue and gluten-sensitive enteropathy, is a multifactor autoimmune disorder, in genetically susceptible persons, located in the small intestine and influenced by environmental factors as well as genetic factors (Hamosh. 2011). The disease is characterized by malabsorption resulting from inflammatory damage of the mucosa of the small intestine due to ingestion of wheat gluten or related rye and barley proteins (Farrell and Kelly. 2002; Hamosh. 2011).

CD is one of the most common immune-related diseases (Hrdlickova et al. 2011), but it is difficult to estimate its true prevalence because many carriers have atypical symptoms or no symptoms at all (Farrell and Kelly. 2002). The incidence of CD in the general Western population is estimated at close to 1% (Dubé et al. 2005). Surprisingly, the prevalence of CD in a Saharawi population living in Algeria was more than five times higher (5.6 %) than in any European population (Hamosh. 2011). People with a purely African-Caribbean, Chinese or Japanese background are rarely affected by CD (Farrell and Kelly. 2002) and in most populations there is a slight female predominance (Farrell and Kelly.

2002; Woodward. 2011). The disease has a strong heredity component, and was long regarded as a gastrointestinal (GI) disorder of childhood, but today it is more frequently diagnosed in adults than in children. Today CD is considered to be a chronic systemic disease (Hamosh. 2011) that meets the criteria of a true autoimmune disease. The genetic predisposition (HLA), exogenous trigger (gluten) and auto antigen (tTG) are known (Fasano and Catassi. 2001).

CD has a wide spectrum of gastrointestinal and extra- intestinal manifestations. In adults iron-deficiency anaemia and diarrhoea are common features. Less common features can be divided into: general features, such as short stature; gastrointestinal features, such as recurrent abdominal pain and steatorrhea; and extra-intestinal features such as nutritional deficiencies, osteoporosis, malignancy, alopecia, anxiety & depression, dermatitis herpetiformis, neurological symptoms etc. (Farrell and Kelly. 2002). There is also an asymptomatic (silent) form of CD, usually detected by screening. A detailed history of these people demonstrates a low-intensity illness often associated with decreased psychological well-being (Fasano and Catassi. 2001). Another autoimmune condition, such as thyroid disease or diabetes type 1, is present in up to 30% of patients with CD, compared to 3% in the general population (Jacobson et al. 1997).

The diagnostic criteria for CD are currently under revision, also with an eye on approaches that imply non-invasive diagnostic strategies (Lindfors et al. 2011). For the time being, positive antibodies or a strong suspicion of CD in a person will give rise to a mandatory small bowel biopsy, which confirms the diagnosis histologically, and then the patient starts the treatment with a GFD (Hopper et al. 2007). The damage to the intestine and associated symptoms are reversed, and both adults with symptom- and screen-detected CD experience an improved state of health and quality of life when treated with a GFD (Ukkola et al. 2011). An abundance of food contains gluten, but it must be clearly labelled by law (Woodward. 2011). New pharmacological treatment based on current knowledge regarding the disease pathogenesis is under scrutiny (Lionetti and Catassi. 2011). The sensitivity to gluten varies, but regardless of that it is important to exclude all dietary gluten (Woodward. 2011). In the current situation, we thus know exactly what the treatment is that heals the damage in the intestine when CD occurs, yet we know very little about what it is really like to be a woman living with this disease. CD and its

impact on this group of patients are generally poorly explored from a nursing perspective and it is therefore considered an interesting area for this thesis.

Subjective well-being

Subjective well-being in people suffering from CD has been identified as a great problem in many studies. Subjective assessment offers perspectives beyond the traditional biomedical markers and shall be a complement to objective evidence of disease (Dimenas et al. 1996). This attitude can be considered as an acceptance of trust in patients' own ability to report symptoms in a reliable and, for healthcare, useful way.

All symptomatic conditions affect a person's well-being significantly, including the potential to function in daily life (Dimenas et al. 1995). The concept "well-being" can be described in terms of being well, healthy, contented etc. and it denotes a state where the individual's basic needs are satisfied. Well-being also refers to the person's state of health, in the sense of including not only the absence of disease, but also the quality of health which the person experiences.

The meaning of the concept well-being also varies depending on where in the world we find ourselves, and can then also be explained by factors such as income, religion etc (Diener,E. Kahneman,D. Helliwell,J. 2010); i.e. the types of factor that we in Sweden might very well connect with the concept of life-satisfaction. In this thesis the concept of well-being refers to the person's state of health, in the sense of including the quality of health as experienced by them. A person's Quality of Health is closely related to the concept of Quality of Life (QoL), which some believe can be measured objectively, while others believe it can only be measured by the individual living that life (Kane. 2002).

An example demonstrating that people with CD experience lower well-being than healthy individuals is the fact that, despite remission with GFD, people with treated CD, particularly women, experience a lower level of QoL than non-coeliac controls of the same age (Hallert et al. 1998; Hallert et al. 2003; Zarkadas et al. 2006). Other factors include the reality that women with CD also report more distress than healthy individuals, which is caused by restrictions to their daily life (Hallert et al. 2003), and the perception, for reasons which are not apparent, that the burden of their condition is worse

than for men (Hallert et al. 2002). Some reports from Italy indicate an increased frequency of depression among adults living on a GFD (Addolorato et al. 2001; Ciacci et al. 1998; Fera et al. 2003), and a new Swedish study even shows that individuals diagnosed with CD have a higher suicide risk than the general population, especially during the first year after diagnosis (Ludvigsson et al. 2011). Also, other problems which impact on a person's well-being, such as GI symptoms, affect both men and women with CD, but in particular women, to a significantly greater extent than other people (Hallert et al. 2003; Midhagen and Hallert. 2003). It has been described that bowel symptoms among women, living on a strict diet, sometimes tend to be so severe that they feel a sense of powerlessness that leads to a decreased motivation to go on with the diet (Hallert et al. 2003).

Patient Education

Different definitions of PE are used in different studies, but Visser et al. (2001) described a working definition: "Patient education concerns all educational activities directed to patients, including aspects of therapeutic education, health education and clinical health promotion" (Visser et al. 2001). Increased knowledge can lead to an improved health-related quality of life (Leino-Kilpi et al. 2005). Current health care policy in Sweden emphasises self-management and support of the patient's participation as ways of improving patient outcomes and reducing costs. The main objective of PE is to motivate the person to comply with the prescribed treatment and adopt a lifestyle that promotes health (Strömberg. 2006). Unfortunately, educational efforts performed by nurses are not always evidenced-based.

PE is not just a way for the patient to acquire knowledge. The knowledge must simplify and improve life for the patients and they must see the benefits of any lifestyle changes (Iqbal et al. 2008; Kennedy and Rogers. 2001).

Knowledge alone is thus limited to inducing a person to promote changes in self-care management unless the person feels motivated. Merely knowing your blood sugar value does not benefit the patient if he or she doesn't understand the inner meaning of this particular value (Harwell et al. 2002; Iqbal et al. 2008). This implies that theoretical knowledge does not automatically mean that the person knows what action should be taken. Therefore it is necessary to start any educational effort directed towards a specific group of patients by investigating their daily life experiences and their

knowledge needs in order to be able to develop an intervention that patients experience as motivating. This action has, to our knowledge, never before been performed with regard to women with CD.

Patient education is widely recognized as a core component of nursing. In order to deliver PE that empowers patients, it is essential for nurses to change from feeling responsible for patients to feeling responsible to patients (Booker et al. 2008), which means to shift from a top-down power relationship and adopt a bottom-up one instead (Cooper et al. 2001). Nurses prioritize PE in their work, but believe that there are different factors, like time, staffing and receptiveness of the patient, that hinder their work. Necessary resources must therefore be provided in order to enhance the provision of PE (Marcum et al. 2002). Roles and responsibilities for different health care professionals should be clarified in order to develop appropriate strategies, concludes (Taylor et al. 2011), and another important component to strengthen the quality of PE is also that nurses must evaluate their efficacy to a greater extent.

