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**A troublesome diagnosis! Perceptions of illness and self in  
adolescents and adults with psychogenic, non-epileptic seizures  
(PNES)**

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Doctoral thesis by  
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## **Publications**

### **Paper I**

Karterud HN, Knizek BL, Nakken KO. "Changing the diagnosis from epilepsy to PNES: Patients' experiences and understanding of their new diagnosis." *Seizure* 2010; 19: 40-6.

### **Paper II**

Karterud HN, Risør MB, Haavet OR. "The impact of conveying the diagnosis when using a biopsychosocial approach: A qualitative study among adolescents and young adults with NES (non-epileptic seizures)." *Seizure* 2015; 24:107-13.

### **Paper III**

Karterud HN, Haavet OR, Risør MB. "Social participation in young people with non-epileptic seizures (NES): A qualitative study of managing legitimacy in everyday life." *Epilepsy Behav* 2016; 57: 23-8.

## Abbreviations

AED	Antiepileptic drugs
BDS	Bodily Distress Syndrome
CATS	Cognitive Activation Theory of Stress
CBT	Cognitive Behavioural Therapy
CFS	Chronic Fatigue Syndrome
EEG	Electroencephalography
FM	Fibromyalgia
IBC	Irritable Bowel Syndrome
ME	Myalgic Encephalomyelitis
MUS	Medically Unexplained Symptoms
MUPS	Medically Unexplained Physical Symptoms
MRI	Magnetic Resonance Imaging
NES	Non-Epileptic Seizures
PNES	Psychogenic Non-Epileptic Seizures
STC	Systematic Text Condensation
TA	Thematic Analysis

## Norwegian summary

Personer med medisinsk uforklarte symptomer (MUS), inkludert personer med psykogene ikke-epileptiske anfall (PNES), beskriver flere utfordringer knyttet til å ha en omstridt og uklar diagnose. De beskriver blant annet at de blir møtt med mistillit både i helsevesenet og omgivelsene ellers. Eksisterende forskning viser at formidlingen av PNES diagnosen til pasienten kan utløse negative reaksjoner, eller benekting av diagnosen. Dette har aktualisert spørsmål knyttet til hvordan diagnosen formidles og forklares til pasientene. På bakgrunn av dette ønsket vi å undersøke pasienters erfaringer med å få «PNES-diagnosen». Prosjektets overordnede målsetning har vært å utvikle kunnskap om sykdomsforståelse hos personer med PNES. Det er utført tre delstudier for å besvare studiens målsetning.

Det er brukt kvalitativ metode med semi-strukturerte intervjuer med 21 personer med PNES. Deltagerne fordeler seg på to grupper. Gruppe 1 (10 voksne) danner utgangspunkt for del-studie I. Gruppe 2 (11 ungdommer) danner utgangspunkt for del-studie II og III. I tillegg er det utført to oppfølgingsstudier via telefon (en for del-studie I og en for II) samt en mindre oppfølgingsstudie (inkludert noe deltagende observasjon, 4 oppfølgingsamtaler og 6 telefonsamtaler) for del-studie III. Materialet er analysert ved hjelp av systematisk tekstkondensering (del-studie I og II) og tematisk analyse (del-studie III). Materialet er i hovedsak fortolket i lys av følgende teoretiske perspektiver: sosial identitet, diagnose og hierarki og syn på sykdom.

Hensikten med del-studie I var å undersøke hvordan det kan oppleves å bytte diagnose fra epilepsi til PNES, med særlig vekt på hvordan pasientene opplevde sin nye diagnose. Vi så på hva som var vanskelig, og hva som bidro til mestring. Da denne studien ble utført var det ingen fastlagte retningslinjer for hvordan diagnosen skulle formidles på institusjonen hvor studien ble utført. Resultatene viste at det var en emosjonelt og kognitivt krevende prosess å bytte diagnose. «PNES diagnosen» var vanskelig å forstå, og den påvirket deltagerens oppfatning av egen identitet. Forsøk på å forstå seg selv i lys av den nye diagnosen kunne medføre en re-fortolkning av selvforståelsen.

Hensikten med del-studie II var å undersøke hvilken betydning det kan ha å få diagnosen forklart ved hjelp av en biopsykososial tilnærming. Ungdommer som hadde vært innlagt til et 2-4 ukers oppfølgingsopphold for bearbeidelse av diagnosen ble intervjuet. Studien har flere begrensinger som gjorde det vanskelig å utforske betydning av den biopsykososiale tilnærming. Det vi oppnår i studie II er økt innsikt i hvordan deltagerne erfarer diagnosen etter oppfølgingsoppholdet. Deltagernes erfaringer peker på noen tema som kan være av betydning for senere studier. Det gjelder særlig betydningen av å bli trodd, og betydningen av å få en forklaring som gir mening.

Hensikten med del-studie III var å undersøke sosial deltagelse hos ungdom med PNES med særlig vekt på opplevelsen av legitimitet. Vi ønsket å undersøke hvordan ungdommene håndterte sin diagnose i hverdagen, dvs i familien, på skolen, på jobben og i fritiden. Resultatene viste at både ungdommene selv og omgivelsene i vesentlig grad oppfattet PNES som en sosialt illegitim tilstand. Ungdommenes handlingsmønster innrettet seg i stor grad etter dette. En strategi for å takle manglende legitimering fra omgivelsene var å holde diagnosen skjult, noen valgte også å isolere seg sosialt. Støtte fra nær familie og venner, og en meningsfull og legitim sykdomsforståelse, kunne derimot bidra til økt mestring og økt sosial deltagelse.

Avhandlingen presenterer følgende hovedfunn:

- Studien viser at det tradisjonelle biomedisinske paradigmet fremdeles kan være et hinder for at pasienter med PNES tar i bruk forklaringsmodeller som overskrider psyke og soma dualismen.
- Deltagernes sykdomsforståelse var preget av et dualistisk og forenklet syn på sykdom, hvor symptomer som ikke kan forklares ut fra organiske funn ble oppfattet som uttrykk for ensidig psykiatrisk sykdom.
- Identifikasjon med en psykiatrisk diagnose kunne medføre identitetstruende prosesser.
- Opplevelse av økt mening og av at diagnosen ble opplevd som mer legitim, kunne bidra til mestring
- Å bli trodd på at anfallene ikke var simulerte kunne aktivere en økt evne til selv-refleksjon, slik at det ble mulig å endre på fastlåste sykdomsforestillinger.

- Studien pekte også på noen sammenhenger mellom sykdomsforståelse og sosial deltagelse, hvor deltagere som selv opplevde sin tilstand som begripelig og legitim deltok mer sosialt.

## **English summary**

People with medically unexplained symptoms (MUS), including people with psychogenic non-epileptic seizures (PNES), describe several challenges associated with having a contentious and unclear diagnosis. Among other issues, they describe that they meet a lack of trust both from the healthcare services and society. Current research shows that being given a diagnosis of PNES can trigger negative reactions, and several patients do not accept the diagnosis. This has highlighted issues related to how the diagnosis is communicated and explained to the patients. On this background, we wanted to examine patients' experiences with receiving a "PNES diagnosis". The project's overall objective was to develop our knowledge regarding how people with PNES understand their condition. The research was divided into three sub-studies in order to address the study's objective.

A qualitative methodology with semi-structured interviews with 21 people with PNES was used to obtain data. The participants were divided in two groups. Group 1 (10 adults) form the basis for sub-study I, and Group 2 (11 young people) form the basis for sub-studies II and III.

In addition, two telephone follow-up interviews were performed (one for study I, and one for study II), and a minor follow up study (including some participant observation, 4 face to face discussions and 6 telephone conversations) for study III. The material was analyzed by systematic text condensation (sub-studies I and II) and thematic analysis (sub-study III).

Essentially, the material was interpreted in the light of the following theoretical perspectives: social identity, view of disease and diagnosis and hierarchy.

The objective of sub-study I was to examine patients' experience when their diagnosis is changed from that of epilepsy to PNES, with particular emphasis on how patients experienced their new diagnosis. We explored what was most difficult for the patients and what factors contributed to coping with the change in diagnosis. At the time that this study was conducted, guidelines for how the diagnosis should be communicated had not been established. Our results demonstrated that it was a demanding process, both emotionally and cognitively, to receive the new diagnosis. The "PNES diagnosis" was difficult to understand, and affected the individual participant's identity. Patient's efforts to understand themselves in the light of the new diagnosis may lead to a re-interpretation of self-understanding.

The objective of sub-study II was to examine the impact of having the PNES diagnosis explained using a biopsychosocial approach. Teenagers and young adult patients who had been admitted to a 2-4 week follow-up hospital stay for assistance with processing the diagnosis were interviewed. The study has several limitations that make it difficult to explore the impact of using the biopsychosocial approach. What we achieve in sub-study II is greater insight into how participants experience the diagnosis after the follow-up stay. The results nevertheless point to some topics that may be of importance for further studies. This is particularly the importance of being believed, and the importance of getting an explanation that makes sense.

The objective of sub-study III was to examine social participation among youths with PNES, with particular emphasis on the experience of legitimacy. We wanted to investigate how young people handled their PNES diagnosis in everyday life, i.e., with family, at school, at work, and at home. Our results indicated that both young people themselves and their social environment largely tended to perceive PNES as a socially illegitimate state. The young people's behavioural patterns were greatly affected by this perception. One strategy for tackling society's apparent lack of acceptance of PNES as legitimate was to keep the diagnosis hidden, some also chose to isolate themselves socially. However, support from close family and friends, a meaningful and legitimate understanding of the illness, may contribute towards an increased ability to cope and greater social participation.

This thesis presents the following key findings:

- The results show that the traditional biomedical paradigm may still be an obstacle to patients with PNES adopting explanatory models that exceed the psyche and soma dualism.
- The participants' understanding of their disease condition was characterized by a dualistic and simplified view, with symptoms that cannot be explained by organic findings being perceived as an expression of unilateral psychiatric illness.
- Identification with a psychiatric diagnosis was seen as a process that may threaten the participants identity.
- The perception of the diagnosis as more legitimate and meaningful may enable increased coping

- Being believed that the seizures were not simulated may activate a greater ability for self-reflection, such that it was possible to modify the rigid understanding of disease.
- The study also pointed out some correlation between understanding of the condition and social participation, i.e. those patients who perceived their condition as being comprehensible and legitimate seemed to participate more socially.



## 1.0 Introduction

### 1.1 Pre-understanding – my path into the study field

Throughout almost all my professional life, I have worked with people with epilepsy and psychogenic non-epileptic seizures” (PNES), both clinically as a nurse and later as the head of a Learning and Coping Centre in the Department of Surgery and Neurology at Oslo University Hospital (OUS). My interest has been particularly related to psychiatry and mental health, and therefore, my choice for continuing my education was in mental health. The knowledge I acquired provided new foundations when meeting people with epilepsy and “psychogenic non-epileptic seizures” and also motivated me towards further study in this field. For my Master’s degree at the Department of Health, University of Oslo, I investigated patients’ experiences of receiving a “PNES diagnosis”. Through this programme, communication between healthcare professionals and people with medically unexplained symptoms (MUS) arose as a new field of interest. As a result, I was invited to contribute a chapter to the book “*Klinisk kommunikasjon i praksis*” (“Clinical communication in practice”) (Johnsen K, Otto H Eds.. Universitetsforlaget 2013). This theme was particularly important and motivated me to undertake my own doctoral research studies.

Regarding my pre-understanding, below I discuss three situations from clinical practice.

The first situation is associated with ethical and professional challenges as a result of misdiagnosis of epilepsy as PNES. It can be difficult to distinguish between PNES and epilepsy and therefore it can happen that people who have PNES are sometimes misdiagnosed with epilepsy, and, conversely, that some people receive a “PNES – diagnosis”, but their seizures are actually epileptic. I have often wondered how it must feel when, having had a full examination at the only specialist centre for epilepsy in Norway, a “PNES- diagnosis” is given, along with a referral to the mental healthcare services, when the patient actually has epilepsy.

A girl in her twenties, who we believed to be suffering from PNES, cried and was clearly frightened during her seizures. During her seizures she was carried into another room, because we used to believe that the symptoms of PNES may intensify if the patient receives too much attention during the seizures. There she lay until the seizures were over, while we looked in on her occasionally. Many years later I met her again, and it was then thought that her seizures

were epileptic. Epileptic seizures may cause anxiety, either as a single symptom, or as part of temporal lobe seizures in which the amygdala is involved. It can be difficult to record such seizures with surface EEG because the epileptic area is deep-seated, and may be necessary to use depth electrodes in order to identify the focus and be able to reach a definitive diagnosis. The patient recognized me, and she said “*do you remember that you carried me into the room and left me alone, but my seizures are based in the “anxiety centre”, and that is why I was so scared during the seizures.*” This and similar episodes demonstrate that this is a complicated field, which demands humility, and also underline the importance of listening to the patient’s perspective.

The next situation originates from the Learning and Coping Centre where I held group programmes for young people with PNES. One project that was organised was creating a collage with the theme: “What is it like having PNES?” One group addressed visualising this theme in the collage by cutting out a red carpet, and gluing onto the carpet pictures of people with epilepsy. Down in the ditch beside the carpet, they glued young people with PNES. Then they cut out pictures of doctors and nurses who covered their ears and thought about what they were going to spend their wages on, while their patients desperately tried to get answers to their questions. This depiction touches on themes related to the different diagnoses resulting in unequal status.

The final situation is also a patient history, this one obtained from an outpatient consultation with a patient in her early teens. In the course of a couple of months, she had changed from being a bright, active girl, to a life that included up to 50 psychogenic seizures daily. During our conversation it emerged that what was particularly difficult for her was dealing with the diagnosis. She expressed this as follows: “*It is ok to have seizures, but it is an unfair diagnosis because they say that it is mental*”. From the way that we discussed her condition, it was apparent that the healthcare services had contributed to the largest part of her burden.

These and similar stories inspired a commitment in me to work on how we, as health professionals, understand and conceptualise the illness conditions that lie in the borderland between psyche and soma.

Clinical problems and patient stories such as the three described here have influenced my pre-understanding and been a motivation for this study. They have provided me with guidelines regarding choice of topic and influenced my theoretical approach to the field. Common to all these clinical situations is that they involve a patient group that is marginalized and does not feel understood.

## 1.2 Research context

The study was conducted at and in collaboration with the National Centre for Epilepsy (SSE), which is a department of Oslo University Hospital. Patients with epilepsy-like seizures of uncertain nature may be referred to SSE. Approximately every 5th patient referred to SSE does not have epilepsy, but rather psychogenic non-epileptic seizures (PNES). PNES is the main differential diagnosis of epilepsy in adults. The hospital has the national responsibility for the diagnosis and monitoring of PNES in children and adults. SSE also provides follow-up stays for patients with PNES, either individually or in groups, which include psychoeducational elements.

## 1.3 The purpose and approaches of the study

The overall scientific aim of the study was to develop descriptions of patients' experiences with a "PNES diagnosis", with particular focus on patients' illness perception and understanding of themselves. One intention of the study was to contribute qualitative perspectives to "PNES research". The study was grounded in a clinical context. First, we wanted to explore the patients' experiences of having their diagnosis altered from epilepsy to PNES. In addition, we wanted to investigate the value of being informed of the diagnosis using a biopsychosocial model. A biopsychosocial approach was used to inform patients of the diagnosis in sub-study II, but the intention was not to *evaluate* the biopsychosocial model. Through interviewing patients who had participated in a "follow-up stay," in which their diagnosis had been explained using this approach, we wanted also to gain insight into the impact such an approach could have on patients' illness perceptions. In sub-study III we examined patients' experiences of managing legitimacy in everyday life.

It was our assumption that research-based knowledge about how patients with PNES understand their diagnosis and their symptoms could have some implications for a better focused and more appropriate health care provision for this patient group. The research has been conducted from the point of view of a health professional. The target audience for our work is primarily healthcare providers. This has influenced the use of terminology, choice of journals for publication, and how the information is presented in the various articles. The research is concerned with a marginalized group, and there is an intention that the results may have implications for changes in practice (1). The study has therefore used a pragmatic approach, in which the clinical utility of the studies' results is emphasized (2).

Although theory is relatively confined in the three articles, in this thesis the theory behind the work is described in greater detail. Interpretation of the empirical material has highlighted theoretical themes such as identity, relationships between body and mind, the importance of social interactions, and various disease perspectives.