There are currently many different educational models for PE. One of them is PBL, which is one way of bringing education, counselling and behaviour intervention together in order to support self-management in people with chronic diseases. Despite a limited number of attempts to use this method, there is some evidence of its effectiveness in promoting self-management with individuals experiencing diabetes, arthritis, asthma and coronary artery disease (Williams and Pace. 2009). There are no studies of the CD population using this approach. These factors combined settled the choice of using PBL as a pedagogical model for the PE intervention in this thesis.

Problem Based Learning

PBL is different from the passive memorization of knowledge that occurs in traditional lecture and reading situations (Barrows. 1985). One theoretical perspective on PBL is offered by constructivist epistemology, a learning theory recently developed which is based on the work of Lev Vygotsky and followers. Central to the constructivism theory is that knowledge is considered as being “constructed” by the learner’s cognitive activities in interaction with other people in a social context, of which the learner is a member (Rideout.

2001). According to Charles Engel the PBL approach can be seen as a way of learning rather than a way of teaching. Don Margetson argues that knowledge valued in PBL is that which can be used in a context rather than what motivates the structure of a particular discipline. He believes that we do students a disservice if we deliver answers without enabling them to learn how they came about and how to find them (Boud and Feletti. 1997).

Three essential characteristics distinguish the process of PBL. The first promotes that learning is based on a problem, a question or a puzzle that the learner wishes to solve. Secondly, a student-centred nature is adopted, with its emphasis on self-directed learning. With the given problem as a start, the learners are responsible themselves for deciding what they want to know, what they need to learn and what resources they have available. Thirdly, the method involves working in small groups (5-10 people) (Rideout. 2001). Small groups promote opportunities for the participants to exchange personal experiences and knowledge in relation to the problem to be solved, and this facilitates the problem-solving process (Barrows. 1985). The PBL approach strives towards independence, functional knowledge, critical thinking and lifelong learning (Rideout. 2001). Problems in PBL are always based on situations drawn from everyday life, and the problem-solving process facilitates the possibility to apply new knowledge in real situations henceforth (Maudslay. 1999).

AIMS

The overall aim of the thesis was to study the lived experience among women with coeliac disease in remission and to assess the effects of patient education with problem based learning on psychological well-being and gastrointestinal symptoms.

The specific aims of the thesis were as follows:

Study I

To describe the life experience of being a woman with coeliac disease in Sweden.

Study II & III

To evaluate the effect of a PE programme with PBL versus participation in a wait-list control group, receiving written information corresponding to the information these patients meet in contact with health care, on psychological well-being (II), and GI symptoms (III) for women with coeliac disease in remission.

METHODS

Design

Different designs have been used in this thesis to answer research questions. Study I is an interview study with a phenomenological approach. Studies II and III are based on the same study, a patient education intervention with PBL, performed as a randomized controlled trial (Table 1).

Table 1. Overview of research design for the qualitative and quantitative data collection in papers I–III.

Paper	Participants	Method of data collection	Method of data analysis
I	n=15	Audio taped personal narrative interviews	A phenomenological 4-step analysis as devised by Giorgi
II	n=106	Randomized controlled trial with a pre-post design	t-test statistics
III	n=106	Randomized controlled trial with a pre-post design	t-test statistics

Participants and procedure (I-III)

All three studies were based on the same sample. To find women candidates, dieticians working in hospitals in five Swedish cities (Linköping, Norrköping, Västervik, Värnamo and Jönköping) were asked for help to identify potential participants through medical records. Additionally, advertisements in coeliac disease association member papers invited women to participate. Participants were invited, by postal mail, to an information meeting in one of the towns in

question. At these meetings, verbal as well as written information was given regarding all parts of the study. A total of 106 women were enrolled.

The inclusion criteria were: Women aged 20 years or above with coeliac disease who had been treated with a GFD for a minimum of five years. Exclusion criteria comprised concomitant serious health disorder. Two women with other ongoing diseases (malignancy and Parkinson's disease), which could affect the results of the questionnaires but could also hinder their chances to fulfill the entire PE program, were excluded from the study. Prior to starting, blood samples were drawn for measuring tissue transglutaminase antibody levels to confirm the CD remission state.

After obtaining written consent, randomization (II - III) was done according to place of residence, with consent forms used as lots. This was done in such a way that consent forms were placed into heaps depending on which city the person who completed it belonged to. Each heap was mixed well by hand, and randomization was thereafter done for one heap at a time by alternately sorting forms to the intervention group and to the control group. This procedure, performed by the author, resulted in 54 participants in the intervention group (divided into 7 groups in the five towns) and 52 participants in the control group. Based on the total number of women randomized to the intervention group (n=54), fifteen women (2-4 individuals per city) were also asked to participate in the interview study (I). These women were selected on purpose; i.e. deliberately selected in a non-randomized approach (Patton. 2004), to achieve heterogeneity with regard to years since diagnosis (≥ 5 years), residence, age, civil status and education. All women who were asked responded positively to participating.

Three women in the intervention group did not complete the trial at the ten week follow-up due to a workload situation (one person), lack of time (one person) or onset of another illness (one person), and one person failed to complete the questionnaire at the six-month follow-up (due to lack of time) within the intervention group. Dropouts from the study were treated according to the principle of "intention to treat"; i.e. analysis was based on the initial treatment intent and not on the treatment eventually administered. After ten weeks, the control group was offered participation in the same "Coeliac school" as the intervention group due to ethical reasons, and was therefore no longer included in the study. Twenty-eight women out of 52 accepted this offer. All control group members completed their questionnaires at the ten week follow-up.

Table 2. Characteristics of women in the interview study (I; n=15), and in the randomized control study (II-III; intervention group (n=54); and control group (n=52).

Group	Age in years		Time since diagnosis (years)		Civil status (n)			Domicile (n)			Education level (n)		
	Mean	Range	Mean	Range	Married	Single	Widow	Town	Village	Rural	Primary	Secondary	Higher
Study I	55.8	30-75	19.7	5-67	10	3	2	13	1	1	5	8	2
Intervention Study II-III	51.8	23-80	14.9	5-67	35	15	4	38	14	2	9	20	25
Control Study II-III	53.6	23-74	15.5	5-51	39	10	3	26	20	6	8	17	27

The studies purposely started with interviews to identify how women with CD experience daily life, and this knowledge was then used in the development of the PE in Studies II-III.

In the next section data collection and data analysis in Study I will be presented. Thereafter follows a description of the intervention, data collection and statistical analyses in Studies II-III.