## 2.0 Literature review

The literature review begins with an introduction to medically unexplained symptoms (MUS) (2.1). This is followed by a section on the diagnosis of psychogenic non-epileptic seizures (PNES) (2.2), where I also give a brief overview of Scandinavian research in this field (2.2.1) and the importance of how the diagnosis is communicated (2.2.2). Finally, there is a chapter on patients' experiences with PNES, and other types of MUS (2.3).

### 2.1 Medically unexplained symptoms (MUS)

The condition PNES is usually placed in the category of medically unexplained symptoms (MUS) (3). People suffering from PNES often also have, in addition to the epilepsy-like seizures, other medically unexplained conditions, such as irritable bowel syndrome (IBS), fibromyalgia (FM), or chronic pain (4,5). Patients with PNES and MUS describe many of the same challenges that are associated with not having a specific somatic diagnosis (6,7, 8).

In cases of MUS, the somatic symptoms with which the patients present cannot be explained by organic findings. In addition to MUS, a variety of different terms have been used to describe such conditions, such as: abnormal illness behaviour, somatization, and somatoform or functional disorders (9). Currently, the most commonly used term is MUS or MUPS (medically unexplained physical symptoms). In my opinion, both these terms are neutral with respect to explain causal relationships behind the symptoms. They show, for example, that there is not to a simple cause-effect understanding regarding how physical symptoms may be thought to be a direct result of emotional or mental problems. Furthermore, use of the word "unexplained" provides an indication that a better explanation of the condition may be obtained when we have achieved a better understanding of the underlying mechanisms.

MUS is not a precise term. It is an umbrella term, and there is no consensus in the literature regarding which conditions should be included under the term. Nevertheless, it is generally agreed that this term should be used to describe conditions when the symptoms cannot be explained by objective findings (10) or as part of a well-defined disease (11). Conditions that

often fall under the MUS term include FM, chronic back pain, IBS, chronic fatigue syndrome (CFS / ME), and asthenia (12).

MUS is common in the general population, in both primary and secondary care (13). The prevalence in primary care varies between different studies, ranging from between 1.1 % and 33 % (14,15). A systematic review showed that between 15 % and 19 % of consultations with a General Practitioner (GP) were concerned with patients with MUS (10). Among new referrals of patients at a neurological outpatient clinic in Scotland (n = 1444), it was found that symptoms were categorized as partially or completely unexplained in 30 % of patients, and that 18 % of these patients had conversion-symptoms, including PNES (16, 17).

Epidemiological studies of MUS among children and adolescents have provided very different prevalence rates. A systematic review showed that between 10 % and 30 % of children and adolescents reported recurrent functional somatic symptoms (18).

The symptoms experienced in MUS vary in intensity and duration from mild and transient to severe and persistent (3, 13), although symptoms are most frequently transient (9, 19-21). The symptoms of MUS are generally less severe and more often transient in patients in primary care than in those referred to specialists (13). A Norwegian study (n = 17,688) demonstrated that of those who visited their GP with MUS, only 3 % had symptoms that lasted for over three months (12). In secondary care, however, it appears as though as many as 40 % of patients with MUS are still experiencing symptoms after one year (13, 22), and this proportion is believed to be even higher among neurological patients (23).

The evidence indicates that the causal associations with MUS are multifactorial (3). It is likely that these factors are best classified as predisposing, precipitating, and maintaining-containing factors (ibid). Genetic factors, chronic and acute physical illness, psychosocial stressors, anxiety, and health concerns are all examples of factors that may play a role in the development of MUS (3).

MUS is a condition that is associated with considerable financial burden on the public purse (17). This is due to the high requirements for healthcare services and also due to considerable sick leave (24).

It must be emphasized that people with MUS comprise a very heterogeneous group. Patients have different medical histories, different symptom patterns, different symptom pressures, use different coping strategies, and have different vulnerabilities. The approach to addressing MUS must therefore be individualized; that is, tailored to the individual patient. The biggest challenges of MUS are associated with the group of patients in whom the symptoms persist over time. Experience shows that many of these patients do not receive adequate help.

## 2.2 Psychogenic non-epileptic seizures (PNES)

### **Terminology**

Epilepsy-like seizures that are not actually epilepsy have been given many names over the years (25), and there is currently no international consensus on what they should be called. It has been difficult to find a term that covers the condition, and at the same time not act as stigmatizing. The term used should not arouse a negative emotional reaction in the patients, so that it does not become an obstacle to coping with the seizures. Simultaneously, the terminology used should stimulate honest dialogue concerning the condition (26).

The current literature uses several different terms to describe such seizures: Psychogenic non-epileptic seizures (PNES), Psychogenic seizures (PS), Non-epileptic Seizures (NES), Pseudo-seizures, Non-Epileptic Attack Disorder (NEAD), Functional seizures, Stress-related seizures (SRS), Conversion seizures and Hysterical seizures.

The terms “pseudo-epileptic seizures” or “pseudo-seizures” are unfortunate because they arouse the suspicion that the attacks are false or have been staged by the patient (25). The term “conversion seizures” was proposed previously (27), but this term is not adequate because there is no agreement according to whether *all* PNES are part of a conversion disorder (25, 28).

In the field of today, most prefer to use the term PNES, and this is also the case in Scandinavia (29). A disadvantage of this terminology is that use of the word "psychogenic" can be problematic, because it implies that the state is generated by psychological or psychosocial reasons. The term therefore reflects a somewhat reductionist understanding, which is not supported by recent research that demonstrates that PNES can best be explained as an interaction between biological and biographical (life experience) factors (30). As with the concept of pseudo-epileptic seizures, the term PNES might also suggest that the seizures are false or simulated.

Studies in which patients and their families have been asked about their preferred terminology have indicated that the terms "non-epileptic seizures", "functional seizures", "non-epileptic attack disorder" (31), or "stress-related seizures" were considered acceptable designations (26). In contrast, the terms "hysterical seizures" and "psychogenic seizures" were perceived as offensive (31).

The term "stress-related seizures" has the advantage that it is relatively value-neutral, as stress has fewer negative associations than psychogenic associations, and this term was suggested as being the optimal choice in a review article (25). However, one objection to this terminology is that stress does not cover those cases in which the basis of the seizures is severe psychopathology.

In this thesis, I have used the term PNES in sub-study I, but switched to NES in sub-studies II and III. The word "psychogenic" was removed, because the results of sub-study I demonstrated that using the word "psychogenic" could provide an obstacle to coping with the condition. In some patients the word psychogenic provoked some resistance and seemed intimidating. I therefore chose to use the term NES instead, because this is more neutral. The term NES is also used relatively frequently in articles that have studied patients' experiences with these seizures (6, 32 -34).

However, the term NES also has weaknesses. It is a very broad, non-specific term and covers all types of epilepsy-like seizures that are not associated with epilepsy, including not only PNES, but also, for example, convulsive syncope, withdrawal seizures, and hyperventilation seizures.



Despite choosing to change the acronym used in the articles during the course of this research project, throughout this thesis I have decided to use the term PNES. The reason for this decision is that the basis for this study is investigating the patients' experiences of being informed about being diagnosed with PNES.

The lack of consensus regarding the terminology that should be used to refer to this type of seizure, probably reflects partly our lack of knowledge and partly the problems in identifying a conceptual framework that accommodates the interaction between psyche and soma.

### **Definition and occurrence**

PNES can resemble all types of epileptic seizures, from small absences to sizeable seizures (28, 30). However, the seizures are not accompanied by epileptic activity in the brain. Reuber has suggested the following definition: "PNES are episodes of altered movement, sensation, or experience resembling epileptic seizures, but are not associated with ictal electrical discharges in the brain. They are a behavioural response to mental, physical, or social distress characterized by a temporary loss of control" (35).

PNES is not listed as a separate diagnosis in the international medical classification systems ICD-10 (36) and DSM-V (37). Such seizures should be considered as a symptom, rather than a separate disease (38) and therefore quotation marks are used when the expression "PNES diagnosis" is used. In ICD-10 PNES is usually categorized under F44.0 (dissociative and conversion disorders), or under F44.5 (conversion disorder with seizures or convulsions). Sometimes R56 (convulsion, not elsewhere classified) or R56.8 (other and unspecified convulsions) are used, particularly if there is uncertainty about the nature of the seizures and there are no indications that the patient has emotional difficulties. In DSM V, PNES can be categorized under Conversions disorder (Functional Neurological Symptoms disorder) (F 44-5) with Attacks or seizures.

The incidence of PNES in adults has been reported to be between 1.4 and 4.6 per 100,000 per year in different populations (39, 40). Because EEG registration is a diagnostic requirement in such studies, and may not have been obtained in all the potential cases, the true incidence is

probably higher. The exact prevalence of PNES in a population is unknown, but has been estimated to be somewhere between 2 and 33 per 100 000 (40). Approximately 20 % of patients who are referred to epilepsy centres for intractable epilepsy are found to have PNES (40).

PNES occur among both sexes and at almost all ages, but is most commonly reported in young women (41-44). Among adolescents and adults with PNES between 70 to 80 % are female (45-47). PNES occur less frequently among children than in adults, but the condition is probably under-diagnosed in children (48). Although exact prevalence data on PNES among children are lacking (49) a study of children using long-term EEG (video telemetry) found that 11-15 % had PNES (50) 2002) and between 10-23 % of children admitted to epilepsy centres are diagnosed with PNES (25, 51).

## **Diagnostics**

PNES is the most common differential diagnosis among adults with suspected epilepsy (52). Unfortunately, there are no measurable physiological markers that can confirm or refute a “PNES- diagnosis”. The diagnostic procedures can be both time consuming and difficult, and misdiagnosis is common (53), including among children (54). Although there are several clinical features that strengthen or weaken the suspicion that the attacks are non-epileptic in nature, it is difficult to rely fully on seizure characteristics only (semiology) for reaching a diagnosis (28, 55).

A thorough medical history of concomitant information, with particular emphasis on seizure form and duration, the situations in which seizures occur, and a complete psychosocial assessment, could provide information to strengthen the suspicion of PNES. Such suspicions would be further strengthened if it is shown that the seizures are not accompanied by epileptiform activity when investigated by EEG using video telemetry (52). Furthermore, patients’ seizures are not always captured successfully during EEG recording. Thus, in some cases many years may pass before the diagnosis is made. Many patients are initially diagnosed with epilepsy and are prescribed antiepileptic drugs for several years on the basis of a misdiagnosis (42). A further complication to the diagnosis is that between 10-30 % of epileptic patients also have PNES (40).

## **Predisposing factors**

Patients with PNES form a very heterogeneous group (56) regarding seizure type, duration, frequency, and underlying causes. Division of PNES patients into sub-groups has been proposed (57).

Among patients with PNES, a higher proportion with epilepsy is found among those with learning disabilities than among cognitively well-functioning patients (58).

The aetiology is often complex (35). A Norwegian study compared the patients with PNES (n = 23) with patients with a somatoform disorder (SD) (n = 23) and with healthy controls (59).

Among patients with PNES, 21/23 had comorbid psychiatric diagnoses, whereas the equivalent proportion among those with SD was 18/23. The average number of comorbid psychiatric diagnoses was greatest in patients with PNES. Higher levels of anxiety, depression, and anger were identified in both patient groups compared with the healthy controls, and those with PNES also scored significantly higher with respect to hostility (59).

In recent years, relatively substantial evidence has been published that PNES can be best understood in the light of a biopsychosocial model (30, 35, 56, 60, 61). In such a model predisposing, precipitating, and maintaining factors are presumed to play important roles (35, 60, 62). Predisposing factors could be traumatic experiences, such as, for example, previous sexual abuse. Triggers (precipitating factors) could be traumatic events and might include bullying, anxiety, or dysfunctional family relationships (ibid). Some studies have shown that PNES can occur when coping strategies are inadequate (63, 64). Patients with PNES have been found to have elevated basal cortisol levels compared with controls (30, 65) which are indicative of higher stress levels.

Some evidence also suggests that neurobiological factors may predispose individual to PNES, however the studies are small and must be interpreted with caution (30, 66). Among other things, an increased incidence of pathological findings on cerebral MRI (Magnetic Resonance Imaging) and non-specific findings on EEG have been reported from patients with PNES (67-69). It has also been shown that PNES may occur after minor head injuries

In children with PNES it has been found that difficult life events associated with increased stress levels may precede seizure onset (38). Bullying, dysfunctional family relationships, inter-

personal conflicts, and social stress have been reported from among children and adolescents with PNES (29, 38, 51, 70). There is also an increased incidence of depression and anxiety amongst children and adolescents with PNES (38, 51, 69). A controlled study in which 55 children with PNES were compared with their siblings showed that the children with PNES had more neurological (including epilepsy), medical, and psychiatric morbidity than their non-PNES siblings (47). The PNES children also had more difficulties at school, were more likely to be bullied, and had greater interpersonal problems (47). In another study it was found that 18 % of children with PNES had been exposed to physical or psychological abuse, and that almost 25 % of them had significant mental health problems (38).

### **Prognosis**

The impact of treatment, and thus the prognosis, for patients with PNES has been explored in only a few studies. Nevertheless, it appears that the prognosis is significantly better in children than in adults (38, 71, 72). It has been estimated that approximately 70 % of children and adolescents become seizure-free after appropriate treatment (38, 72 -74) whereas a study of adults with PNES (N = 164) showed that 71 % still suffered from seizures four years after the diagnosis had been made and 11 years after seizure onset (75). These results support those of other studies that have investigated seizure development in adults (39). Early intervention and correct treatment are important in order to avoid the condition becoming chronic and with increasing psychopathology (5, 25, 38). One possible reason for a better prognosis in children than adults is that it is often easier to identify underlying or associated causes of PNES in children, such as problems at school or in the family. Thus, it is also easier to provide appropriate interventions (72).

When considering the prognosis for people with PNES, focus should not only be directed on the seizures, but also on psychological difficulties and quality of life (52, 76). Many patients with PNES drop out of school and lose their social networks. In a study of adults with PNES, nearly 50 % of those who became seizure-free did not return to work (77). Another study found that during follow-up of adults with PNES over 5-10 years, less than 23 % were in paid work (78).

## **Treatment**

The heterogeneity regarding the underlying causes of the seizures in PNES means that treatment should be adapted to the individual (28, 63, 79, 80). There is currently general agreement that all patients should be offered psychoeducation at the time of being informed of the diagnosis (52, 81). It is also recommended that patients and their families should be given some explanations about the possible mechanisms involved in dissociation (52, 55, 60, 82).

For patients that have neither experienced severe trauma nor have psychiatric comorbidity, a good, empathetic method of informing them about the diagnosis (83), together with a brief cognitive intervention focusing on stress management and "empowerment" can be sufficient (80).

Traumatized patients, however, and patients with clear associative difficulties and psychiatric comorbidity, usually need a more prolonged treatment in a safe environment with time to develop a positive relationship with the therapist (80). If dysfunctional family relationships are a triggering and/or sustaining factor, the whole family should be involved in the treatment (60). It is also important to identify and treat comorbid psychiatric disorders (84). Untreated depression and anxiety have been shown to be associated with persistent seizures (53). Because some patients with PNES try to avoid situations associated with unpleasant and overwhelming feelings (85, 86) therapy that is directed towards working on emotions can have a beneficial effect (85). Treatment of PNES also involves use of anti-epileptic drugs being gradually reduced, right down to full withdrawal (52).

Cognitive behavioural therapy (CBT) is the treatment method that has been tested the most (60). Although a few individual studies have shown a positive effect of CBT (87), psychodynamic psychotherapy (88), and brief psychodynamic interpersonal therapy (89), a recent Cochrane review demonstrated that there is little evidence to recommend a single treatment above another (90). In general, there is a paucity of randomized controlled trials in this field.

Nevertheless, there appears to be consensus that the PNES condition requires a comprehensive approach to treatment and close cooperation between neurology and psychiatry (52, 53, 91).

However, what commonly occurs in practice is that the diagnosis is made by neurologists, whilst the treatment is usually a psychiatric concern (52, 53)

This means that, usually, when the diagnosis has been made, follow-up in neurology ceases, and the patients are referred to a local psychiatric unit. Unfortunately, it can sometimes be difficult to reach a common understanding of the condition (92, 93) and some patients find that they fall between two chairs (53). Reaching a common understanding of the diagnosis might also be challenging, and the lack of evidence-based treatment programmes across neurological and psychiatric disciplines makes the challenges even greater (94).