Study I

Phenomenology

Study I was performed in the year 2008. Phenomenological research in general strives to clarify situations that people experience in their daily lives, i.e. to study specific phenomena in which the interviewee has first-hand experience and can describe how it actually took place (Giorgi and Giorgi in Smith, 2003). In the first study of the current thesis, descriptive (eidetic) phenomenology has been used as the research approach. It was chosen because life experiences

were the focus for this research, but also this approach has been demonstrated as being useful in uncovering essences of phenomena that haven't been completely conceptualized by prior research (Beck. 1992). The central figure of phenomenology, Edmund Husserl (1859-1938), argued that the way that such a phenomenon appears in everyday life, the so-called lifeworld, constitutes valuable knowledge, and with a scientific approach the essential components, i.e. the essence, of the experience will emerge (Lopez and Willis. 2004). The essence of consciousness is intentionality, which implies that mental acts of all kinds are intentional in such way that they point to something or some object that is not consciousness itself (Aanstoos and Giorgi. 1985). Husserlian phenomenology emphasizes the importance for the researcher of shedding all prior knowledge in order to grasp the essential lived experiences of those people studied (Lopez and Willis. 2004); i.e. one must be present at the "given" as it presents itself, without either adding or subtracting (Giorgi. 2009). For this reason there was no question guide, other than the one opening question with the desire for a description of the lived experience of being a woman living with CD, which was constructed for this study (I). The goal is, as a researcher, to achieve "transcendental subjectivity", which means that the impact on results derived from the researcher her/him-selves must be constantly assessed and confounding factors neutralized (Lopez and Willis. 2004). Within the descriptive phenomenology "bracketing" or "epoché" is proposed for this purpose; i.e. the researcher puts past knowledge about what is presently given aside so the attention can be directed to what is present (Giorgi. 2009). The author's pre-understanding of the subject of coeliac disease was derived from occasional meetings with children and adolescents with CD as a nurse in a paediatric unit and privately with people living with the disease. The pre-understanding regarding the studied phenomenon was written down by the author as a way to identify own knowledge and preconceptions in order to raise awareness of them. Phenomenology is an approach that takes informants' subjective experiences as its main focus which, from a nursing perspective, must be considered of superior value to study.

Data collection - Interviews

The data in study I comprises 15 tape recorded interviews. All interviews were conducted by the first author (LRJ). Personal narrative interviews (Kvale and

Brinkmann. 2009) were held before the start of the “Coeliac school” because it was considered important that participants had not been affected by this. The interviews took place in a suitable room at the hospital in each city involved, or in the woman’s home if this was the choice of the informant. The interviews lasted 30-60 minutes. In order to focus on the phenomenon, i.e. what life is like as a woman living with coeliac disease, all interviews started with the same question, namely: Will you please tell me what it is like to live with coeliac disease? Informants were encouraged to talk freely and probing questions were asked when the interviewer searched for a deeper explanation of the phenomenon, such as: “What happened then? Can you say some more about that? Can you give me an example?” One pilot interview (not included in the analysis) was carried out specifically to test the question and interview skills. The interviews were transcribed verbatim by the first author (LRJ) and emotions that emerged during the interviews, i.e. laughter, tears, snorts etc. were noted in the transcribed text.

Analysis

The analytic process in study I followed the general outlines derived from Giorgi (2009). The aim of the analysis is to, as close as possible, capture the phenomenon as experienced within the context in which it takes place. The phenomenological analysis seeks to discern the essence of the phenomenon, i.e. it looks for the meanings that constitute the phenomenon by analyzing the contexts of the participants’ lives (Smith. 2003).

The text was analyzed in four steps:

1. Naïve reading, i.e. the interviews were read through, at the same time as listening to the tapes, in order to get a first superficial interpretation. This step provided ideas about the sense of the whole and how to proceed with further analysis.
2. The interviews were then read once again, but this time with the goal of identifying smaller parts, meaning units, within the perspective of the phenomenon of being a woman living with CD. To avoid theoretical explanations, data was kept at as concrete a level as possible. A meaning unit could be part of a sentence or a paragraph.

3. Transformation. In this step, the content of each meaning unit was made clear from the scientific perspective of nursing. Such a description can be conducted on the basis of the question: What does this particular meaning unit tell me as a health care professional about the experience of living with CD? This is a process of reflection and imaginative variation, i.e. an analytic process where the content is varied until the most accurate description is obtained. The transformation of meaning units is helpful in identifying the components of the phenomenon, the so-called constituents.
4. In the last step of the analysis the relationships between the meaning units and the constituents and between the constituents themselves were investigated in order to understand the deeper structure of the phenomenon. This understanding requires consideration of the data, both in parts and as a whole. The general structure, i.e. a new whole, was synthesized into a depiction of the phenomenon of being a woman living with coeliac disease.

The result of the descriptive approach includes descriptions of findings rather than theories or hypotheses (Giorgi. 2009).

In order to assure trustworthiness in study I, various actions have been taken. According to Lincoln and Guba (1985) a qualitative study has to compile certain criteria. Credibility, i.e. confidence in the truth of the findings (Lincoln and Guba. 1985), was in this study pursued by carefully following the research design step by step (Aanstoos and Giorgi. 1985; Giorgi. 1997; Giorgi. 2009), and by providing examples of results by using quotations. Transferability shall be sought by giving a thick description, with sufficient details, that enables the readers to evaluate to what extent conclusions drawn are transferable to other settings (Lincoln and Guba. 1985). The aim was to provide as rich as possible a description of the informants' experiences. Dependability means the way the researcher shows that the results are consistent and could be repeated (Lincoln and Guba. 1985). In this study the first author (LRJ) performed and transcribed all interviews. The supervisors gave comments and criticism on the interview technique as well as concentrating on validation of the results by questioning each step in the analysis to find possible alternative interpretations. The analysis was discussed until agreement was reached.

Study II-III

Intervention

Study II & III was performed during the years 2008-2009. An expert in the field of PBL was linked to the project and influenced the design of the intervention. The groups were conducted by a tutor, and it was this person who, after receiving names from the first author of the women randomized to the intervention group, invited participants to the educational program, the so-called "Coeliac school", which started a few weeks after the completion of interviews in study I. Women (n=54) randomized to the intervention group took part in a ten-session educational program, with weekly meetings in groups of 7-9 people. The meetings were held, for the purpose of ensuring suitable premises, in hospitals (4 groups) or universities (3 groups). Each meeting lasted about 1.5 hours, but the time varied slightly depending on the content of the meeting in question. A total of five tutors, all women, were involved in the project, of which four were nurses and one was a behavioural scientist. It wasn't necessary for the tutors to possess special knowledge regarding gastroenterology or coeliac disease, but it was regarded as crucial that they were familiar with the PBL pedagogy. In cases where the tutor had no prior knowledge at all regarding the pedagogy of PBL (one tutor), or if the person's experiences had faded over time (one tutor), the expert in the field who was assigned to the project trained these tutors in their home town. This field expert was also available to the tutors via email and telephone throughout the intervention period to answer pedagogical issues that arose in the groups.

In accordance with the PBL philosophy (Maudslay. 1999) three main features were emphasized in the education program of this study: working in small groups, starting from real life situations, and using a problem-solving process that stimulated self-directed learning. The main purpose of the programme was to support and encourage the participants to find possible facilitating changes in lifestyle, i.e. tools to master their own disease, and thereby increase their subjective well-being and achieve new knowledge within the area.

At the first meeting, tutors introduced PBL to the group participants, as a pedagogical philosophy and method of learning. Each session considered a predetermined specific topic, presented as a scenario that illustrated the subject of the session in some way. The different scenarios in the course could be selected in the order that participants thought was appropriate. The ideas when developing the scenarios used originated from the results of study I and prior studies (Sverker. 2005; Sverker et al. 2007; Sverker et al. 2009). The scenarios took their starting points from everyday problems, which are part of the fundamental ideas of PBL (Rich et al. 2005) and had the form of a short text, a picture or a section of text from a daily paper or periodical. Areas covered were: anxiety and fears associated with CD, obstacles in daily life (Being in control), attitudes to surroundings (Being seen and included), psychological reactions, coping strategies, (Being secure), new knowledge in the field and various questions associated with food and cooking. The work in the groups with the scenario in question was based on the problem-solving process, whose steps can be seen in Table 3 (Moust et al. 2005). The process of learning in PBL starts with a brainstorming of the current scenario, which concludes with presenting problems of interest. The participants then summarize tasks to investigate together (Kamwendo et al. 2004). The group members performed self-studies between the meetings, and tried to answer the agreed questions. At the next meeting, group members discussed new information they had collected and reflected upon what impact this information would have on their daily lives. New questions of interest then arose after a new brainstorming and so on (Wikblad et al. 2004) .

Table 3. The problem-solving steps used in the “Coeliac school”

Step 1	Clarification of terms and phrases
Step 2	An exact description of the phenomenon that needs to be explained or understood is formulated
Step 3	Brainstorm to formulate various explanations
Step 4	Elaboration of a “theory” regarding the process underlying the phenomena
Step 5	Formulation of issues for self-directed learning
Step 6	Self-studies in order to fill the knowledge gaps
Step 7	Integrate the new knowledge from the group members and evaluate if this knowledge is sufficient and the process to get there

According to (Boud and Feletti. 1997), the tutor shall guide and assist the group through each stage of their discussion and decision making. The tutor shall also prevent and mediate difficulties in the group interaction if such occurs. The tutors' role in the groups was to act as moderators and not as teachers.

The expert on the issue of PBL served as a supervisor for the tutors in the current study, throughout the intervention period. The participants in the "Coeliac school" received a folder with information about the PBL method and also different areas of the scenarios.

All groups had the opportunity, if requested by the participants, to invite various external resources to the groups, such as nutritionists, chefs, physicians etc. These people did not function as lecturers in the groups but were included in particular meetings and answered the questions which had either been compiled in advance or which arose during the meeting. In all groups, at some point, group members discussed where to find reliable information about the disease, and also investigated what books about CD had been published in recent years. The groups started at intervals of a few weeks in order to give the supervisor the opportunity to help one group at a time to get started. Group attendance was high (one person was away for three consecutive meetings before she took the final decision, because of a new work situation, to quit the study. In addition, a total of five people were absent at some point).

Women (n=52) randomized to the control group got information about which group they belonged to by postal mail. They then received a total of five circulars by postal mail over a ten-week period, containing written information. The information covered evidence-based details on CD corresponding to that offered to adults with established CD seeking medical advice for the disorder in primary care. This information consisted of brochures dealing with origins, symptoms, diagnosis and treatment concerning CD, and information about current research within the area.

Enrollment

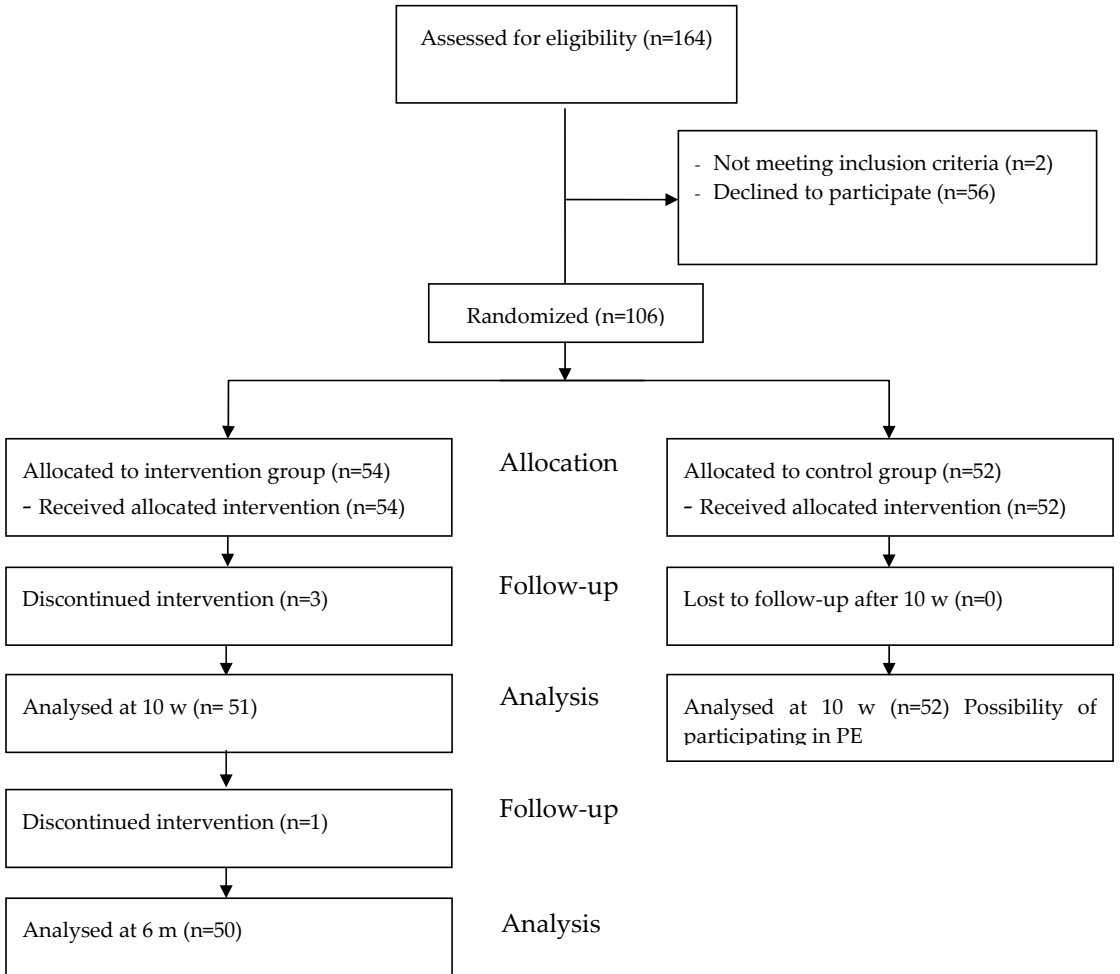


Fig 1. Flowchart of events in study II-III, from recruiting participants to study completion.

Data collection

Data in studies II and III was collected at baseline and after 10 weeks in both the intervention group and in the control group. Six months after completed intervention, data was collected again with regard to intervention group

members. After randomization, at baseline, all questionnaires were sent out by post, as was the case for the control group at 10 weeks follow-up. The intervention group received the questionnaires from the group tutor at the two follow-ups, but completed them at home. These questionnaires were then sent back in a common envelope for the whole group at the next group meeting. The members themselves put the completed questionnaires in the envelope, which was then sealed. The tutor of the group posted the envelope. The two questionnaires used to assess subjective well-being over the past week were self-administered. Both instruments had well-documented reference values for a healthy normal population (Dimenas et al. 1996).

For the assessment of general well-being (II), the Psychological General Well-Being (PGWB) index was used. It was developed specifically to measure subjective well-being or distress (Wenger et al. 1984). The PGWB protocol consists of 22 items that, apart from combining into a global overall score, are combined in six dimensions: *anxiety* (nervousness, tension, anxiety, relaxedness, stress), *depressed mood* (depressed, downhearted, sad), *positive well-being* (general spirits, happy, interested in daily life, cheerful), *self-control* (firm control, afraid of losing control, emotionally stable), *general health* (bothered by illness, healthy enough to do things, concerned about health) and *vitality* (energy, wakes feeling rested, vigorous, tiredness). Each dimension comprises three to five items (see above) using a six-grade Likert scale (Glise et al. 1995). The maximum value of 132 indicates optimal well-being and the minimum value of 22 corresponds to a very poor level of well-being (Dimenas et al. 1995). Mean scores in a healthy normal population are 101 for females (Glise et al. 1995). The PGWB index has good evidence supporting internal consistency, validity and test-retest reliability, and has also been shown to be sensitive to the presence of GI disease (Dimenas et al. 1996; Glise et al. 1995; Revicki et al. 1996).