Although the focus and research on PNES have both increased significantly over the past 20 years, many unanswered questions still remain. In short, it must be admitted that the mechanisms behind PNES continue to be poorly understood. There is therefore an urgent need for large, multicentre studies with the goal of improving the diagnosis, recognising causal relationships, and identifying the most effective treatments.

### *2.2.1 Scandinavian studies of PNES*

There are few Scandinavian studies in this field, and, to my knowledge, none have been published in which qualitative methods have been used. With respect to treatment of PNES, a Norwegian clinical review article has recently been published (52), in which the need for training and informing patients of their diagnosis in a respectful and empathetic manner was emphasized. Likewise, closer collaboration between psychiatry and neurology was identified as a requirement. In another Norwegian article on differential diagnoses of epilepsy, the importance of understanding the interaction between somatic, autonomic, and emotional aspects of non-epileptic seizures, including PNES, was highlighted (55). Education about the underlying mechanisms of these conditions was considered to be essential for the patients being able to cope (55). In a third Norwegian article, it was noted that a wide range of psychological conflicts or traumas could be the basis for PNES, and thus individual treatment measures are necessary (28).

A further Scandinavian study reported an increased incidence of psychiatric comorbidity, in terms of anxiety, depression, and aggression, in patients with PNES (59).

A recent Danish survey of doctors showed that, of those included in the study, only 49 % used video EEG as a diagnostic tool for suspected PNES (29). The authors conclude that there is a need to improve and systematize the diagnosis of PNES in children. The study also showed a lack of consensus regarding the diagnostic codes and terminology that were used by the doctors (29).

Two other Danish studies investigated the use of an automated algorithm based on surface EMG (electromyography) to distinguish PNES from epileptic seizures. The authors concluded that the algorithm was a good tool to distinguish between the two seizure types (95, 96)

A fourth Danish study reported that 30 % of children who had been referred to a tertiary care service because of a difficult seizure situation did not have epilepsy, despite there being no expression of doubt regarding the epilepsy diagnosis in the referral papers. The authors suggest that misdiagnosis was common, and urged colleagues to refer difficult cases for further diagnostic review in the tertiary care services to avoid incorrect medication with anti-epileptic drugs (AEDs) (54).

The authors of a Swedish review article conclude that a patient's acceptance of the diagnosis and good patient information are crucial for achieving good results (82). Another Swedish study found that people with PNES used AEDs for shorter periods than people with epilepsy, but that they used several types of AED (97).

In summary, this overview indicates that most Scandinavian articles about PNES are review articles that are published in national medical journals. Such reviews are considered important as experience indicates that GPs in particular, but also specialists in paediatrics, neurology, and psychiatry, may be unsure how this patient group should be managed.

Qualitative Scandinavian studies that aim at exploring the patients' perspectives are completely absent. Given that we know that a patient's own understanding of the condition is important for

optimizing the individual approach to treatment in these patients, this lack of studies is disappointing.

### *2.2.2 Communicating the “PNES diagnosis” and health professionals' understanding of PNES*

A few decades ago some articles started to describe how patients reacted negatively to the “PNES diagnoses”. Clinical experience showed how communicating a “PNES diagnosis” to patients may elicit aggression and denial, and may even increase the risk of suicide (41, 98). More recently, this topic has become even more relevant because it has become apparent that the way in which the diagnosis is communicated may affect whether the patients accept the diagnosis or not, and thus also their motivation for obtaining treatment and their prognosis (76, 93, 99, 100, 101).

In the book *Pseudo-Epileptic Seizures* (102), which was written at the initiative of the International League Against Epilepsy, an overview of how the field looked at PNES at that time (1993) was presented. Amongst other information, an introduction to contemporary views on how the diagnosis should be communicated was provided. It was believed then that the treatment team would achieve the best results if the diagnosis was presented to the patient as an indisputable truth, even if some people within the team had some doubts about the diagnosis (102)(p. 79). If the patient refused to accept the diagnosis or reacted negatively in any way, this was interpreted as a confirmation that the diagnosis was correct. The rationale for this interpretation was because protest was considered to be a natural defence if the seizures were an expression of repressed conflict or trauma (103) in (102).

In the same book, some Norwegian authors write (104) in (102) that the multidisciplinary team should not have a fixed position on the diagnosis, as there will always be some risk of misdiagnosis. Moreover, efforts should be made to avoid the patients being intimidated by the diagnosis; the importance of respect for the patients' own understanding of the condition is highlighted (p. 139). This attitude is supported by several articles about PNES that were published later. Several authors argue that one should be honest and inform the patient if there is any doubt about the diagnosis (93, 105).



In recent years several standardized manuals on how to communicate the “PNES diagnosis” have been published (106 -108). In these publications it is recommended that considerable time be used helping the patient and family to accept that the seizures are not epileptic, and ensuring that the patient receives help in addressing any shame and stigma (108). The different attitudes described above illustrate how the approaches, understanding, and attitudes of health personnel are modified with increased knowledge. In my opinion, this shows there has been a development in this field, in which we have moved away from a paternalistic attitude towards a more humble approach, in which we increasingly attempt to incorporate the patient’s own perspective. History shows that a diagnosis of PNES brings about both professional and ethical challenges for health professionals.

A prolonged and ongoing debate is concerned with whether one should tell the patients that their seizures can be controlled by a conscious voluntary effort. During the 1990s several authors were proponents of this approach (109) Cited in:(102) (p76).

Ten years later, arguments are now presented *against* such an attitude (93). The author gives three reasons:

1. The author did not believe that the patients themselves had control over the seizures (except for a few individuals who simulated seizures).
2. The patient and family could easily interpret this as the patient being understood to be simulating the seizures, and this could result in resistance or lack of acceptance of the diagnosis.
3. Control of seizures was usually first achieved through treatment.

In 2014, a study that investigated patients’ levels of consciousness during PNES seizures was published (Roberts 2014), and concluded that the level of awareness varied between patients. Today, most researchers seem to agree that the majority of PNES are beyond patients’ willing control (110). Nevertheless, it can appear that some health personnel who are working clinically with this population believe that patients with PNES have more control over the attacks than they actually do (111). A qualitative study from England based on interviews of 22 neurologists showed that the neurologists had an unclear view of the relationship between "game" and conversion (112).

Although many clinicians do feel empathy with this group of patients, many also recognize that they may have stigmatizing attitudes. This applies especially to cases where they believe patients 'act' seizures, or if the attacks come at very convenient times (94). It should be emphasised that there are also examples of health personnel being an important resource for patients with PNES (6, 113).

PNES continues to be poorly understood by both laymen and professionals (38). In the years ahead, knowledge of PNES should be strengthened among health personnel. How the diagnosis is communicated is essential for achieving a common understanding of the seizures, and important, not least, for attaining an alliance with the patients. Communication of the diagnosis is thus a critical point, and there is no doubt that more research is needed in this field (91).

### 2.3. Patients' experiences with MUS and the “PNES- diagnosis”

#### **Patients' experiences with MUS**

There is rather much literature on patients' experiences with MUS. I only present certain aspects that are particularly relevant to this study. Patients with MUS meet several challenges. One challenge is that they experience a lack of acceptance of the symptoms or the condition (6-8, 114-118). Young people with CFS may describe finding it difficult to explain their condition, and that they struggle because their condition is controversial (114, 119 -122). Being mistrusted by family and friends can be perceived as personal rejection (116).

One consequence of MUS can be that the patients live restricted lives. Many become socially isolated, they become less active, and several miss out on their education and thus on employment opportunities (7, 123).

People with MUS also describe communication with the clinician as being difficult. This may be because their frames of reference are not the same. Whereas patients are concerned about their subjective symptoms, doctors may be more interested in the results of tests and investigations (124). This can easily result in a patient feeling that he/she have been poorly treated and misunderstood by the healthcare system (123, 125). It may appear as though doctors in the primary care facilities have a tendency towards referring their patients to somatic

examinations against their patients' wishes. One possible explanation may be that the doctor is trying to avoid addressing patients' psychosocial problems (124).

It is also problematic for patients with MUS that the healthcare services are lacking in knowledge about their conditions. It has been said by some that this lack of knowledge about the suffering is experienced as a greater burden than the symptoms *per se* (125). Patients with MUS can find it threatening when therapists refer to psychosomatic explanations. This may be due to a fear of being labelled as mentally ill, marginalized, or that they "play at being ill" (117, 126).

### **Patients' experiences with PNES**

Although patients with MUS have some common traits, conditions may be considered to differ depending on the pattern of symptoms. There are also aspects of PNES that mean that the patients' experiences differ somewhat from those of patients with other types of MUS. People with PNES generally experience that the seizures may occur in any social setting. Although it is possible to conceal a diagnosis, seizures are more difficult to hide. This contrasts with patients with only subjective MUS that are not visible to others (127). As the "PNES diagnosis" provides an immediate link to psychiatry or psychosomatics, via the term "psychogenic", this also provides parameters regarding how patients interpret their condition.

How a patient reacts to receiving a "PNES diagnosis" varies greatly from individual to individual. Many react with denial, astonishment, anger, or confusion (33, 34,108).

A study from Ireland (33) demonstrates some of the difficulties in being informed of a "PNES-diagnosis". The study was based on telephone interviews of 84 patients between 1 and 7 years after they had received a "PNES-diagnosis", and showed that only a third of them had some understanding of diagnosis. Even among those who had some understanding, there was still considerable confusion regarding the causes of seizures, and 63 % felt unsure about how they could get better. Although 65 % of the interviewees had received psychological intervention, the average period of treatment with a psychologist was only 2 hours per patient. The authors concluded that the health information and psychological support provided to the patients was far

from satisfactory. It was recommended that the patients' own understanding and reaction to the diagnosis should be the basis for developing a more appropriate treatment programme (33).

Based on those studies that demonstrate the difficulties experienced by people receiving a "PNES diagnosis", some recent research has looked more closely at those factors that result in these difficulties. It appears as though it is particularly difficult for patients to identify causal explanations that are understandable and meaningful.

A study of nine adult outpatients with PNES concluded that it was often challenging for clinicians to help patients to understand their condition in light of more scientific terms (34). Patients had a tendency to perceive the condition as dualistic; that is either physical or psychological. On the one hand, patients were looking for biomedical explanations of their symptoms, but, at the same time, their understanding of the "PNES- diagnosis" was that it was psychiatric (34).

Communication between clinicians and patients with PNES is difficult, as with communication regarding MUS in general. Monzoni and colleagues used conversation analysis to investigate the challenges in communication between neurologists and their patients with regard to functional symptoms (n = 20, 17 with PNES). All the patients expressed resistance when their doctors provided psychological explanations and suggested that treatment should be in mental healthcare (128). Indeed, the patients' resistance could be so great that it became pointless for the doctors to mention mental healthcare (129).

Other studies investigated the patients' experiences with the diagnosis from the aspect of how the diagnosis was communicated. Thompson and colleagues developed a stepwise procedure to be used by neurologists when communicating the diagnosis to patients (108). The results showed that the participants in the study felt stigmatized. They felt ashamed and found it difficult to receive a diagnosis that was perceived as "only mental". The diagnosis indicated to them that the seizures were self-inflicted or that they were mad. It is clear that health professionals need to spend time working on patients' prejudices against psychiatric illnesses. Some patients refused to be referred to a psychiatrist (108).

In another study, a brief (four session) psycho-educational intervention was carried out subsequent to the diagnosis. The authors argued that the method that they used to convey that the aetiology of the seizures was psychological was both effective and acceptable. In evaluating this study it should be taken into account that the 50 patients enrolled were only followed for 3 months (107).

In a subsequent study, Baxter and colleagues (81) interviewed 12 of the same 50 patients who had received psycho-educational intervention by Hall Path 2010. (107). The aim of the study was to provide insights into the participant's perceptions following the intervention. The authors report that psycho-education can help patients to see a logical association between emotional stress and seizures. Six of the 12 participants had increased their general understanding of the relationship between seizures and life experiences. Nevertheless, the study showed no clear link between improved understanding and acceptance of the "diagnosis" and a tendency towards fewer seizures (81).

It is obvious that it can be difficult for patients to accept a "PNES diagnosis". The explanatory models that have been used have not been optimal. Thus, the question arises of how we can best help our patients to recognise the associations between biography and seizures during the initial phase while patients are still hospitalized. It may appear as though a salient point is related to the patients being helped to understand relationships in their own lives.

Two recent studies have shown that those who integrate *personal* stories into their understanding of their condition demonstrate greater coping ability and acceptance of the diagnosis (6, 32). Thompson and colleagues (6) used interviews and interpretative phenomenological analyses to explore the understanding of the illness with eight adult patients who were on the waiting list for treatment of PNES. Although the patients described isolation, helplessness, shame, low self-esteem, and a feeling of being abandoned in "no man's land", those patients who understood the cause of the seizures, showed increased coping abilities. Those patients who were aware of past trauma were able to understand that the diagnosis made sense and were motivated towards obtaining treatment (6). However, those patients who did not remember having been subjected to trauma, were afraid that they had experienced trauma that they had now forgotten.

The work of Dickinson and colleagues was also based on interviews and involved five adult patients who had received the “PNES diagnosis”. Those patients who incorporated a psychosocial explanatory model into their understanding of their condition experienced that the diagnosis made sense and they had greater motivation for treatment (32). The authors emphasize the importance of exploring the reasoning that is used by people with PNES when they are reflecting on their illness.

In summary, it appears that individuals with MUS experience significant challenges as a direct result of the imprecise definitions of their condition. There is a lack of studies on how social interactions affect patients’ daily lives. This is a key area, as social interactions can have an impact on how patients perceive their illness (130).

Regarding MUS in adolescents, some studies have investigated how young people understand their condition and manage their symptoms (114, 119-122). MUS in children and adolescents has been the subject of few research studies (131). This is unfortunate because such conditions should be identified early in order to avoid them becoming chronic (121, 132). Studies investigating relational aspects in adolescents with MUS are particularly lacking (121).

In brief, it appears that patients diagnosed with PNES often struggle to understand the diagnosis. There are very few qualitative studies on PNES (56, 133), and those that exist are largely directed toward adults. While some studies have investigated interactions between patients and treatments (128, 129) others have had greater focus on the patients’ experiences with the diagnosis (6, 32, 81).

As previously stated, qualitative studies of PNES in young people are lacking (6). I have been unable to identify any studies that have been specifically aimed at the importance of social interactions in everyday life in patients with PNES, whether adults or adolescents and children.

Based on my review of the literature above, I wanted to examine patients’ experiences with the diagnosis. When we planned the first study (sub-study I), the evidence regarding patients’ experiences with the diagnosis was largely unexplored. We therefore chose a relatively open approach and examined how patients whose diagnosis had been changed from epilepsy to PNES

experienced being informed of their new diagnosis. The results of our first study, along with other research that had been performed, demonstrated that the explanatory models presented provided little sense. Based on this, we opted for a more targeted approach in the second study (sub-study II). In this second study, we investigated the impact for the patients of using a biopsychosocial model to communicate the diagnosis. Whereas the two first studies had the patients' experiences with healthcare as their main point of focus, the third study concentrated on the participants' social interactions in everyday life. This field had been the subject of very few previous investigations. Furthermore, as sub-study II had also demonstrated the patients felt delegitimized when confronted with a contested diagnosis, sub-study III had a particular focus on delegitimization.

On the basis of these three studies, I had the hope that by increasing our knowledge of the patients' own experiences, the foundations could be laid for putting in place a more optimised "treatment plan" for this patient population, which has often been "given the cold shoulder" treatment by the healthcare services.





### **3.0 Objectives and research questions**

- 1) What are the experiences regarding their condition of patients whose diagnosis of epilepsy is changed to a diagnosis of PNES?
  - a) Which aspects of being diagnosed with PNES were particularly difficult for the patients?
  - b) To what extent did the patients feel that their needs were met by the Norwegian National Health Service?
  - c) Which factors were associated with a successful outcome?
- 2) What is the impact of using a biopsychosocial approach for communicating the diagnosis to adolescents with NES?
- 3) How do the young people with NES describe their social participation in everyday life?



## **4.0 Design, materials and methods**

This chapter describes and explains the methodological choices used in the PhD project. Qualitative research methods were used to investigate the problems described. Qualitative methods are well suited to exploring human experiences, expectations, motives, attitudes, and practices (134).