Gastrointestinal symptoms (III) were evaluated with the Gastrointestinal Symptom Rating Scale (GSRS). The instrument consists of 15 items, each asking how bothered a person is about the presence of commonly reported gastrointestinal symptoms (Glise et al. 1995). Subjects respond using a seven-grade Likert scale defined by verbal denominators that range from no symptoms to the most pronounced symptoms (1=no discomfort at all, 2=minor discomfort, 3=mild discomfort, 4=moderate discomfort, 5=moderately severe discomfort, 6=severe discomfort, and 7=very severe discomfort) (Wiklund et al. 2006). Based on a factor analysis, the 15 items break down into five

dimensions: *Abdominal Pain* (abdominal pain, nausea, and hunger pains), *Reflux syndrome* (heartburn, acid regurgitation), *Indigestion syndrome* (borborygmus, abdominal distension, eructation, increased flatus), *Diarrhoea syndrome* (diarrhoea, loose stools, and urgent need for defecation) and *Constipation syndrome* (constipation, hard stools, feeling of incomplete evacuation) (Dimenas et al. 1995). The gastro intestinal (GI) total index is the mean of these 15 symptoms, and the index value for a clinical syndrome is the mean of all items within an individual scale (Wiklund et al. 2006). Mean scores for GI-total index in a healthy normal population are 1.56 for females (Dimenas et al. 1996). In European patient populations the GSRS received good internal consistency reliability (Dimenas et al. 1995), and acceptable construct validity and responsiveness (Dimenas et al. 1993; Dimenas et al. 1995; Glise et al. 1995).

Demographic data was collected by a questionnaire constructed by the authors that was distributed together with the consent form during the information meeting in the cities included. Issues included information concerning other ongoing diseases, age, time since diagnosis, civil status, domicile and level of education.

Statistical analyses

A parametric test was regarded as appropriate for analysis, since data in studies II and III was normally distributed and summarised in an index.

ANOVA was used for comparison between the assessments at baseline, ten weeks and six months after completing the intervention in the intervention group. A paired t-test was used to compare within each group, from baseline to ten weeks in both groups and from baseline to six months in the intervention group. An independent T-test was used to analyse the difference between groups at baseline and also to analyze the difference in improvement between groups from 0-10 weeks.

A comparison of scores between the intervention group and the control group from baseline to ten weeks was performed with a t-test for Equality of Means where equal variances were not assumed. The significance level was set at $p < 0.05$.

Statistical Package for Social Science, version 17 was used for statistical analysis.

Ethics

All participants who took part gave their informed consent and the studies were conducted according to the Declaration of Helsinki. The studies were approved by the Regional Ethical Review Board at Linköping University, Linköping, Sweden (Dnr M218-08). Studies II and III have been registered at www.clinicaltrials.gov: (NCT 010 88152).

At the first meeting, all intervention groups wrote a contract in which the members agreed that everything that was said during the group meetings would stay in the group and should not be passed on. For ethical reasons, all members of the control group were offered participation in PE, corresponding to “Coeliac school”, after 10 weeks.

RESULTS

The life experience of women with coeliac disease (I)

The general structure of what it is like being a woman with coeliac disease was described as a striving towards a normalised lifeworld. Different experiences of the phenomenon appeared, expressed here as the constituents, and the balance of thoughts within each constituent proved to be crucial for the individual woman's ability to experience a normal life despite the disease. In time, when one of the constituents was disturbed, a normalised lifeworld could not be experienced. The women's narratives revealed both positive and negative sides of these constituents, and life with CD meant a constant movement between conflicting feelings.

The three constituents found in the material were:

Being secure vs. being insecure in different situations in life was crucial to whether life would be perceived as normal or not. Their own homes offered security for all informants, but as soon as they left it the problems started. Insecure informants often experienced distrust and a limited ability to make choices in life, while secure informants considered themselves able to make deliberate choices that facilitated daily life by choosing the "right" sort of thoughts.

// I'm fortunately the kind of person that tends to turn most things into something positive in life. I think that it is lucky that I cannot eat a lot of cakes!

Being in control by planning vs. loss of control was an experience that informants sometimes could maintain control over, mainly through good planning, for example by bringing something yourself to eat when invited to someone. Other situations were experienced as difficult to maintain control over, such as the days when informants had to be close to a toilet, or thoughts on possible consequences when getting old.

// As long as you yourself can control this I do not believe that there is any major risk, but, but the day you cannot control yourself.....What will happen then? It is easy that you then get both this and that (food)!

Being seen and included vs. not seen and excluded were experiences that emerged in relation to how women were responded to in relation to their disease. When other people showed positive attention in terms of knowledge and caring, such as cooking the same food for all guests, feelings of being seen and included arise. When, instead, people around acted with forgetfulness, unwillingness and ignorance, informants perceived themselves to be ignored and badly treated, almost invisible.

// Oh no! It is impossible to invite you home! It is so hard! You cannot eat anything! (Talks about what people often say)

Effects of the PE-programme on psychological well-being (II)

At baseline the participants in the control group stated a better total PGWB-index value ($p=0.027$) compared to the intervention group (Table 4).

An ANOVA of total PGWB-index values showed a significant difference within the intervention group between baseline, ten weeks and six months after completed intervention ($p=0.024$).

Table 4. Index outcome for PGWB in intervention group: at baseline, after 10 weeks and 6 months after completed intervention and in control group: at baseline and after 10 weeks. Positive mean diff means improvement of symptoms.

PGWB	Intervention group								Control group					
	Mean index and SD				Mean diff (10w-0w)	Mean index and SD			Mean diff (6m-0w)	Mean index and SD				Mean diff (10w-0w)
	0w	SD	10w	SD	n=51	6 m	SD	n=50	0w	SD	10w	SD	n=52	
Anxiety	22.1	5.2	23.4	4.9	1.3*	22.9	5.4	0.7	23.8	4.2	23.6	4.3	-0.2	
Depression	15.0	3.1	15.9	2.6	0.8*	15.3	3.0	0.3	15.9	2.5	15.0	2.7	-0.8	
Well-being	15.6	3.7	16.5	4.2	0.9*	16.4	4.2	1.0	17.2	3.6	15.2	4.0	-2.0**	
Self-control	14.8	2.9	15.0	2.7	0.3	14.8	3.3	0.0	15.3	3.0	13.3	4.8	-2.0**	
General health	13.3	2.1	14.4	2.7	1.2**	13.8	3.1	0.6	14.4	2.8	13.5	3.3	-0.9*	
Vitality	14.6	4.4	16.1	4.7	1.5**	16.1	4.9	1.5**	16.9	4.1	13.9	6.2	-3.0**	
Total	95.4	18.3	101.4	19.1	6.0**	99.3	21.0	4.1	103.3	17.2	94.4	22.9	-8.9**	

Note. PGWB = Psychological General Well-Being

*p<.05, **p<.01 vs. baseline

The answers to questionnaires within each group revealed a significant increase in total PGWB-index value in the intervention group, after the ten-week PBL program (p=0.001). This was particularly the case for the indexes: Anxiety (p=0.016), Depression (p=0.023), Well-being (p=0.026), General health (p=0.002) and Vitality (p=0.002). This was not the case in the control group, which instead presented a significant worsening of total PGWB-index value (p=0.006) particularly for the indexes: Well-being (p=0.002), Self control (p=0.007), General health (p=0.03) and Vitality (p=0.002).

Six months after completing PE, improvement in the intervention group, compared with the baseline, remained significant for the index of Vitality (p=0.009) (Table 4).

A comparison of scores between the two groups showed a positive development, reflecting the participants' subjective general well-being from baseline until after ten weeks in the intervention group. This was found regarding all indexes, compared with the development in the control group. The difference in improvement was significant concerning the indexes: Depression (p=0.003), Well-being (p<0.001), Self-control (p=0.004), General health (p<0.001), Vitality (p<0.001) and for the total PGWB-index value (p<0.001) (Table 5).

Table 5. Comparison of mean PGWB improvement from baseline to 10 weeks between intervention group (n=51) and control group (n=52). Positive mean diff means improvement of symptoms.

PGWB	95% CI of the Difference		
	Mean diff	Lower	Upper
Anxiety 0w-10w	1.5	1.35	1.65
Depression 0w-10w	1.7**	1.60	1.80
Well-being 0w-10w	2.9***	2.76	3.03
Self-control 0w-10w	2.2**	2.05	2.35
General health 0w-10w	2.0***	1.90	2.10
Vitality 0w-10w	4.5***	4.31	4.63
Total 0w-10w	14.9***	14.20	15.60

Note. PGWB = Psychological General Well-Being

*p<.05, **p<.01, ***p<.001

Effects of the PE-programme on gastrointestinal symptoms (III)

At baseline the participants in the control group stated a better total GSRS-index value (p=0.009) compared with the intervention group. After ten weeks there was no longer any statistical significant difference in GI symptoms between the groups (Table 6).

Table 6. Index outcome for GSRS in intervention group: at baseline, after 10 weeks and 6 months after completed intervention, and in control group: at baseline and after 10 weeks. Negative mean diff means improvement of symptoms.

GSRS	Intervention group									Control group				
	Mean index and SD				Mean diff (10w-0w)	Mean index and SD			Mean diff (6m-0w)	Mean index and SD				Mean diff (10w-0w)
	0w	SD	10w	SD		n=51	6 m	SD		n=50	0w	SD	10w	
Indigestion	3.0	1.2	2.8	1.3	-0.2	2.7	1.2	-0.3	2.4	1.0	2.4	1.1	0.0	
Diarrhoea	2.9	1.8	3.0	1.7	0.1	2.4	1.6	-0.5	2.3	1.4	2.4	1.6	0.1	
Constipation	2.5	1.5	2.1	1.1	-0.4*	2.2	1.3	-0.3	2.2	1.4	2.3	1.3	0.1	
Abdominal pain	2.7	1.2	2.1	0.9	-0.6**	2.2	1.0	-0.5*	2.0	0.9	1.9	0.9	-0.1	
Reflux	1.6	1.0	1.6	0.9	0.0	1.7	1.1	0.1	1.4	0.6	1.3	0.5	-0.1	
Total	2.5	1.0	2.3	0.9	-0.2*	2.2	1.0	-0.3*	2.1	0.7	2.1	0.8	0.0	

Note. GSRS = Gastrointestinal Symptom Rating Scale.

*p<.05, **p<.01 vs. baseline

The answers to questionnaires within each group revealed a significant improvement in total GSRS-index value in the intervention group, after the ten-week PBL program ($p=0.013$), and for the indexes: Constipation ($p=0.032$) and Abdominal Pain ($p=0.001$). Six months after completing PE the total GSRS-index, for the intervention group, still showed statistically significant improvement in comparison to baseline ($p=0.029$), with general improvement in all dimensions (except for Reflux), with only Abdominal Pain showing a statistically significant improvement ($p=0.01$). The control group did not report a significant change in GSRS-index value (Table 6).

A comparison of scores between the two groups showed a significant positive development in the intervention group compared with the control group, regarding the index Abdominal Pain ($p=0.007$), but no significant difference regarding the other indexes or the total GSRS-index (Table 7).

Table 7. Comparison of mean GSRS development from baseline to 10 weeks between intervention group (n=51) and control group (n=52). Negative mean diff means improvement of symptoms.

GSRS	95% CI of the Difference		
	Mean diff	Lower	Upper
Indigestion 0w-10w	-0.17	-0.20	-0.14
Diarrhoea 0w-10w	0.03	-0.02	0.08
Constipation 0w-10w	-0.42	-0.46	-0.38
Abdominal pain 0w-10w	-0.47**	-0.50	-0.44
Reflux 0w-10w	0.08	0.05	0.11
Total 0w-10w	-0.19	-0.21	-0.17

Note. GSRS = Gastrointestinal Symptom Rating Scale

* $p<0.05$, ** $p<0.01$

DISCUSSION

This thesis found that coeliac disease can influence women's lives in different ways, and that a PE based on PBL improves psychological well-being and gastro-intestinal symptoms to some extent. But irrespective of what impact the disease has on an individual woman's life, they all have a common goal, which is a striving towards a normal lifeworld (I). The desire to achieve some semblance of normality in life has previously been highlighted among chronically ill people (Miles et al. 2005; Sturge-Jacobs. 2002). In this study (I) women's goal of living a normal life was described as requiring three conditions, namely: being secure, being in control, and being seen and included. One can reflect on what the term "normal life" means in a deeper sense. Is it about living like you did before the diagnosis or living like other people in society, or something else? 'Normality' can constitute a significant factor towards how a person with a chronic condition chooses to respond to illness in a family setting (Gregory. 2005). Women (I) who felt secure in most situations expressed that they themselves, by deliberately choosing the right kind of thoughts, could decide how their life would look. It therefore appears that these women are successful in finding coping strategies on their own, to handle everyday problems in relation to their chronic illness, and can thus achieve a high level of well-being. Other women considered that the disease had very negative consequences on their well-being.

Patient education with PBL can be useful in order to help women with CD to achieve higher levels of well-being in life. In this study, participants in the "Coeliac school" reported significant improvement in psychological well-being (II) after ten weeks, whereas the controls, given normal care, reported a worsening. Also, with regard to gastrointestinal symptoms (III), it was found that women participating in PE reported significant improvements. On measuring this outcome again six month later, the effect remained significant.

There are several possible educational choices which are useful in the context of PE. The choice of educational model for these studies (II-III) was based on the fact that PBL seemed to fit well with the requirements that previous studies (Booker et al. 2008; Harwell et al. 2002; Iqbal et al. 2008; Kennedy and Rogers. 2001) have described that PE should meet, but also because it hasn't been tried for this group of patients before. Within the PBL pedagogy the learning process starts with patients' beliefs about the disease and its

treatment. The patients' interpretations of the disease may not correspond with current medical knowledge, and unexplored feelings may hinder the patients learning process (Jayne and Rankin. 2001). Therefore, it was thought a good idea to start with a qualitative study to identify the core beliefs that existed, as has been done in this thesis.

One of the positive aspects of PBL is that not only is the patients' immediate need for knowledge remedied, but they are also given the tools to be proactive, by searching for reliable information for use in the future. This helps improve their self-care behaviour. However, it is not only knowledge that is required to enable changes in ingrained patterns in life. Motivation is crucial, and PE must be designed in such a way that the person sees the benefit of any change in habits and, according to this study (I), ways of facilitating their strive for a normalised life. Another important factor that must not be overlooked when planning interventions is that PE cannot be administered as a single dose. People have to remain motivated in order not to return to their old habits. In the present studies (II-III) the effect had already waned at the follow-up 6 months after completed intervention, and in particular regarding women's psychological well-being. Management for a person with a chronic disease also necessarily varies over time (Hepworth and Harrison. 2004), with adjustments based on changes regarding symptoms and fluctuations in the disease process. This poses a challenge when designing PE for people who have had their diagnosis for different lengths of time. Could these variations in disease process be the reason why our result (I) showed two poles (e.g. Being secure-Insecure), between which women's relationship with the disease fluctuates?

The impairment in psychological well-being in the control group at the ten week follow up may emphasise the need for and importance of discussing new knowledge which has been attained about one's disease with others. One can also ask if the procedure of receiving printed information brochures sent to one's home may result in greater anxiety than receiving no information at all. In future studies, researchers should consider having also a control group not receiving printed information.