### **The study's scientific basis and analyses**

Patients' experiences with the healthcare services, and their experiences with the diagnosis of PNES, cannot be considered as unambiguous observable facts. Patients' experiences will be affected by various factors, including the cultural and historical situations of the patients. Thus, in investigating patients' experiences, we rely upon an interpretative paradigm in which the study's empirical results are interpreted using various theories and philosophies (135).

The knowledge derived from the study should thus not be considered as an established truth, but as constructed consequentially from an interpretive argument. Contextual conditions have affected all stages of the research process (136). The choices made during the study, including the choice of theory, have influenced the study results (137). Knowledge results from an interaction between empirical data, analysis, and theory (2).

Elements of both phenomenological and hermeneutical philosophies have been used in this research. According to Malterud (135) there is often a relationship between phenomenology (lived experiences) and hermeneutics (the interplay between parts and whole) in most qualitative studies (135). Descriptions and interpretation often go hand-in-hand (134).

Phenomenology represents an understanding in which human experiences are considered valid information (134). We wanted to explore how the participants made sense of their experiences with PNES. This means that both the unique individual opinion of each participant and the commonality in the participants' experiences were considered as valid information sources (138). In order to accommodate this approach, we made a sustained effort to bracket our own preconceptions during the initial steps of the analyses. This is a methodical concept that is based on phenomenological understanding (134).

In order to develop a broader sense of study participants' statements, we used a hermeneutical or interpretive approach in the subsequent analytical steps. Hermeneutics is concerned with the interpretation of texts (2). In interpreting the data acquired during this research, we have asked ourselves how the participants' descriptions can best be understood and what is at stake. In attempting to answer these questions, we have moved back and forth between the empirical data and different theoretical perspectives. We considered various theoretical approaches and chose those that we believed could best elaborate the central categories of analysis in the data (138).

Although various theories were considered in the analysis and elaboration of the findings, it is those theories that were first and foremost employed that are described in this thesis. Theories on "different perspectives of disease" and "psyche / soma" dualism were found to be of particular relevance because of their consequences regarding how the patients' cope with their condition. By using theory concerning identity, we hoped to obtain a better understanding of what can be at stake for patients when they are diagnosed with PNES. Our intention of using such theories during this research has not been to develop new theories nor to basically challenge existing ones, but to develop knowledge that is of such a nature, that it has the potential to improve clinical practice (135).

#### 4.1 Data collection methods

Both qualitative interviews and participant observation (fieldwork) were used to collect data. The empirical material consists of transcribed text from 21 semi-structured interviews with 21 participants. In Study I, there were 10 participants (henceforth referred to as Group 1) (see Figure 1), and there were 11 participants in sub-study II and III (henceforth referred to as Group 2) (see Figure 2). In addition, the material consisted of notes from a smaller follow-up study involving follow-up calls and home visits (sub-study III) and two follow-up studies by phone (sub-study I and sub-study II). The analytic strategies used are exploratory and interpretative. The data has been analysed by using systematic text condensation (STC), based on the Malterud Model (139, 140) (sub-studies I and II), and thematic analysis (TA) (137) (sub-study III).

## **Interviews**

Qualitative research interviews were chosen as the primary means of data collection, because qualitative interviews are well suited to obtaining information that will assist in understanding the world from the interviewee's perspective (2). In this study, it was important to emphasize the importance that the patients themselves attributed to being informed of the "PNES diagnosis". Our intention was to obtain data that was as rich and diverse as possible (134).

The format of interviews may vary from using structured, established questions, to completely unstructured and free-ranging discussions (141). We chose an open-ended, and thus semi-structured approach to the interviews. Due to our considerable prior knowledge about the topic, this characterised the interview guide. However, at the same time, we wanted the participants themselves to have the opportunity to shape the content. By following a thematic guide, we ensured that whilst we were able to obtain those data that were of relevance to the issue, the interviewees simultaneously were able to provide their own reflections. Their contributions were generally followed up with new questions (141).

Within the context of this study, the interview process is understood as a route by which knowledge is created actively by the interviewer and interviewee together, through questions and answers (2). Communication between interviewers and interviewees will always be on the basis of their different perspectives, and this also includes the social positioning that emerges in the interview context, in the form of an interaction between interviewer and interviewee / questioner and respondent (142). The context in which the interview takes place will thus affect the content. According to Warren (142), it is more useful to understand the people that are being interviewed as creators of meaning, rather than as purveyors of fixed facts. Thus, the creation of qualitative data is not simply making a passive record, but an interpretive work in which the researcher's preconceptions and perspective (research paradigm) also affect the process. What is most important is not whether the researcher affects the process, but how (136). Therefore, in the following text, I will try to describe the research process - from data collection to final results - as clearly as possible, so that my perspective, my choices, and my reasoning are available to the reader (140).

## **Participant Observation**

As a supplement to the transcribed interviews, in Study III we wanted to conduct some participant observation in the adolescents' home environments (family, school, work, and leisure time). We thought that observation and informal conversations could be better suited to gaining insights into the significance of the social interactions in everyday life on the participants' illness perceptions than by using interviews (2). Furthermore, by using a variety of methodological approaches, we have the opportunity for acquiring different types of data. During an observational approach, the researcher is present and can witness events directly, and this provides the participants with the opportunity to offer further information that may have been missed by other methods (143). Nevertheless, participant observation was not such a large part of the research as we would have liked, because we found that gaining access to the field was problematic. This is discussed in greater detail later in this chapter, under the heading "Collection of Data".

### **4.2 Sampling of study participants**

Staff on the wards at SSE recruited patients consecutively, provided that they met the inclusion and exclusion criteria of the study. Sampling was conducted by staff contacting relevant patients and providing them with verbal and written information about the study (see chapter section "Collection of Data"). Folders for the wards were provided containing: the letter of approval from the hospital's management, a copy of the approval from the Regional Ethics Committee (REK), the topic guide, the project plan (detailing the inclusion and exclusion criteria), guidelines for informing patients of their diagnosis (sub-study II), the names and contact details of the members of the study's reference group, and an informed consent form for distribution at study inclusion. As shown below, it was necessary to modify the inclusion and exclusion criteria used in sub-study II and sub-study III due to recruitment difficulties.

Inclusion and exclusion criteria common for all three studies:

- The patient should be an inpatient at SSE during the empirical phase of the study.
- Both sexes can be included.

- The patient must have sufficient cognitive functioning (as determined by the ward staff) to be able to reflect upon their own situations.

Additional criteria for sub-study I:

- The patient must be over 14 years of age (the lower limit was later raised to 16 years).
- The patient's seizures should have previously been considered to be due to epilepsy, and the patient should use anti-epileptic drugs (AED).
- During the period hospitalization the patient shall be introduced to the "PNES-diagnosis" as an explanatory model of the same seizures that were previously regarded as epileptic. Video-EEG recording of seizures typical for the patient shall not reveal epileptiform discharges.

Additional criteria for sub-studies II and III:

- The patient should be between 13 - 16 years (upper limit later raised to 24 years).
- The patient must have had their diagnosis explained via a biopsychosocial approach, and should have received psychoeducation about PNES, in which PNES is considered in the light of a biopsychosocial understanding (cf. "Description of Guidelines for the "follow-up stay").
- The diagnosis should be based upon typical seizures on telemetry without clinical correlates. (This was changed to: the diagnosis should be determined by a neurologist and the patient should be referred for treatment in mental healthcare).
- The patient's PNES seizures shall be the main problem, although epileptic seizures cannot always be excluded.
- Patients with severe psychopathology, e.g., recognised borderline diagnoses, shall be excluded from the study.

Recruitment to sub-study I took place in one of the adult wards immediately following the patient receiving the diagnosis. Inclusion to the study took seven months. Recruitment to sub-studies II and III took place in both the adult ward and the paediatric ward. Inclusion to sub-studies II and III took 1 year.

One reason for the prolonged inclusion periods in the studies was the fluctuations in admissions. There were significantly fewer admissions during the relevant period in comparison with the previous year. Several potential participants also withdrew immediately before admission because they were having a “good period” regarding seizures or because the teenagers did not want to miss any school attendance. In some cases, the parents wanted their children to be hospitalized, but the young patients themselves did not. In the initial recruitment period, some potential participants were excluded after they were admitted, because the diagnosis was not made based on video EEG monitoring during seizures, because they had epilepsy as well, or because their level of cognitive functioning was not considered sufficient for them to benefit from the biopsychosocial model. Two further potential participants were excluded after the upper age limit was raised to 24 years due to severe psychopathology.

Following new approval by the Regional Ethics Committee, the upper age limit was raised in two stages, first to 18 years, and then to 24 years. An upper age limit of 24 years could, in our opinion, be justified because the experiences of the young people in this context are not necessarily associated with a strictly defined age group. This also provided the opportunity for inclusion of patients from the adult wards. Patients from the adult wards should receive the psychoeducation in groups. However, this also led to delays, as identifying patients who belonged in the same group was not easy.

Eventually, several changes were made in order to speed up the inclusion process. Firstly, it was decided that patients could be included even if their seizures had not been recorded by video EEG. However, this was conditional upon the diagnosis being made by a neurologist and the patient being referred for treatment in mental healthcare (this applied to two of the participants). Next it was decided that patients could be included in the study even if they also suffered from epilepsy. However, the “PNES seizures” should be the main problem, and the seizures should be surveyed by video-telemetry such that the patient was aware of which seizures were epileptic



and which were PNES (this situation applied to one, or possibly two, of the participants). Finally, we also informed other neurological departments in Norway about the study and requested neurologists to identify patients suitable for the “follow-up stay” at SSE. The consequences that these changes may have had on the study’s results are discussed in the “Discussion of Methods” section of this thesis.

#### 4.3 Descriptions of guidelines for the follow-up stay

One of the inclusion criteria for sub-studies II and III was that patients had had their diagnosis explained via a biopsychosocial approach and had received psychoeducation about PNES. In practice, this meant that the patients had to attend a follow-up stay to give them the opportunity to process their diagnosis. In this section the guidelines for the inpatient follow-up stay are described.

Participants were interviewed at the end of the follow-up stay. The purpose of this stay was to increase their understanding of the disorder. The process could be divided into three phases. The *first* phase was to build trust and to get to know the patient. Emphasis was placed on processing previous unproductive encounters with the healthcare system, and recognizing the patient’s perception of the illness. Patients were asked to talk about those factors that they believed might have had significance for the development of seizures.

In the *second* phase of the “follow-up stay” the neurologist and nurse went through the “PNES diagnosis” with the patient and/or patient’s family. Biological, emotional, and social factors that were believed to underlie the diagnosis were then organised as being predisposing, precipitating, or maintaining factors, and written down on a whiteboard or paper. At the same time, information was provided on how different pressures (stresses) may initiate organic processes. The intention of this process was to generalize the condition by demonstrating how life conditions, psyche, and soma interact with each other in the development of all types of illness. The intention was also to try to avoid searching for a specific cause of the seizures, but instead to emphasise that many factors may play a role. Furthermore, an effort was made to avoid dualistic concepts, such as psychogenic and “psychological cause”.

In the *third* phase, the patients were offered follow-up meetings with the neurologist and a nurse. (The guidelines are also described in Section 8.3).

### **Different offers from the different departments**

Six of the seven participants who were recruited from the adult department were offered a longer stay with a broader course of material than for those who were admitted from the paediatric ward. The adult ward implemented a 4-week stay, while the paediatric ward offered a 2-week stay. Patients with the extended stay were also offered:

- 4 group discussions with other patients with PNES.
- Instruction in techniques to halt seizures (breathing techniques) and offers of physiotherapy.
- One hour of teaching with a PowerPoint presentation (see Appendix 5). Key topics included in the presentation were: different causal relationships of PNES, diagnoses, treatment, and bodily reactions to anxiety, stress, and hyperventilation. The participants were provided with a printout of the presentation afterwards.

## 4.4 Collection of data

### **Interview**

During the collection of data for sub-study I, I worked in a clinical capacity in the hospital's paediatric ward, while the participants were recruited from the hospital's adult department. Collection of data for sub-studies II and III took place while I was employed as a research fellow and as the leader at the Learning and Coping Centre at SSE. Thus, I did not work in a clinical capacity with the inpatients.

Two of the participants in sub-study I could not be interviewed. One declined to be interviewed because she had not accepted the diagnosis. The other participant felt mentally unstable and therefore did not have the energy to be interviewed. Two other patients, who had been selected for sub-study I, were also not included. For one of these, this was because the seizures had not previously been regarded as epileptic, and the other because it was not determined that the

seizures were PNES. None of the participants selected for sub-studies II and III declined to participate.

Prior to the interviews, the participants were told that they could terminate their interviews at any time and without needing to provide any justification. In order for the interviews to run smoothly, two different topic guides were developed and studied prior to the interviews. The interviews took place in an office on the ward or in my own office.

The interviews in Participant Group 1 lasted, on average, 50 minutes, and I transcribed the information. The interviews in Participant Group 2 had an average duration of 65 minutes, and were transcribed by office staff at SSE. We aimed to transcribe the text so that it was as close as possible to what was actually said (136). For reasons of anonymity, dialect was not transcribed, and place names and personal names were replaced by alternatives.

As mentioned previously, we endeavoured to ensure that the topic guides were as open as possible such that the interviewees could influence the direction of the interview. The interviewees were encouraged to follow associations and talk freely about what was important to them, even if this did not provide direct answers to the questions. In retrospect, it became clear how anecdotes that might have seemed irrelevant to the focus of this study during the interview, nevertheless gave useful information when the interview transcription was read in its entirety. This was either because such anecdotes provided a backdrop against which other statements could be considered, or because such anecdotes provided information that was of relevance to the problem, but in a more indirect and unexpected way.

The relationship between an interviewer and interviewee is basically asymmetrical, because it is the interviewer who defines the theme, decides on which topics should be followed up, and has the greatest scientific expertise on the subject (2). I attempted to reduce this power imbalance by using meta-communication with the participants during the interview. Participants were reminded that they could talk about what was important to them, and it was made clear that they could take breaks during the process. I also repeated the information that I had received back to the interviewee, to ensure me that I had understood it correctly (144). In this way, I tried to

determine whether the participants were comfortable with the situation and the form of the interview.

Two pilot interviews were conducted before sub-study 1, and these resulted in some additions / changes being made to the first interview guide.

### **Telephone interview, sub-studies I and II**

In sub-studies I and II follow-up data were also collected via telephone interview about 1.5 years after the initial interviews, concerning the prognosis of the seizures and factors influencing the prognosis study (sub-study I) and seizure development (sub-study II). The telephone interviews were conducted by me (sub-study I) and by the contact nurse (sub-study II).

### **Participant Observation**

Participant observation was difficult in practice, due to limited access to the private spheres of the patients' lives. Most participants wanted to keep their diagnosis hidden from others. Several isolated themselves and would rather just stay at home. Participants also were relatively mature (14 - 24 years). This made it difficult to determine the role that I would take if I should be with young people in their social settings. Moreover, the participants were spread out across the country, and therefore only those who lived closest were offered home visits. One participant said in advance that she did not want to have a home visit.

One goal of participant observation is to describe what people say and do in circumstances that have not been prepared by the researcher (145). Should researchers conduct more participant observation studies, there would be greater opportunities for capturing more subtle situations that even the participants themselves would not have noticed. Moreover, participant observation provides direct information about events, because the situation does not need to be interpreted by the participants. Participant observation also provides the opportunity to capture events that are not included in the researcher's preconceptions and thus the researcher does not ask about during an interview (145).

Instead of the planned participant observation, we facilitated a more participatory approach with 4 “face-to-face” discussions (average duration of 65 min), that were conducted while the participant was readmitted to the hospital, 6 telephone conversations (average duration of 25 minutes), and 3 home visits. In total, this amounted to 26 pages of notes. Home visits were mostly characterised by informal conversations. On one occasion, I joined the participant in attending a handball match, but otherwise the observation occurred at the participants’ family homes.

The information obtained from the home visits did not result in much new material in relation to the focus of this study, but they did provide information about the dynamics within the participants’ families. Thus, the research study mostly utilises the field notes from the home visits to set contextual frameworks for the individual participants’ lives. The follow-up discussions provided information on how the participants experienced coming home with a “PNES diagnosis”. Again there was not much new data and, in practice, this material was largely elaboration and confirmation of the information that the interviewees had already provided during the interviews.

## 4.5 Presentation of participants



**Figure 1.** Group 1. Participants – sub-study I.

Ten participants aged between 16 and 61 years. Mean age, 27.3 years. Seven women and three men participated.