Despite the improvements in psychological well-being and gastrointestinal symptoms for participants in "Coeliac school", the mean total index reached the normative values for PGWB, valid for women, only at ten weeks but not after six months. At no point in time did this happen for GSRS normative

values (Dimenas et al. 1996). This indicates that the treatment effect of PE as described here (II-III) does not seem to be sufficient for women with CD. It has been found that the presence of a chronic illness is associated with a substantially reduced quality of life and happiness, and increased levels of mood depression (Wikman et al. 2011). Of course, any kind of improvement is important for such people, but it is a bit of a mystery why these women, who are being treated with a GFD and who should be free of symptoms and experience the same level of well-being as men with treated CD, still do not experience this. Although there is little doubt that biological factors are part of the explanation of gender differences in health, researchers in general agree that biological factors alone cannot account for all observed differences. Biological factors probably interact with psychosocial factors like work- and family-roles etc, to produce gender differences in health (Rieker and Bird. 2005). The results from study I have provided a description of the life experiences of women in Sweden living with CD. It remains, for the future, to study the gender-related aspects that can explain the differences in CD symptoms between women and men. The results from such an investigation could be used in the development of more effective PE, with greater impact on these people's well-being, and hopefully with a more pronounced long-term effect. Even today there are traditional gender role stereotypes in the home and in the workplace, cultural expectations of women's bodies and stigmatized beliefs about women's diseases in healthcare. Can these things together result in many women with CD experiencing difficulties in their daily management of the disease, and if so, how can we influence these factors in a PE context? According to the reasoning of Wikman et al. (2011), we know too little about those aspects which, together, contribute to a person's quality of life from a broader perspective, such as material and emotional well-being, autonomy, self-realization, control over important things in life, and meaning and fulfilment (Wikman et al. 2011). We have learned that (I) women with CD are exposed daily to mental stress and a lot of ignorance in society, and sometimes even an attitude among some people who believe that people with CD actually tolerate a certain amount of gluten, leading to these women constantly having to be on their guard. This could be a challenge for public health to educate society.

Another important factor in the well-being of these women is the way in which they are likely to be received by healthcare. It often takes many years to get a proper diagnosis of CD, and before that they have to undergo several diagnostic procedures, incorrect diagnosis, gender stereotyping and scepticism

from medical care which can lead to a choice to suffer in silence (Johnson and Johnson. 2006). CD is a disease that can also have a profound influence on an individual's social life, as many women with CD feel different, excluded, a nuisance, finicky, suspicious, and so on (I). It has emerged that 70% of women with CD exhibit the presence of social phobia of a mild or severe nature, compared to 16% in a healthy control group that moreover showed no severe cases (Addolorato et al. 2008). This says a great deal about the difficulties women with CD encounter in daily life and proves that these women's problems cannot be brushed aside with stigmatizing beliefs. These people deserve to be handled with respect in society.

A nurse's professional role has changed over time and today includes responsibility for providing support to chronically ill individuals so that they can meet the goals set as a result of self-care behavioural changes (Laughlin and Beisel. 2010). Nurses therefore have a central role in providing advice, guidance, education and support. Self-management does not only benefit patients but also allows nurses to use and develop their clinical and interpersonal skills (Davies. 2010). Regarding future PE in order to empower self-care for people suffering from CD, perhaps nurses should encourage women's awareness of social demands originating from gender roles in society and provide them with resources to assert their rights in various situations.

Methodological issues

A qualitative approach was chosen in study I to reach a deeper understanding of what life is like as a woman living with coeliac disease. A phenomenological design was judged to be the most accurate means to describe the essence of the reality of women's life experiences. The number of women included in the study is relatively high for a phenomenological study. The reason for this is that some of the first interviews did not give such a rich description of the phenomenon. One limitation in study I may consist of the fact that all the women who were included had been accepted to the PE program. This circumstance may have influenced the results of this study, because the women either did not experience their problems so greatly at the time, as they believed that help was near, or they unwittingly magnified their problems to prove that they deserved their place on the program. Another consequence of this selection procedure may be that the transferability of results to the whole group of CD women may have been affected, since it was

found that women in the intervention group initially had a lower level of well-being compared with members of the control group.

It is, to our knowledge, the first time that PE with PBL has been carried out for this patient group. The strengths of the study are that the dropout rate was very low (II-III), and the rate of attendance at group sessions was good (II-III). The instruments applied to measure the outcomes (II-III) have been extensively used and their validity and reliability are well documented.

The goal for study II and III was to recruit a larger number of participants. However, this is a relatively small patient population, and we included everyone who was interested in participating and who met the criteria for inclusion in the participating counties. The optimum would have been to include only those individuals who expressed the lowest levels of well-being in relation to the disease, and in a larger number. No power calculation has been performed for studies II and III. To permit such a calculation, a level of what was considered to be an acceptable improvement regarding well-being should have been defined before the studies began. An acceptable improvement in this study could be considered that women in the study would achieve the same levels of well-being as men with treated CD.

Various factors, which can be influenced to a greater or lesser degree, affected the possibility of evaluating the outcome of the intervention (II-III). One is the fact that there was a discrepancy in total mean index value between the groups at baseline, despite randomization, where the controls stated a better psychological well-being (II) and fewer GI symptoms (III). It can be assumed that, for this reason, women in the intervention group were more motivated initially, compared with the control group, since it seems natural that motivation to carry out changes in one's life would increase, the greater the problems are perceived.

The question is, to what extent can the procedure for randomization (I-II) explain the differences between the groups at baseline? It would have been more reliable if women were randomly assigned on the basis of one sample. However, this was not possible as we couldn't request a person living, for example, in Linköping to go to Jönköping to receive PE. For this reason we had to perform randomization for one city at a time. Out of the total number of women randomized to the intervention group, 15 people were interviewed (I) before intervention started. Can these interviews

have affected the outcome of PE, in the sense that these women began to think about factors associated with their life with CD that they had not been so aware of before; i.e. that they normally might not be considered to be a major concern in their everyday life?

The circumstance that control participants were offered PE after only ten weeks constitutes perhaps the greatest limitation in studies II and III, as this procedure precluded a six-month follow-up with this group. This means that it is difficult to determine with certainty that it really was participants of the intervention group that were feeling the best. Also a qualitative evaluation of participants' experiences regarding the intervention, i.e. what was good, or not good, in relation to the content and structure, should have facilitated guidance for the design of future interventions.

Concluding remarks

The reason seem to be very complex why many women who, despite having treated coeliac disease, and thus a healed bowel, are not doing well. Experiences of insecurity and of losing control, and a sometimes uncomprehending attitude of society can contribute to difficulties in relation to a life with the disease.

A PE based on PBL (II-III) can, to some extent, improve psychological well-being and gastro-intestinal symptoms in women with CD, however not sufficiently. It is unclear to what extent stress due to role strain and other factors, which have not yet been researched, may contribute to many women with CD experiencing difficulties in their daily life management of the disease. Future research in this area should focus on finding the answer to this issue in order to develop more effective educational approaches for women with CD.

Clinical implications

This thesis indicates that the lives of women can be influenced by coeliac disease in different ways. The possibility for a woman with coeliac disease to experience a normalised life-world rests on a number of conflicting feelings, and the results of this study provide knowledge, for healthcare and others, regarding the necessary conditions. To facilitate the women's striving for a

normalised life, feelings of security and being in control, seem to be important goals for healthcare who meet women with CD.

A patient education with PBL can be helpful for these women in order to identify strategies that facilitate everyday life. Future intervention should primarily approach the group of women who, themselves, have difficulty in finding coping strategies. Another action that is highly likely to benefit these patients would be if PE was offered shortly after diagnosis. This would probably counteract many of the adverse consequences of this illness in the everyday life of women, before they occur.