*Psychosocial aspects:* Six of the patients had considerable psychosocial problems, including anxiety, depression, and somatisation, isolation, and personality deviation. Four patients had no apparent emotional or psychosocial problems.

*Employment/schooling:* None of the patients were in employment; six were in education.

*Seizure characteristics:* All ten patients had frequent and disabling seizures with extremely varied semiology. Nine of the patients exhibited three or more seizure types, and nine of them had convulsive attacks, two with opisthotonus. Seizures lasted between a few seconds and more than one hour.



**Figure 2.** Group 2. Participants – sub-studies II and III.

Eleven participants aged between 14 – 24 years. All were female.

*Psychosocial aspects:* The majority (7/11) reported that prior to the onset of the condition they had experienced stress, being a victim of such as bullying, exclusion, or family difficulties. Some of the participants (4/11) nevertheless reported that they had no such negative life experiences.

*Employment/schooling:* Nine were in school or were studying, and two were in employment. All of them had considerable sick leave due to their seizure situation. Several reported that they originally had good results at school and in sporting activities, but their seizure condition resulted in poorer performance at school and fewer social activities.

*Seizure characteristics:* Almost all the young people involved in this study experienced daily, prolonged seizures that often came without warning. Several types of seizures were reported. Almost all had seizures with convulsions and some with falling. Seizure frequency varied, but many had several seizures per day. Seizures lasted between one minute and several hours.

Most of the participants (9/11) also reported that paramedics had been called upon on several occasions or they had been hospitalized on several occasions due to seizures.



## 4.6 Analyses

In the initial sections of this chapter, it is mentioned that the data was analysed using systematic text condensation (STC) and thematic analysis (TA). We have followed these analytical methods as an inspiration for the analytical processes. We have particularly followed the methods described by Malterud 2012 (140) and Braun & Clarke 2006 (137) (see also the section regarding Discussion of Methods).

The analyses build a bridge between raw data and results, via interpretations and summaries (136). In order to have a record of the analytical process afterwards, a continuous log was maintained of all the analytical phases throughout the three studies.

In the descriptions below I have attempted to make the analysis process transparent, so the reader can follow the procedures and be aware of the choices that were made during the process (140, 146).

This section of the chapter is structured such that it begins with a short description of the theory of STC. This is followed by a description of how the analytical process was conducted in Studies I and II. Finally, the theory of TA is described, together with a description of the analytical process used in sub-study III.

### *4.6.1. Systematic text condensation (STC)*

Systematic text condensation (used in sub-studies I and II) is a pragmatic, descriptive, and exploratory method (140). STC is inspired by Giorgis phenomenological psychological analysis (139) and has been modified by Kirsti Malterud (134, 140). The method is well suited to development of descriptions and concepts, and is primarily suitable for analyses across multiple participants. The method of analysis is inductive. The main intention is to present the experience of participants as they themselves express it, and not initially to explore the underlying meanings behind their statements. The analytical processes in STC move between decontextualisation and recontextualisation.

## **Description of the analytical process in Study I**

In the first phase of the analyses, we tried, wherever possible, to bracket our preconceptions (140). A nurse at one of the wards helped the researcher with the selection and definitions of the themes. The interview transcripts were read by both of us and each of us made a summary, of about one page for each participant, regarding what we considered to be the main theme of the participant. We also made a short title or heading for each abstract that covered the essence of each participant's perspective and proposed sub-themes. The core citations supported the abstracted text and the theme suggestions. By discussion, we identified the preliminary topics that were of relevance in each individual interview. In the course of this work we were able to identify four main headings that we believed covered all of the text that was relevant to the focus of this study. These four headings are listed below:

1. The patients' understanding and description of the epilepsy diagnosis.
2. The patients' reactions to the epilepsy diagnosis.
3. The patients' understanding and description of the "PNES- diagnosis".
4. The patients' reactions to the "PNES- diagnosis".

The meaning units for each interview were now categorised below one of the four headings above. Under headings 1 and 3 we categorised the meaning units that addressed how the participants understood the two conditions' explanatory models and treatment models. Under headings 2 and 4 we placed the meaning units that were associated with the emotional reactions of the participants to the diagnosis.

The meaning units were then taken out of their original context and put together with text from other participants under the same headings. In this phase the text was also sorted according to themes (140). During this process, some separate themes were merged, while other new themes emerged. Several meaning units appeared contradictory. These contradictions were primarily concerned with meaning units among the group members, but on some occasions we observed that an individual participant could make opposing statements at different points during the interview. We noticed that on these occasions this was often an expression of ambivalence, and was associated with particularly vulnerable themes. One example was that one participant emphasized that she neither needed nor wanted a diagnosis of epilepsy, but at another time she

stated that she wished she had an epilepsy diagnosis, because then she would receive more help. Such contradictory statements were emphasized in particular in the interpretation.

When all the meaning units had been united under a single theme, the text was sorted into code groups. In this phase, tables and forms were made in which internal links were used for horizontal and vertical associations. During this work, the code groups were divided into sub-groups. These sub-groups reflected the different ways in which the content of a specific code group could be understood. In the subsequent phases, the text was interpreted and condensed. Finally, we recontextualised the material (140) by developing content descriptions that were compared with the summaries from the first analytical phase and from the original interviews. We investigated whether there was an overall correlation between our results and the collective summaries, and at the same time ensured that there was no erroneous interpretations of the information from each of the participants.

### **Description of the analytical process in sub-study II**

In this study, the primary author and collaborators worked closely together throughout the analytical process. First, the interview transcripts were read separately by the individual researchers in order to get a total impression (140). The primary author made a single page summary of each of the participant's main themes, with selected key quotations to illustrate the contents. The primary author also drafted an overview of the preliminary themes. This document was used during the analyses as a reminder of the main themes of the participants. Each of the researchers then made a draft of the themes, and these were then discussed by teleconference. Although different headings had been used, the themes identified by the different researchers largely coincided with each other. By discussion, we reached agreement on the following six preliminary over-arching themes, with sub-themes beneath. We negotiated our way through the following six preliminary key themes, along with sub-themes:

1. Relational effects
2. Communication of the "PNES- diagnosis"
3. Illness beliefs
4. Coping strategies

5. Symptoms and personal meaning
6. Medical history / symptom descriptions

In the second analytical step, the text was entered into NVivo 10, a computer software package for qualitative data analysis, which has been designed for research in which the data is very rich text-based material. All meaning units were coded on a line-by-line basis, and grouped under one of the sub-themes under the over-arching themes listed above. At this stage in the analysis, new sub-themes were created. The meaning units were marked by code numbers, participant number, and using a colour-based system in order to identify to which of the 6 main themes the meaning units belonged. In practice, the coding was accomplished by the researchers first marking the appropriate text colour in the printed version of the material, and then the text parts were added into NVivo. By using this approach it was easier to maintain an overview of the material. In addition, this was a quality assurance procedure, because it required that the text was read through twice.

Two versions of the text were created. A theme-based (or decontextualized) version in which the meaning units from all the participants were grouped together under a single sub-theme, and a participant-based version in which the respective meaning units were grouped together under the heading of the appropriate participant.

We now had over 600 meaning units. In order to limit ourselves to a manageable analysis, we chose to explore in greater detail just one of the six over-arching themes. The theme “Relational effects” was selected because this was considered to contain the most significant content in relation to our aim, and in relation to the analytical emphasis in the empirical data. The theme “Relational effects” consisted of the text in which participants went into their relations with others. We had already identified the following seven sub-themes within the main theme of “Relational effects”: Stigma; Credibility; Distrust; Transparency about the diagnosis and the seizures; Legitimization attempts; Explanation problems; Experiences of being understood / misunderstood.

During this phase the sub-themes were slightly adjusted; for example, the sub-theme “Explanation problems” was considered to be too narrow and was exchanged with “How is the

condition talked about”. The sub-theme “Credibility” was expanded to “Credibility / mistrust”. In addition, two further sub-themes, “Fighting for dignity” and “Support”, were added. Thereafter, the meaning units in each of the sub-themes were encoded into code groups.

During this work “trustworthiness” was found to be central in the participants’ statements. We therefore created mind maps of the meaning units that were listed under “trustworthiness”. This approach clearly showed how closely trustworthiness was associated with “meaning”. We therefore returned to the over-arching themes of “Symptoms and personal meaning” and “Illness beliefs” (numbers 3 and 5 of the six preliminary main themes) and then related these to credibility. During this work, we arrived at the five main headings listed below, and then coded the text under these headings.

1. Fear of being considered to be simulating / faking
2. Actually being found to be simulating / faking
3. Correlation (or lack thereof) between the explanatory models of the condition and own understanding of the condition
4. Holding on to (or loss of) own identity
5. Trust (or lack of trust) that others hold onto the identity of the participants

In the final phase the text was recontextualised, using the content descriptions of the sub-codes. We had narrowed our focus of the analysis by looking in detail at the “Relational effects” and “Symptoms and personal meaning” and the sub-theme of “Credibility”. The preliminary results were now examined in the light of the other four preliminary over-arching themes from the first analytical phase. In this final phase, the results were also validated by comparison with the summaries that had been developed during the first analytical phase.

#### *4.6.2 Thematic analysis (TA)*

##### **Description of the analytical process in sub-study III**

According to Braun and Clarke (137) thematic text analysis is a flexible analytical method that is not based on a specific methodological framework, but can be used to describe experiences,

meaning, and real-life events of participants. In this study, TA was used to identify, analyse, and find patterns across the dataset. According to TA, a theme should contain something that is important in relation to the research question and represent a set of patterns, responses, or meanings within a dataset. How frequently a topic occurs, does not necessarily indicate the importance of that topic. A theme must not occur in all interviews (137).

The starting point of the third study was exploration of the participants' experiences with PNES in everyday life. This final issue was developed during the analysis process.

The first phase of TA is concerned with familiarisation with the data (137). We started by coding all the text from the original interviews with the 11 young people, in which they went into their relationships with others in everyday life. This involved considering which people they interacted with, the reactions from these various people, what was difficult, and how the difficulties were resolved. This material amounted to the article dataset.

We then wrote down *preliminary* themes / patterns and what it was they focused upon. The text was then sorted into codes. The qualitative computer software (NVivo) was used to systematize and structure codes in the first analytical steps. Following Braun and Clarke (137) we tried to ensure that the data were coded as closely as possible.

In the second phase, we continued to look for patterns that began appearing across the dataset (137). The dataset was now coded in several rounds. With the starting point in different analytical questions, we “cross-checked” the data throughout the analytical process. We found that it was useful to divide the participants' behavioural patterns into two categories, “open” and “closed”. In the category “open”, we coded text containing meaningful units where the participants spoke openly about their condition and participated socially. In the category “closed”, we coded text where the participants tried to conceal their diagnosis, and isolated themselves socially because of their condition.

A table was made to show the number of “open” and “closed” meaning units (here called extracted data) per participant. Most participants had multiple data extracts that were divided into both “open” and “closed” patterns of behaviour, but one of the participants had only a few

data extracts. A one-page summary of “openness” and “closedness” was also developed so that it was clear what was encompassed by the themes. Finally, a written text was prepared for each of the extracted data sets and how it referred to the following points: What characterized the situation? What types of dynamics were seen? What is the mood of the situation, and who did what?

In the third phase, the codes should, according to Braun and Clarke (137), be analysed and sorted into themes. In this phase, we used mind maps and tables to look for patterns throughout the material. We posed the following questions to the material:

- What are the reasons for, and the consequences of, the choice of this action pattern?
- How might this action pattern affect the participant’s illness perceptions and vice versa?

In this analytical phase, it became clear that “openness” versus “closedness” occurs in three areas:

- 1) The specific seizure situation
- 2) The diagnosis itself
- 3) In relation to the consequences of the seizures for the patient (tiredness, social withdrawal, etc.).

It now seemed natural to divide the codes among the three different arenas in which the participants belonged:

- 1) Immediate family and friends
- 2) School, work, recreation
- 3) The public sphere

At the same time, a more precise problem approach was developed and a storyline established that contained shared aspects for each participant, but to different extents. The behavioural patterns of each interviewee were considered in relation to their own understanding of the illness. The final themes had now been established. The route from code to theme included interpretation of the codes that was purely empirical (137). Finally, we checked the themes in a

comparison with the transcribed material and notes from the follow-up calls to see if this perspective could provide new light on the results.

In the fourth analytical phase, the themes were reviewed and controlled such that there was certainty regarding which themes were to be included, and what they were concerned about (137). The themes were now validated by new questions being posed to the material. In this phase the analyses were longitudinal. A summary of each interview was also made, with quotations that supported the summaries attached. All the analytical phases were performed in collaboration with the whole research team and the articles' authors. Key quotes were used to corroborate the findings made in the final presentation. When the finished results had been achieved, these were then considered in the light of Goffman's theory of identity (147).



## 5.0 Discussion of methods

Validity in qualitative research and the way in which the validity is conducted is a constant discussion point among leading researchers (2). For most researchers, validity in itself refers to both internal and external validity. However, the techniques for achieving this and the measure that is being sought, for example consistency, reliability, objectivity, verification, generalizability, or transferability to others, appears different from that of quantitative research (2). According to Morse (144) use of the concepts of reliability, validity, and generalizability (or external validity) is recommended. For quality and validity, the extra dimensions of 'reflexivity' and 'relevance' can also be added (134). In my discussion of this study's validity and quality, I have mainly used concepts of reflexivity and internal and external validity, as those terms are understood by Morse (144, 148, 149) Malterud (134, 140, 150), Kvale and Tong (2, 146). In order to avoid too much repetition, I have reflected only on some of my choices in the description of the methods. There is also some degree of overlap between reflexivity and internal validity.

### 5.1 Reflexivity

My role as a clinical nurse for this patient group for many years, as well as my field of interest and my theoretical framework, will have affected the entire research process (146). This may have led to results and interpretations that concur with my preconceptions, despite actively adopting strategies that may challenge the findings (150). This chapter is used to reflect on my role in this study, and to comment upon how attempts were made to offset bias.

As a psychiatric nurse, I am used to assuming a supportive and affirmative attitude when difficult subjects arise during discussions with patients. A research interview, however, has a different agenda than a clinical conversation, and, moreover, the relationship between interviewer and interviewee ends after the interview. Participants may feel rejected if they have opened up too much. I tried to find a balance between maintaining a research role, while simultaneously trying to be supportive. In a few cases, I invited the participants into more informal discussion after the tape recorder had been turned off, because the young interviewees had opened up a lot during the interview. This resulted in new information that was not included

in the study because the participants had not provided the information while the discussion was being recorded.

My role as a professional, working at the same hospital as where the study participants were patients, may have created a power imbalance in which the participants, either consciously or unconsciously, tried to answer the questions in line with what they considered to be my expectations (146). This possibility is also reflected upon in the discussion of the results. An interview should be understood as human self-representation, which also means that the interviewees have the opportunity to try to show themselves as they want to (145). When the participants were informed about the study prior to the interview, I said that I wanted to investigate how it can be experienced to have seizures for which the cause is not properly understood. My preconception was that this was difficult. The patients may have noticed this preconception, either consciously or sub-consciously, so that the focus on their *positive* experiences with the diagnosis was too restricted. For example, it came as a surprise to me when one of the participants spontaneously told me that the condition had led her to have a greater appreciation of life. This statement could form the basis for a new pattern in the analysis, but the dimension of coping was not followed up because it was considered to lead the study in a direction that was away from the study's main theme. The participant's statement had more focus on positive experiences with "being ill" than with have a "PNES diagnosis". Although the coping aspect is included in all the articles, it is largely in relation to factors that contributed to coping with the diagnosis.

Use of a semi-structured interview guide, in which we endeavoured to follow up participants' associations with specific questions, has contributed towards validation of the study. This meant that the participants took up themes that were beyond those that I could anticipate and ask questions about. One example of this was when several participants spontaneously related that feeling believed was essential for them to be able to advance in their processes in relation to understanding the relationships that could explain the seizures. This was a key finding of the study, which we did not go into in the researcher preconceptions.

My clinical and theoretical preconceptions influenced the choice of problems to approach and the direction of analysis. The three studies are based upon each other. The results that we chose

to continue further with in the last two studies were determined by several factors. Sub-study I resulted in findings in different research areas, but it was the area concerned with the diagnosis being difficult to understand that we chose to explore further in sub-study II. This area was selected for further study both because it was within the researchers' field of interest, and also because it was perceived as being of significant relevance to the field of research. Furthermore, it was possible to explore this subject using the study's methodological approach.

The material obtained in sub-study II was so rich and diverse that it could have provided the opportunity for many directions in sub-study III. However, we chose to move forward with the text that was concerned with situations where the patient interacted and had social relations with others. This particular field was selected primarily because the social interactions in adolescents with PNES have been little explored, but also because this topic was within the researchers' interest. Furthermore, the young people who were participating in the study were themselves concerned about their relationships with friends, school, and family. The participants expressed different reactions and challenges that were related to these, and it appeared as though this was an issue where something was "at stake" for them. Thus, the direction of study has been characterized by: exploring areas where there is a lack of knowledge in the research field, exploring areas which participants themselves have emphasized as being of importance, the researchers' preconceptions, and which theories have been selected.

## 5.2 Internal validity

This section discusses the study's internal validity; that is, whether the selected methods are relevant and appropriate for answering the study question (2, 150). The question of relevance occurs in all phases of the study. It applies to whether the methods that we have used are relevant for addressing the study problems, and whether the results that the study has produced are of relevance to the field (134). In the following paragraphs I have selected different aspects of the study's internal validity and relevance that I think are most relevant to discuss within this context.

*Sampling:* Inclusion of study participants was performed using purposeful sampling. That is, we included patients consecutively, provided they complied with the study's exclusion-inclusion

criteria (see Chapter 4.2). A strategic choice presupposes that we have identified the direction of research that we wish to explore (134). We chose to include patients who had recently had a diagnosis of epilepsy changed to a diagnosis of PNES (sub-study I), and patients who had received their “diagnosis” via a biopsychosocial model (sub-study II). We believed these patient groups represented a relevant choice, because they had just been in a situation where the focus of this study was the relevant topic. The data from sub-study III was the same as for sub-study II. This provided the opportunity to study some patterns between illness understanding (which had been explored in sub-study II) and social participation, which we might otherwise have missed. In retrospect, we have nevertheless asked ourselves the question of whether we should have interviewed the participants again in sub-study III. This could have given us descriptions of their social participation when they were back in their home environments again. An attempt was made to add this on afterwards with follow-up calls and home visits (see Chapter 4.4).

Preconceptions, the problem itself, and the theoretical framework determine which considerations are of importance in order to obtain data that provide sufficient depth and breadth of the phenomenon that is to be explored (134).

*Saturation:* Regarding the number of participants and analysis by qualitative methods, the term “saturation” is used (134, 148). According to Morse (148) the participants in the study must be *adequate* and *appropriate* for reaching saturation in a study. *Appropriate* means that the participants must be suitable to provide information on the phenomenon of interest (148). On the basis that we required participants who had knowledge of the study theme and could provide new insights (144), we chose to select participants from a specialist hospital. This also means, however, that our participants were amongst those most severely affected by the condition. As mentioned, patients with PNES represent a very heterogeneous group. It is possible that had we recruited participants from primary or secondary healthcare settings, then they would have contributed different experiences. This bias must be taken into consideration when determining for which group the study results are of relevance.

That the sampling should be *adequate* means that the number of participants must be sufficiently large for replication to occur (148). In Study I, inclusion of participants was stopped

at ten patients, three men and seven women. Because we worked in parallel with the analyses, after a while it became apparent that the participants' descriptions divided into three patterns.

- 1) Those who found that the diagnosis provided meaning and had associated hopes for treatment.
- 2) Those who accepted the diagnosis, but did not find that it provided any personal meaning.
- 3) Those who rejected the diagnosis.

Within these three groups, we found both similarities and differences, and we therefore considered that the data for the current study regarding categories and themes were saturated. Although inclusion of more participants could have brought some new nuances, the participants within these three categories provided the same responses overall. Thus, we had achieved a basis for advancing an argument that was rich and meaningful.

We completed recruitment of participants to sub-study II after several rounds of sampling and on the basis of an assessment of the ongoing interviews. These assisted in elucidating the focus of this study in a way that “made a difference” from what had been reported from other studies, and from what was manageable with regards to the size of the study (134). The heterogeneity of the group contributed to the diversity and variation within the material. The data contained contradictions and paradoxes, and was well suited to the exploration of internal relationships.

Nevertheless, in retrospect, we see that a broader variation in the choice based on who showed themselves to be most information-rich according to the objective of the study, could have strengthened the study. This is further reflected upon in the discussion of the results (Section 8.3).

When it comes to sub-study III the participants' experiences could be divided into two categories of “open” and “closed” behavioural patterns, with copious descriptions in both categories.

It also became apparent during the analyses of the material from sub-studies II and III that we should have included more participants who could have provide further information about the coping aspects. This had not been properly considered during the inclusion phase. Ideally, we should also have had male participants in sub-studies II and III, as it seems probable that the experiences of men with the diagnosis are somewhat different to those of women. However, the difficult recruitment process precluded the inclusion of male participants.

The analyses in all three studies were based upon detailed, comprehensive, and rich descriptions of the selected themes (148). Comprehensive descriptions contribute to internal reliability because data will overlap. Key issues that have resemblances and overlap enable the researcher to identify replication (148).

Thus, in this study *saturation* does not imply that all aspects of the phenomenon are covered, but that we have a sufficiently rich and diverse material to address the study question adequately, that the interviews fall into patterns that repeat themselves, and that a convincing and justified argument was possible to develop (148).

*Changes in the inclusion criteria:* Difficulties with recruitment led to changes in the inclusion criteria in Studies I and II, and this may have affected the studies' validity. The studies have several methodological limitations that make it difficult to state with certainty the consequences of how a biopsychosocial approach may affect a patient's understanding of their condition. (This is also further discussed in Chapter 8.3).

Difficulties with recruitment also led to a wide range in the participants' ages, which, in turn, meant that patients received somewhat different subject matter during the follow-up stay. This also led to the patients being in contact with various different personnel groups. All these factors may have affected the participants' experiences.

We found some patterns in the participants' experiences that were related to different their age group. Those who got the broadest range of " follow-up stay" were the oldest, and also those who described that they had had the greatest benefit from their stay. Five of the six participants

who received the broadest range of material described that their stay had resulted in improved understanding of the illness. They described a greater acceptance of the diagnosis and increased understanding of the relationship between seizures and life events. Three of these were seizure-free, whereas the other two still had some seizures at the time of follow-up.

The attitudes of the personnel were also highlighted as an important positive factor by the participants who had the longest follow-up stays. It also appeared as though the starting point for the older participants differed from that of the younger participants. Whereas the older participants talked a lot about the diagnosis being difficult to understand and accept, several of the younger participants had no need to explore the explanatory models of the condition. They themselves even had ideas about why they had developed seizures and they were primarily interested in addressing the practicalities of everyday life. This may be understood on the basis that the aetiology of PNES in children is probably somewhat different to that of adults, with the seizures particularly associated with interrelated external factors of a child's environment (see Literature Review, Chapter 2.0).

In one sense, the large age range among the participants and the necessity to recruit participants from two departments may have resulted in an enrichment in the data, as it appears as though some nuances regarding differences between the two groups have been exposed, about which we would otherwise have been unaware. Indeed, if we had had more participants from each age group, we might perhaps have been able to explore further subtle distinctions within each group (see Chapter 8.3)

Difficulties with recruitment also resulted in inclusion of patients who had epilepsy in addition to PNES. This was relevant for one, or possibly two, of the participants. In my opinion this did not reduce the strength of validity of the study, because the patient who had both PNES and epilepsy was himself aware of which of his seizures were epileptic and which were PNES. In the second case, not all the seizures were captured by telemetry, and therefore there was a lack of certainty regarding whether some attacks could actually be epilepsy. This patient was unsure about whether PNES was the correct diagnosis. This situation is a reality for many patients with PNES and therefore enriched the material of the study.

*Validity of the analyses:* Reflections regarding the validity of the analyses are also described under reflexivity. The validity of a study is strengthened if the data collection and the analyses may run parallel to each other (149). In Study I, this situation led to the development of new research questions. Because the participants, without prompting, talked a lot about their experiences with the healthcare system during the initial interviews, the following research question was included: *How do the patients feel that their needs are met by the Norwegian healthcare system?*

We chose to use TA in the final study, because we wanted to see if a different analytical method than STC would reveal other dimensions. As the transcribed empirical material in Study III was the same as that in sub-study II, it was important for us to explore it from a new angle and with a new method for analysis. One advantage of TA is that it is well suited to bringing out a coherent storyline. This enabled us to bring out an association between the participants' understanding of disease and their social participation.

The study's validity is strengthened by several researchers working together (146, 150). The bias introduced due to an individual researcher's preconceptions is counteracted by active participation of all the researchers/co-authors in all the phases of the study. During sub-study I, the primary author worked closely with a nurse during the coding and selection of themes. As nurses have different awareness of the research field, we had different assumptions. In sub-studies II and III the coding was conducted solely by the primary author, but was discussed both with a study group and with the other researchers / authors. That the co-authors had different scientific backgrounds was an asset as it meant that several perspectives were included in the study design and interpretation. In sub-study I, the co-authors included a neurologist and a psychologist, and in sub-studies II and III the co-authors included a physician and a medical anthropologist.

The pragmatic validity of a study includes, among other issues, how broadly statements of knowledge are accompanied by action (2). In order to achieve pragmatic validity, a more indirect validation format must be adopted, which means that statements also result in action (2). One ambition of this research was that the three studies should provide some implications in terms of changes in practice in the approaches of health professionals when meeting patients



with PNES. Whether the results of this research will actually lead to changes in practice is unknown, but we contend that the knowledge produced by the study contains some new ideas that may have consequences in terms of a more patient-centred approach, if taken seriously by healthcare professionals.

### 5.3 External validity /analytical transferability

External validity refers to the contexts in which study results can be applied (150). In our study we interviewed people with “psychogenic non-epileptic seizures” who had been admitted to a specialist hospital. Questions thus arise regarding the extent to which our findings can be extrapolated, firstly, to other patient groups with PNES, and, secondly, to people with other types of MUS.

Regarding the first question, people with PNES represent a very heterogeneous group, of which only a small selection has been included in our research. Although we have rich material with many variations, it is likely that there are some aspects of the phenomenon that we have not captured in our material. As mentioned in the section on internal validity, because our patients were recruited from a specialist centre, it may also be that we have interviewed people with PNES who have particularly serious problems. It is therefore not certain that everyone with PNES will feel that our findings reflect their own experiences.

Regarding extrapolation to people with other kinds of MUS, patients with PNES and patients with other forms of MUS often describe the same challenges related to living with a controversial diagnosis. We therefore believe that our results show some patterns regarding human experiences that we suggest could be usefully extrapolated to understanding people with MUS in general, and PNES particular. This particularly applies to what it means that the disease provides a personal meaning, and how the interplay between perceiving the condition as meaningful and legitimate may impact upon social participation.

## 5.4 Ethical considerations

Sub-study I has been reported to the Norwegian Social Science Data Services (NSD) and approved by the Regional Committee for Medical Research Ethics (REK South). Sub-studies II and III were approved by the REK North and reported to the Privacy Ombudsman for Research at the Norwegian Social Science Data Services (NSD).

The ethical rules for research on humans set requirements in the following areas: 1) informed consent, 2) confidentiality, and 3) impact (2). In the paragraphs below I have described how these rules have been followed in this study.

1. Informed consent: Informed consent forms for children under 16 years, and young people / adults over 16 years were prepared. These described the purpose of the research and the practicalities involved from participating in the study. In addition, verbal information about the project was provided by the departmental staff, and a period (a few days) to think over the project was allowed before the first contact was made by the researchers. It was emphasized that participation in the study was voluntary and that a participant could withdraw from the project at any time. In the case of withdrawal from the study, all data would be deleted (136). The participants were also informed about how the final material would be produced and the sort of journal in which the results would be published.

2. Confidentiality: Confidentiality during the process was ensured by storing the participants' names or ID numbers separately from the data. The interview tapes were only available to researchers and the person who transcribed the text. The person who did the transcribing was a signatory to the hospital's confidentiality agreement. The interview tapes were kept locked in a fireproof safe. Dialect, place names, or other information that could be linked to a specific patient were not transcribed. Different names were allocated to the interviewees, and personal data that could reveal the interviewees' identity were not presented. Medical histories and presentation of the interviewees were anonymised in the final presentations. In order to protect the interviewees' anonymity, an analysis model was chosen in which the results are produced crosswise. In preparation of the results, ethical considerations were continually evaluated with regard to maintaining the participants' anonymity. Information on the year of data collection

and the departments from which each of the patients were recruited is not included in the articles.

3. Impact: Research on children involves the same dilemmas that arise with all research, in which the benefits must be weighed against the drawbacks. In this case, any positive outcomes will primarily benefit their successors. Children and adolescents may be vulnerable, partly because they often trust figures of authority. In addition, young people who participated in this study also already have a disorder that indicates that they struggle with emotional difficulties. By agreeing to take part in this project, the participants are likely to be interviewed about topics that may be difficult for them. The interview could precipitate difficult emotions, and thus be perceived as an additional burden. For children whose background for PNES may be traumatic stress, it may take time before they can access the repressed memories and emotions. Furthermore, an interview may increase the focus on the patients' symptoms. Therefore each case was considered on an individual basis, considering whether an interview could have an adverse effect on a particular patient.

The adolescents were admitted to the hospital during the interview period. The intention with hospitalization was that, together with healthcare professionals, the participants could explore new ways of understanding their seizures. That somebody was willing to listen to the participants' own perspectives could also be perceived as a positive experience. Should the interviews activate difficult emotions, the patients were in a system with professionals who could provide help and support following the interviews. Therefore the interviews were conducted in good time before the participants were discharged from the hospital.

Research on children and adolescents may result in situations in which notification to the child protection authorities may be mentioned. It was clarified in advance that in the event of such situations, there was the right to notify the child protection authorities.



## **6.0 Theoretical approaches**

### **6.1 Illness concepts and dualism.**

Many people have tried to define disease as a concept, but without reaching any adequate and satisfactory definition (151, 152). Although the study did not have any ambition of providing detailed philosophical explanations for the term, in this Chapter I will nevertheless give a brief account of how disease is considered in this study.

One natural scientific paradigm has deep roots in western culture (153) and has left a significant imprint on biomedicine. Based on such a paradigm, disease can be understood from the model of disease occurring when the body's organs deviate from their natural functions. Christopher Boorse has become the spokesperson for this view of disease. According to (154), the presence of disease is determined by fundamental, objective, and value-neutral facts. The human body consists of organ systems with natural features that may deviate from the norm in many ways; disease occurs when there is dysfunction in one or more of the organs. "Normal function" is based on given frames of reference; although some deviations from normal functioning may be harmless or even beneficial, others are not, and the deviations in this latter category are "diseases" (154). A biomedical concept of disease thus involves searching for unambiguous, objective biological causes for a patient's symptoms (155). Subjective symptoms, which cannot be measured and thus do not manifest as being deviations from the normal range, will, on the basis of this model, not be classified as being due to disease. With MUS, in which the state cannot be explained on the basis of clear organic findings, the model therefore provides a poor description. Thus, according to this study's view of disease, a strictly biomedical view of illness is considered to be reductionist.

The biomedical model has evolved further. In medical practice today, holistic and patient-centred models are increasingly developed (156, 157). Biomedicine currently comprises many different biological processes, including also those illnesses that we describe as MUS. One example of this is irritable bowel syndrome (IBS), in which we now have a better understanding of many of the biological processes taking place (158).

The position taken within the current study is that disease cannot be understood on the basis of a single specific standpoint. In Norwegian we have only one single word for disease (sykdom), but in English there are at least three concepts:

1) *Disease* denotes the condition as a reduction in biological function compared with normal function and is therefore adequate for a biomedical model. According to Hoffman (159), disease is a health professional's perspective, and having a disease involves the view that health professionals can localise and classify the phenomenon such that targeted treatment can be initiated (159). According to Richard Mayou (160) disease implies the presence of biologically structural abnormalities in the body organs and systems (ibid). 2) *Illness*, however, covers the patient perspective and involves "being sick" (159, 160). Illness is characterised by a patient's subjective perception of their pain and suffering (159). 3) *Sickness* addresses the social perspective, which, among other things, means "to have the role of being sick", that is to be perceived as being ill in a social context and has an impact on social status and rights (159).

All three aspects of the disease are considered as important in this study. Based on the definitions above, illness and disease can occur independently, although they often co-exist (160). A person may have disease without illness, because somatic disease does not necessarily give a subjective experience of being sick. A person may also have an illness, although the symptoms cannot be explained by biomedical investigations.