CONCLUSION

The following conclusions can be drawn from the three studies in this thesis:

- Coeliac disease can affect women's lives in various ways, but one thing in common was a constant striving towards a normalised lifeworld. To achieve a normalised lifeworld three preconditions were expressed, namely: feelings of security; a sense of having control of the situation; and feelings of being seen by other people. This knowledge can help healthcare professionals to support these women in their aims and in the development of coping skills.
- A patient education program based on an active method for women with coeliac disease can improve the psychological well-being of participants. However, PE cannot be administered as a single dose, but should be replenished regularly, since its effects tend to wear off over time.
- The effect of the same PE on GI symptoms was not sufficient in comparison to the outcome in the control group since the comparison of changes regarding development of scores, from baseline to 10 weeks, in the two groups were not significant.

SUMMARY IN SWEDISH

Svensk sammanfattning

Dagliga livserfarenheter, symtom och välbefinnande hos kvinnor med celiaki. En patientutbildningsintervention.

Bakgrund

Tidigare studier visar att kvinnor med kronisk sjukdom upplever mer symtom än män, medan andra studier hävdar att dylika skillnader inte kan påvisas. Celiaki är en kronisk autoimmun sjukdom som drabbar personer med en genetisk betingning för sjukdomen. Här har forskning upprepade gånger påvisat könsskillnader gällande upplevda symtom och sjukdomsörda. Diagnosen celiaki ställs då en person uppvisar onormal struktur på slemhinnan i tunntarmen som ett resultat av glutenintag, samt en påföljande histologisk och klinisk tillbakagång efter glutenfri diet. Sjukdomen kännetecknas av malabsorption, men många atypiska symtom och ibland inga symtom alls, förekommer. Förekomsten av celiaki i den västerländska befolkningen beräknas ligga nära 1 % och är något vanligare hos kvinnor än hos män. Även efter behandling med glutenfri diet, då tarmen är läkt, upplever personer med celiaki, och i synnerhet kvinnor, lägre välbefinnande jämfört med friska kvinnor i befolkningen, men även jämfört med män med behandlad celiaki.

Patientutbildning intar en central roll i sjuksköterskans omvårdnadsansvar och är en av de åtgärder som kan hjälpa patienter med kronisk sjukdom att uppleva högre livskvalitet. Patientutbildning är inte bara ett sätt att inhämta kunskaper utan måste även bidra till att förenkla och förbättra livet för patienterna, så att de kan se de eventuella vinsterna av genomförda livsstilsförändringar. En annan viktig komponent för att stärka kvaliteten av patientutbildning är att sjuksköterskor måste utvärdera dess effektivitet i större utsträckning.

Det finns i dag många olika pedagogiska modeller användbara i patientutbildningssammanhang. En av dessa är Problembaserat Lärande (PBL). Denna metod strävar mot självständighet, funktionell kunskap, kritiskt

tänkande och ett livslångt lärande. Signifikanta förbättringar efter patientutbildning med PBL har rapporterats för kroniska sjukdomar som diabetes, astma, artrit och hjärtsjukdom. Trots dessa positiva resultat har PBL aldrig tidigare använts i patientutbildning vid celiaki.

Syfte och delsyften

Det övergripande syftet med avhandlingen var att studera kvinnors erfarenheter av att leva med celiaki i remission samt att bedöma effekten av en patientutbildning baserad på PBL avseende psykiskt välbefinnande och tarmsymtom.

Syften med studie I var att beskriva upplevelsen av att som kvinna i Sverige leva med celiaki. Syftet med studie II & III var att utvärdera effekten av ett patientutbildningsprogram baserat på PBL, för kvinnor med celiaki i remission avseende psykiskt välbefinnande (II) och tarmsymtom (III).

Deltagare och tillvägagångssätt

Alla tre studierna baseras på samma deltagare, vilka identifierades via patientjournaler hos dietister på de fem ingående sjukhusen (Linköping, Norrköping, Västervik, Värnamo och Jönköping) samt via annonser i Celiakiföreningens medlemstidningar. Alla kvinnor som uppfyllde inklusionskriterierna blev inbjudna till ett informationsmöte i någon av städerna några veckor innan patientutbildningen skulle ta sin början. Vid dessa möten gavs såväl muntlig som skriftlig information om alla i studien ingående delar. Totalt 106 kvinnor, 20 år eller äldre, med bekräftad celiaki som behandlats med GFD i minst fem år randomiserades (II & III) med utgångspunkt från bostadsort till antingen interventionsgruppen den s.k. "Celiakiskolan" (n=54), eller kontrollgruppen (n=52).

Studie I genomfördes 2008 i form av en kvalitativ intervjustudie med fenomenologisk ansats. Individuella bandinspelade intervjuer utfördes med ändamålsenligt utvalda kvinnor (n=15) tillhörande interventionsgruppen före starten av Celiakiskolan. För att fokusera på det studerade fenomenet startade samtliga intervjuer med frågan: Kan du berätta för mig hur det är att leva med celiaki? Intervjuerna transkriberades ordagrant av författaren och analyserades i enlighet med riktlinjer utarbetade av Giorgi.

Studie II och III är baserade på samma intervention, en celiakiskola baserad på PBL-pedagogik där deltagarna träffades en gång i veckan à 10 tillfällen. Totalt sju utbildningsgrupper startade under hösten 2008. Grupperna bestod av 7-9

deltagare. Det huvudsakliga målet för programmet var att stödja och uppmuntra deltagarna att finna möjliga livsstilsförändringar och därmed redskap att handskas med svårigheter förknippade med sjukdomen. Kontrollgruppen fick information beträffande celiaki hemskickad regelbundet och vid totalt fem tillfällen. Efter tio veckor erbjöds även kontrollgruppens medlemmar att delta i samma patientutbildningsprogram som interventionsgruppen. Celiakiskolan utvärderades före dess start och efter tio veckor beträffande båda grupperna, samt sex månader efter avslutad celiakiskola för deltagarna i interventionsgruppen. Enkäten "Psychological General Well-Being" (PGWB) användes för utvärdering av subjektivt välbefinnande. Tarmsymtomen utvärderades med enkäten "Gastrointestinal Symptom Rating Scale" (GSRS).

Resultat

Studie I visade att celiaki kan påverka kvinnors liv på olika sätt, men gemensamt för alla kvinnorna var en ständig strävan mot en normaliserad livsvärld. Tre förutsättningar för att uppnå detta uttrycktes som: Känslor av trygghet; Känslor av att ha kontroll över situationen; Känslor av att bli sedd av andra människor.

Studie II & III visade att deltagarna i celiakiskolan rapporterade en signifikant förbättring beträffande subjektivt välbefinnande ($p=0.001$) och tarmsymtom ($p=0.013$) efter tio veckors intervention, medan kontrollgruppen inte förbättrades signifikant. Vid den första utvärderingen, dvs. före celiakiskolans start, rapporterade kontrollgruppens deltagare av okänd anledning och trots randomisering, signifikant bättre subjektivt välbefinnande och signifikant mindre tarmsymtom jämfört med celiakiskolans deltagare. Sex månader efter avslutad celiakiskola hade dess positiva effekter avtagit till stor del.

Avhandlingens slutsatser är att celiaki kan påverka kvinnors liv på olika sätt, men gemensamt är en strävan efter en normaliserad livsvärld. En patientutbildning med PBL kan hjälpa kvinnor med celiaki att uppleva en högre grad av välbefinnande. Det är viktigt att erbjuda patientutbildning till de kvinnor med celiaki som inte själva kan finna strategier som underlättar livet med sjukdomen. En annan åtgärd som sannolikt skulle gynna dessa patienter är att erbjuda patientutbildning i samband med diagnos. Detta skulle förmodligen vara kontraproduktivt mot de negativa aspekter denna sjukdom kan medföra i det dagliga livet för de drabbade. Interventionsmetoden bör

dock förfinas i syfte att få en tydligare och mer långsiktig effekt av utbildningen.

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