The significance of social interactions can be an important factor in understanding disease (130). The importance of seeing a sick person within his or her psychosocial and cultural context is therefore considered important (130, 160). According to the perspective of this study on disease, the absence of disease does not mean that the individual is not sick. It might only mean that the presentation of the conditions cannot yet be explained on the basis of biomedical findings.

Regarding the causes of disease, this study supports a multifactorial understanding that goes beyond the classical body-mind dualism. Based on such an understanding, a simple linear cause-and-effect approach, in which PNES is based on a single primary psychological cause and the seizures are considered as secondary to the primary psychological cause, will not be adequate. Although individual cases in which, for example, a sexual assault can be a significant

factor for a person developing PNES are conceivable, the conditions such as the individual's coping strategy, social support, previous emotional vulnerability, identity etc. also are of relevance regarding whether or not the person develops PNES.

Body-mind dualism is deeply rooted in western society and continues to affect our health care system. Although we know that the aetiology of, for example, ME cannot be understood as *either* physical *or* psychological (161), such thinking characterises debates in the media and the understanding of the general public (162). Some argue that ME is due to a biological cause, such as a virus, whereas others argue that the condition is due to emotional conditions. A dualistic view also influences the treatment apparatus, which is divided into either psychiatric or somatic treatment. For conditions such as PNES, which is located in the borderland between neurology and psychiatry (48), this division could lead to inferior health care possibilities because the diagnosis is made by neurologists, but with treatment by psychiatrists. There is often a waiting list for treatment, such that there is little continuity in patient care. A division of disciplines also means that competence is lessened, due to the risk that no one sees the whole picture.

The terminology for MUS and PNES is also characterised by a dualistic mindset, because the terms are based on the exclusion of biomedical findings. Terms such as “psychogenic” and “pseudo” also easily provoke associations that the symptoms are “not real” or are “all in the mind”. Thus, the biomedical paradigm and dualistic approach contribute to people with MUS feeling that they are delegitimised as their symptoms are not considered “real” (113, 163, 164). Regarding the terminology used for MUS and PNES, our study supports the results of a review of functional somatic symptoms that concluded that the terminology used, which presupposes a mind-body dualism (such as somatization, medically unexplained) is unfortunate (11). In practice, however, I think this may be difficult, because we lack concepts that are appropriate for these conditions.

Health care providers probably have a more nuanced and comprehensive understanding of MUS than the general population. However, because the concepts are characterised by dualism, it can be difficult for patients to "hear the entirety". When journals become filled with negative (or non-confirming) test results, the patient may feel that they are not being trusted. The challenge

therefore lies in finding a technical language for clinical work that integrates body and mind into an undivided whole (161). Although it is difficult to conceptualise the body and mind as being integrated with each other, there are now good models that demonstrate how physical and mental processes and life conditions interact closely with each other, such as the Cognitive Activation Theory of Stress (CATS) model (165). The model shows how emotions, cognitive functions, subjective experiences, the nervous system, and physiological stress reactions cooperate through complex signalling systems in order to restore balance (165).

The term bodily distress syndrome (BDS) also gives some indications regarding treatment of MUS. The term includes most of the conditions usually classified under MUS (166) and, amongst other things, also provides treatment guidelines related to the patient's coping strategies and measures that could be implemented to inhibit activation of the autonomic nervous system (167). Such models can help in the creation of an understanding of how all types of pressures (stresses) can affect balance and cause physiological activation. The resulting stresses on the individual depend partly on interpretative pressures; previous experiences have an impact on interpretations and expectations. Cognitive processes may affect whether the condition will develop towards a disease state or in the direction of health. Persistent activation that is beyond an individual's control may result in chronic maintenance of inappropriate activation and may explain disease mechanisms in MUS (165).

In summary, this study distances itself from a stringent biomedical model of disease. The biomedical paradigm is seen as a barrier for patients with MUS to adopt explanatory models that override the psyche and soma limitations. Thus, this study relies on a multifactorial understanding of disease that goes beyond the body-mind dualism.

## 6.2 The biopsychosocial model

In response to the inadequacy of the biomedical model, Engel developed the biopsychosocial model (168). The model is based on the biological, psychological, and social factors that can have a role in the development of disease. Thus, the model makes a break from a dualistic frame of reference in which symptoms are interpreted as either somatic or psychological (168). Nevertheless, the model is criticized because it is not sufficiently comprehensive. The criticism



is primarily associated with our recent knowledge that it is impossible to distinguish between biological, psychological, and social factors (156, 169). On a theoretical level such an understanding provides meaning. An understanding that every thought activates “complex neuronal processes”, makes it impossible to say that something is just physical and something else is just psychological. All aspects of “lived lives” thus incorporate biological effects. The question in this context regards the practical implications provided by this understanding. Clinically, I believe that this can be challenging, due to the prevailing mind set and conceptual framework that remain characterised by an underlying dualism separating psyche and soma. We therefore continue to lack a vocabulary that covers the entirety.

Henningsen writes in an editorial that if the model is to be used in the 21<sup>st</sup> century, then continuous work with the model is necessary, and this is particularly relevant with respect to the biological mechanisms that underlie the body-mind-environment interactions (156). The term “bio” in biopsychosocial has expanded since Engel’s time, and we know more today about the biological underpinnings of, for example, empathy, cognition, attachment, and early adversity (156). The knowledge base on the biological mechanisms of PNES is also steadily increasing (65 -67, 206) but the field is still complicated and the studies small, and therefore it can be difficult to communicate this knowledge to the patients.

Although the model needs to be developed further, in step with our growing knowledge base, the model is considered by many to be suitable for both theoretical understanding of PNES and for clinical use (30, 85). Clinically, the model can provide a better understanding of how conditions beyond the purely medical may have a role in the development of disease (157, 170). At the same time, the model can elucidate that biological processes are involved in disease states that are currently categorized as “unexplained”.

Traditionally, communication between clinician and patient has been characterized by being “doctor-centred interviews” in which the doctor decides on the direction of the conversation and where the purpose is to make a diagnosis on the basis of biomedicine (157). This can contribute to a form of communication in which the patient’s perspective is barely put forward and is thus poorly suited to cases of MUS, in which the patient and clinician must work together in order to

achieve a common understanding of the symptoms. There have now been more patient-centred forms of communication developed, including the biopsychosocial model (170). During the last 10 years such patient-centred approaches have also been included as a part of medical studies (157).

In a textbook of child psychiatry (171) a biopsychosocial model is described as having the potential to be a good clinical tool in both *diagnosis and treatment* of children who have symptoms that cannot be explained on the basis of organic findings. The table below is taken from Grøholt 2008 (Chapter 26 p 325) (171) and gives an example of how such a model might appear.

<b>Factors</b>	<b>Predisposing</b>	<b>Triggering</b>	<b>Maintaining</b>
<b>Biological (the body)</b>	Physical diseases in the family (genetics, model-learning) Patient's previous injuries from disease Neurobiological vulnerability (delayed or disturbed motor development or learning disabilities)	Physical illness or injury Age-related biological changes, such as puberty	Inactivity, disturbed circadian rhythms Constantly requiring new medical tests Other self-perpetuating biological mechanisms
<b>Psychological (feelings, thoughts)</b>	Personality, temperament	Insurmountable stress and intolerable pressure Trauma, abuse, violence, bullying, social exclusion	Individual and family disease understanding, focus on symptoms, anxiety Ongoing psychosocial burdens
<b>Social (surroundings)</b>	Family, school friends, leisure activities	Life events and pressures, family conflicts, divorce, family illness, relocation, change of school Demands and expectations that exceed individual abilities to cope	School absenteeism with increasing academic concerns Social isolation, depression

Although we have used the biopsychosocial model in this study, this does not mean that I think that the model is optimal for understanding PNES completely, either clinically or theoretically.

It is also unfortunate that a division into biological, psychological, and social conditions is conducive to maintaining a hierarchical system in which the biological aspects of disease are ranked above psychosocial factors (156, (169). This provides the normative problems associated with the diagnoses' different values. However, a model for disease in which psyche and soma are understood as a single unit restricts the unequal ranking of disease (169). If clinicians explain to the patients that there are also biological processes involved in MUS, the delineation of disease into purely physical and purely psychological may be increasingly overcome, and thus, consequently, the stigma associated with MUS may be reduced.

From a philosophical perspective, the study thus supports an understanding that is based upon how biological, psychological, and social/cultural issues are so intertwined that it is not possible to distinguish them from each other (156, 169). In my opinion, conceptualisation of such an understanding may be difficult and, therefore, I believe that the model can be a useful tool *on the path towards* a broader understanding and interpretation of the individual as an indivisible whole. The model can assist in coming closer to the lived lives of the patients than can be achieved by the biomedical model alone (157, 170).

In this study the healthcare personnel kept a biopsychosocial understanding "in mind" during initial discussions with the patients. Further on in the course of the discussions, the various biological, social, and emotional relationships that the patient themselves had brought forward were summarized for the patients by the healthcare professionals. The intention was to give the patients the opportunity to perceive their condition in the light of a multifactorial understanding. This is explained in greater detail in Section 8.3.

### 6.3 Diagnoses and hierarchy

Diagnoses as a concept is of interest in this research because different sub-sections of the study use different approaches to investigate patients' experiences with the "PNES diagnosis". In the first sub-study, we explored what it can feel like to have the diagnosis altered from being neurological (epilepsy) to PNES, which is a more unexplained diagnosis. In sub-study 2, we examined the significance of having the "PNES diagnosis" explained to the patient using a

biopsychosocial model. In the third sub-study we examined experiences of living with a diagnosis of PNES in everyday life. Because PNES, like other conditions with MUS, is often perceived as a contested diagnosis, this aspect is emphasised throughout this section.

A diagnosis is a classification tool for medicine and is decisive regarding how medicine performs its role in society (172). Diagnoses govern medical treatment and care, they give structure to the clinical picture, and provide guidelines for medical education. At the same time, diagnoses guide how health care is organised in various disciplines. Diagnoses provide access to services and status (172). What can be classified as a diagnosis is determined by, among other things, what is culturally acceptable in society and the extent to which it is believed that the condition can be considered as normal or that it should receive treatment. For example, homosexuality would not be classified as a diagnosis today, but did so previously (172). In addition to directing medical treatment and creating the hope of a cure, a diagnosis is intended to provide meaning and legitimacy to the symptoms, both for the patient and for society (172, 173). Thus, patients that have their condition labelled as a disease can obtain both emotional and financial benefits.

Medical specialties and diseases are ranked differently and have different levels of prestige (174, 175). Diagnoses of disease or somatic diagnoses are given greater value and recognition than symptom diagnoses that are based to a greater extent on a patient's subjective symptoms (174). Those conditions that cannot be associated with a particular location in the body and chronic diseases are often ranked relatively low (174, 175).

There are several conditions that are not perceived as disease states (152) and not everyone with a condition with MUS receives a diagnosis (176). Without an acceptable diagnosis, the patient risks not having social recognition, because society rarely acknowledges illness in the absence of disease (176, 177). This patient group describes considerable fear that society will not believe them, fear of stigmatization due to their symptoms being construed as psychogenic, and fear that they will not qualify for social security benefits such as sick leave, etc (172, 177, 178). Diagnoses that are not perceived as legitimate, that do not make sense to the patient, and that do

not provide much indication regarding treatment may be perceived as additional loads, rather than being of help.

In this study, our focus was directed towards how the biomedical paradigm provides constraints regarding which diagnoses give recognition, and the consequences that this may have for patient groups who have symptoms that cannot be explained by organic findings.

#### 6.4 Concepts of social identity

The concept of identity is well known and is used in many different ways. In this study, the term is limited to identity in association with disease and diagnoses. The concept of identity has a long tradition in sociology, and has its roots in psychology (179). It is usual to distinguish between two basic types of identity: personal identity and social identity (179). Social identity is also known as group identity (180). Whereas personal identity is related to the individual, social identity is related to interaction with others (179). A simplification of these concepts is that personal identity can be described as what makes a person the unique individual that he/ she is - the factors that distinguishes one person from another (e.g., appearance, values, body posture etc.), whereas group identity or social identity implies that we can be divided into different groups, social communities, interest organisations, etc. The concepts cannot be considered to be independent of each other (179).

From a sociological perspective, identity is related to a number of criteria that play a role in the interaction, when an individual considers themselves in relation to others (179). We belong to different social groups such as family, neighbours, workplaces, religious communities etc. Given that these groups become internalized as part of our social identity, this also affects our psychology. These groups can offer social life, emotional support, and personal safety, they can strengthen our self-esteem and self-worth. Such positive groups provide meaning, purpose, and identity, thereby contributing to affirmative psychological consequences. However, if an individual should be rejected by the group or the group changes, these factors can have a negative influence on an individual's mental health (180).

Social identity theory can be understood as those aspects of self-image of an individual that originate from those social categories with which the individual is associated (181). Any individual generally tries to maintain a positive social identity and a positive self-image; a positive social identity may be associated with preferential feelings for the group to which an individual belongs, i.e. “in-groups” in contrast with “out-groups” to which an individual does not belong. Should an individual not consider that their group is satisfactory in comparison with other groups, then the individual may attempt either to move to another group or to alter their own group. An individual’s identity is not only based on membership of a particular group, but also on the condition that members of that group differ from members of other groups. The “us versus them” distinction not only helps people to understand themselves, but it is also of relevance for assessment and experience of value (181).

An internal and external dialectic movement occurs between self-image and the public image. Similarities and differences are implicit in each other; one does not make sense without the other (183). Social categorization is a cognitive tool that helps us to systematize the world (182). Without categorization, it is not possible to master the complexity of the social world (183). Identification, that is, recognition of what something is and what it is not, involves some specifications of properties, which are fundamental for categorization (ibid).

Goffman (147) distinguishes between the concepts *apparent social identity* and *actual social identity*. According to Goffman, apparent social identity is the first impression that one person makes on another. This first impression is based on our stereotyping of people and is transformed into normative expectations regarding how the other person is and which qualities he/she has. An individual’s actual social identity, however, is the category that the individual person actually does belong in, and the abilities or qualities that the person actually possesses. When we first meet with a stranger, we believe that we can predict the category in which he belongs, and the qualities he possesses, or, in short, the social identity of the stranger. Impersonal contacts with strangers are therefore particularly vulnerable to generating these stereotypical images. The categorical setting is, however, weakened when others know a person in advance of meeting them (147). According to Goffman, a *stigma* is the extreme disapproval of a person and may arise if there is a discrepancy between an individual’s actual identity and apparent identity (147).

Disease and diagnoses will affect an individual's identity (7, 116, 184 -186). With MUS, identity can be challenged in various ways, including because the diagnostic explanations did not make sense or are perceived as illegitimate (7, 114, 116, 126, 178) as a result of the patient feeling misunderstood and not trusted (7, 164), and as a result of a past life as a healthy individual also being lost (184, 187). Social isolation as a result of a passive life can result in loss of contact with important social links (7), thereby limiting opportunities for confirmation (188). The diagnosis of mental illness can also mobilize some cultural stereotypes and negative prejudices about mental illness that have consequences for an individual's identity (147). p 27.

A review that has considered the importance of social identity and self-categorization for health (180), demonstrates that *social identification*, among other factors, is of importance for how we assess our own health and whether we seek, and accept, help for ailments. An example of this is how children with MUS reflect other family members with respect to the impact of the symptoms, and to the question whether help should be sought for treating the symptoms (131).

Social identity is also considered to be a basis for social support. Social support is often offered and accepted by those who share a sense of social identity. Shared identity is positive for work life, contributing toward greater satisfaction and preventing burnout (180). Patients with MUS often describe themselves as feeling alone, socially marginalized, and experiencing little support from others. This underscores the importance of individuals with MUS feeling acknowledged and experiencing their state as being legitimate.

Overall this shows that although the social group identity that individuals already possess affects their health behaviour and ability to cope with a condition, at the same time the symptom burden itself and diagnoses (or lack of diagnosis) will also affect identity.





## 7.0 Summary of results

### Article I

*Karterud HN, Knizek BL, Nakken KO. Changing the diagnosis from epilepsy to PNES: Patients' experiences and understanding of their new diagnosis. Seizure 2010; 19: 40-6.*

The purpose of the study was to explore the experience for patients of having their diagnosis of epilepsy switched to a diagnosis of PNES and how patients experienced their new diagnosis. Ten adult participants were interviewed and we investigated what was difficult and what contributed to coping. When this study was conducted, there were no established guidelines regarding how a diagnosis of PNES should be communicated to the patient.

According to our results, PNES is a difficult diagnosis to understand. When the causes of the seizures were unclear, this resulted in feelings of hopelessness and helplessness. The change from a neurological diagnosis to a diagnosis of PNES meant that the patients had to re-evaluate their self-understanding. The term "psychogenic" was immediately associated with severe mental illness. In this way, the diagnosis was considered by the patients as a threat to their identity. Patients felt that the healthcare services lacked expertise in their condition. With the change in diagnosis to PNES, the patients felt that responsibility was transferred from the health professionals to themselves.

Some patients also felt they were not sufficiently included in the diagnostic process. If the patients felt that the diagnosis was communicated to them in a categorical manner, it resulted in a lack of trust. This, in turn, made it difficult for the health providers and the patients to reach a common understanding of the disorder. Coping with the condition was strongly associated with understanding what caused the seizures. Other factors that contributed towards being able to cope included being taken seriously by the health providers and meeting other people with PNES.

The study concluded that changing the diagnosis of epilepsy to a diagnosis of PNES is a demanding process. The manner in which the patients are informed of the new diagnosis can be crucial for the patients' ability to cope.

A follow-up study was also conducted one year after discharge. Here we found that there were fewer who accepted diagnosis at follow-up, than when they were first informed about it. This could be related to the fact that the participants still did not understand why they had developed seizures.

## Article II

*Karterud HN, Risør MB, Haavet OR. "The impact of conveying the diagnosis when using a biopsychosocial approach: A qualitative study among adolescents and young adults with NES (non-epileptic seizures)". Seizure 2015; 24:107-13.*

The aim of the study was to explore the impact on participants of using a biopsychosocial approach to explain their non-epileptic seizures. Participants in the second and third study had attended a two to four weeks follow-up stay in which they were informed of their diagnosis in the light of a biopsychosocial approach. Eleven young people were interviewed. They were asked how they understood their condition from the time at which they were first informed about their diagnosis up until the end of the follow-up stay.

The results showed that the diagnosis may initially be perceived as threatening to their self-image. Most of the participants initially perceived their condition as being purely psychological in origin, as a result of being given the "PNES diagnosis". As they felt mentally healthy, they interpreted this as frightening and threatening. Participants had many previous experiences of being suspected by healthcare providers of staging their seizures. Some had even begun to have doubts themselves as to whether the seizures were voluntary or not.

The study also identified some factors that contributed to mastering. Explaining that unconscious processes are involved in NES resulted in the patients themselves believing that the seizures were real and not faked. When patients felt believed, they were able to develop the confidence and legitimacy that they needed to explore the associations between their life experiences and the development of seizures.

The participants needed an explanation of NES that made sense, and matched their self-image and illness perception. It seems like an understanding of PNES where many different factors are at play, helped the participants to develop recognition of their own lives. The study concluded that being believed was the most important factor for coping with the condition

### Article III

*Karterud HN, Haavet OR, Risør MB. Social participation in young people with non-epileptic seizures (NES): A qualitative study of managing legitimacy in everyday life. Epilepsy Behav. 2016; 57: 23-8.*

The aim of this study was to explore how young people with NES experience social participation in everyday life settings, with particular emphasis on how they manage the legitimacy of their condition.

Whereas the two first studies were based on the participants' encounters with the healthcare services and their understanding of the diagnosis, the starting point of the third study was the participants' social interactions with their environments (i.e., family, school, work, and leisure time activities).

According to our results, the participants' behavioural patterns could be divided into two main categories: "open" and "closed". Whereas some participants spoke openly about their condition and participated socially (i.e., open behavioural patterns), others tried to conceal their condition and withdrew socially (i.e., closed behavioural patterns).

Both the young people themselves, and their environments, tended to perceive NES as a socially illegitimate condition. The participants described frequent delegitimizing experiences from families, schoolteachers, colleagues, and employers. The delegitimizing experiences included, among others, evidence that other people hinted that they had more control over their seizures than they themselves expressed and were convinced about. For example, when employers and colleagues insinuated that it was up to the participants themselves to recover from the seizures.

Closed behavioural patterns: Fear of being exposed to delegitimizing events could result in the participants trying to conceal their diagnosis and this could result in social withdrawal. For some it resulted in isolation from all social arenas, apart from with their closest relationships. Concealing the diagnosis was understood to be a protection against stigmatization.

Open behavioural patterns: Support from close relationships was a protection against delegitimization and contributed towards greater social participation. The study also revealed a relationship between legitimacy of illness experienced by the participants and the extent to which they either participated or retreated socially. Those who had an illness perception that was personally meaningful experienced their condition as being more legitimate and thus participated socially to a greater extent.

The study concluded that fear of delegitimization because of a controversial diagnosis may result in young people with NES isolating themselves socially. Support from close friends and family, together with the participants themselves having a good illness perception, can be a protection against delegitimization, and thereby contribute to increased social participation.



## 8.0 Discussion of results

The main purpose of this study was to investigate patients' experiences with the "PNES-diagnosis". The discussion is organised around the overarching results in the articles and themes that are of relevance across all sub-studies. The themes are discussed in relation to recent studies that go beyond the references already included in the articles.

In the discussion of the study results, the terms illness perception and self-understanding are used. These are broad and comprehensive concepts. In this study, illness perception is restricted to meaning how participants understand their symptoms / health problems and their diagnosis. Similarly, self-understanding is, in this study, limited to referring to how participants understand *themselves* in the light of their symptoms and their diagnosis.

### 8.1 How to understand the challenges faced by patients diagnosed with PNES

#### *8.1.1 Contextual factors related to communication of the diagnosis*

As described in the literature review, communication of the "PNES diagnosis" is a critical point in the dialogue between a health worker and his/her patient. This is partly because it is considered important that patients accept the diagnosis as early as possible so that they are motivated for getting treatment. The question of acceptance of the diagnosis, and when and how the diagnosis should be conveyed, has therefore regularly been a topic in PNES research. This topic has also been part of this study, both as a starting point for the problem and as a common theme that often emerged via the empirical data. The discussion starts regarding contextual factors related to the patient's acceptance of the diagnosis. Factors relating to the context of the interview itself could also be discussed in the discussion of methodology, but are included here because they can clarify some of the premises that underlie patients' experiences with the diagnosis.

Although we did not ask participants directly whether they accepted the diagnosis, it was not difficult to obtain their perception of the disorder. Participants were asked how they understood their seizures both before and after they received the "PNES diagnosis" and how they

understood the explanatory models from the clinician. Some participants said that they agreed with the clinician's explanations, but others disagreed.

We found a discrepancy in the two groups regarding acceptance of the diagnosis the first time it was communicated. In the first group that we interviewed, eight out of ten patients stated that they *accepted* the diagnosis, whereas in the second group eight out of eleven patients said that they had initially *rejected* the diagnosis. One approach for understanding this difference was based on the context in which the interview occurred.

In group 1 the interview took place directly after the diagnosis was communicated, while the patients were still in hospital in the neurological department where the diagnosis had been made. The interviewers were employees at the same hospital, and this may have resulted in the patients wanting to stay loyal to the hospital diagnosis. However, the analyses showed that of the eight whom initially accepted the diagnosis, six had no idea of what could be the basis for their seizures. This lack of understanding of their symptoms may have engendered feelings of inferiority in the patients, and contributed towards a subordinate attitude. There were also examples of participants feeling guilty about their condition, and therefore taking responsibility for the condition itself. As a result of this feeling they reported that they withdrew from the consultation with the doctor without any understanding of the new diagnosis, and without offering objections. When the participants were interviewed again between one and two years later, fewer accepted the diagnosis, and there were still only a few who had some idea about causal relationships.

In group 2, however, the patients were interviewed at the end of their follow-up stay. They had received the diagnosis *before* admission to the hospital. These patients were thus describing their reactions to receiving a diagnosis that had occurred sometime back. Furthermore, the interviews in group 2 did not occur in the same place as where they had received the diagnosis. Thus, they may have felt less obligation to be loyal to those who had made the diagnosis, and it was therefore easier to disagree with it.

In another study (33) it was reported that more than one third of the patients still did not accept their diagnosis between 1 and 7 years after the diagnosis had been received. Among the patients (N = 84), 63 % did not have a good understanding of the diagnosis at the time of follow-up.



Most of the participants were unclear about the precipitating factors, and the most common reaction to the diagnosis was confusion (33).

In my opinion, this suggests that it may be unrealistic to expect that patients will accept the “PNES diagnosis” at an early stage. Many of the patients are left to themselves with a diagnosis that does not make sense to them, that they find unacceptable as time goes by, and they do not receive adequate assistance. In practice there is often a long delay between the patient receiving the diagnosis and treatment being started. This is unfortunate because an understanding of what might underlie the seizures is often only first realised during treatment (93). From a scientific perspective, I think that this means that studies in which different strategies for communicating the diagnosis are tried out should follow the patients over time in order to know with certainty how the communication format affects the patient.

The organization framework, in which the field is divided into psychiatry and neurology, has implications for the timing of communicating the diagnosis. The consequences may be that the patients are forced into a framework of understanding of their symptoms that does not agree with their perception of the condition, nor with their self-understanding. Communication of the diagnosis is therefore critical, and, in my opinion, an expectation that patients will accept the diagnosis early may cause some professional and ethical challenges.

### *8.1.2 Threatened self-understanding*

In the continuing discussion, the perspective moves from that of contextual conditions and the external framework for communication over to the patients’ own experiences. Here the discussion is based upon what might be at stake for patients when they are introduced to the PNES explanatory model of seizures.

Diseases and disease diagnoses may have impact on a person's identity. Charmaz (188) describes how people with chronic illness may experience a loss of self as a result of four factors: 1) Leading restricted lives, 2) Experiencing social isolation, 3) Being discredited, and 4) Being a burden to others.

Similar experiences are reported in individuals with MUS (7, 184, 187). Biographical disruption can be experienced via: self-image, social role, and loss of valued activity. Reductions in strength, activity level, and social participation could result in individuals feeling that they do not know themselves (187). A study of women with ME and FM found that the biographical disruption was particularly evident in connection with work and social life (7).

All these aspects were also highlighted by the participants in our study, but it was the significance that the actual diagnosis had for their self-understanding that was described as the biggest challenge, and to which space will be devoted in discussing the study results.

### **The participants understanding of the diagnosis**

As the interviewers were not present when the diagnosis was communicated, only the participants' interpretation of the diagnosis is described. Although the two groups had the diagnosis communicated in different ways and by different people, there were some similarities in their immediate experiences that will be discussed.

Some of the participants understood the "PNES diagnosis" as being a reaction to prolonged stress and as "the body's way of stating this", but most of them understood the seizures as being a reaction to *one* particular *repressed trauma*. They expressed this as "the subconscious remembered something that they had suppressed or forgotten, but which appeared via a signal from the body, that is, the seizures". This was a difficult explanation model to deal with, and the search for *one specific* suppressed stress factor or life event that could have caused the seizures, made it difficult to develop good coping strategies and made the participants feel incapacitated. Coping with the condition was strongly associated with identifying factors that could precipitate seizures. Nevertheless, eight out of ten patients in our first study believed that neither they, nor anybody else, could identify any specific stress factors (189).

### **Lack of meaning**

Although one purpose of a diagnosis is to provide a meaning for the symptoms, and a treatment route (173) it could appear that the "PNES diagnosis" was a burden rather than a help. The diagnosis often came unexpectedly, and many had believed that their seizures were epileptic. The change from a neurological diagnosis, to a more diffuse state such as PNES, resulted in

some existential questions arising that were related to experienced meaning and experienced legitimacy of illness. According to a meta-synthesis of patients' subjective experiences of FM obtaining a meaning in illness seems to be central for being able to cope (115). The association between a logical reasoning related to symptom understanding, and development of successful coping strategies, is also known from research on patients with medical problems (190).

The participants' understanding of the diagnosis for which the seizures are an expression of a repressed trauma, have some similarities with theories of conversion of anxiety into physical symptoms. Conversion disorder may refer to a disturbance of body function characterized by neurological, sensory, or motor symptoms for which known medical explanations do not explain, or fail to account for, the severity of the patient's impairment (191). Conversion *symptoms* include medically unexplained motor weakness, loss of other sensory functions such as touch, sight, or hearing, and non-epileptic seizures (also known as pseudoseizures). Symptoms are experienced by patients as involuntary and vary in severity from mild, transitory somatic concerns to chronic functional impairment. The diagnosis of a *disorder* requires that the symptoms are associated with significant distress or disability (191).

If PNES is a form of conversion in many cases, and part of a dissociative disorder, as indicated by various factors (192), this could have an impact on the patients' interpretation of their physical symptoms and their ability to understand the diagnosis. With dissociation, a "link" between symptoms and their cause may be missing. The individual thus has a limited ability for integration, and this can result in traumatic memories being avoided (193).

A recent study of youth with PNES (N = 55) suggest that PNES could be assumed to be a symptom of conversion disorder (47). Another study found that patients with PNES may tend to deny the influence of stressful life events (194). This may mean that there are some characteristics of patients with PNES, at the group level, which can contribute to the explanation that it is particularly difficult for this patient group to find a meaning in this diagnosis. This will not be discussed further here, because it lies beyond the framework of this study to explore the psychological mechanisms of PNES. It has, nevertheless, been mentioned here because it provides an important "backdrop" for understanding the patients' experiences with the diagnosis. Probably this is not of relevance for all, as people with PNES represent a

very heterogeneous group (79). In group number 2 in our study we observed that some of the youngest participants already had in mind what might underlie their seizures as soon as they received the diagnosis. They understood that the seizures could result from a difficult life-situation. These participants experienced diagnosis as a relief rather than a burden. This can be understood in the context that in children PNES is probably, to a greater extent, due to environmental circumstances (72). It is the experiences of those patients that describe the diagnosis as a challenge that form the basis for discussion in this current study.

### **Threatened identity as a result of a psychiatric diagnosis**

For patients who themselves had no idea of what might underlie their seizures, their condition was interpreted as unilaterally psychiatric. The basis for this understanding was a dualistic view of disease that divides illness into either somatic or mental disease, and for which only symptoms that can be explained by organic findings are classified as somatic disease (155). The participants immediately categorised themselves negatively, as a result of identification with a psychiatric diagnosis. The terminology contributed further towards the condition being interpreted as mental. Identification with the diagnostic label “PNES” was difficult, with the term “psychogenic” immediately giving associations with serious mental illness, and provoking a fear of being categorised as mentally ill. This was frightening, and it was difficult to understand that such a powerful physical symptom as seizures could simply be an expression of emotional difficulties.

It appeared as though the “PNES diagnosis” triggered some stereotypical beliefs about mental illness with which the participants could not identify. This can be understood on the basis of the concept that categorisation results in the formation of prototypes (183). These prototypes can increase the differences between the groups. Members of “out-groups” are not perceived as unique individuals, but as representatives of a particular prototype (183). The opinions were related to general categories of disease that are divided into physical and mental illnesses, and in which mental illness was associated with specific cultural values and assumptions.

We noticed that the patients used the terms “psychiatric” and “psychological” interchangeably, such that it seemed as though they were not really aware of which term they used. When they discussed how they understood the diagnosis, it seemed as though they meant to use the word

psychiatric, because they associated the condition with serious mental illness and hospitalisation in psychiatric clinics. Or it could appear as though it was important for them to distinguish between physical illness and mental illness.

The fear of being defined by a psychiatric disorder could be understood from two perspectives: on the one hand this could trigger a fear of “loss of self” (by which I mean a fear of losing their previous self-understanding). Simultaneously, however, a fear of “loss of social identity” was also triggered (by which I mean a fear of how others will define them). Both perspectives are concerned with the patients’ self-understanding as being mentally healthy was coming under threat.

Regarding fear of loss of self, it can be understood that the participants’ identity was being threatened due to a discrepancy between their own self-perception as being mentally healthy, and the diagnosis that was perceived as entirely psychiatric. This elicited a re-interpretation of the previous self-understanding, in which they wondered how they could understand themselves and their previous lives in a new way.

Regarding the fear of loss of social identity, this concerns the patients’ identity being threatened due to the assumption that others would define them solely on the basis of their diagnosis. This can be understood on the basis of Goffman’s concepts of actual identity and apparent identity. According to Goffman (1959), first impressions about another person encompass their *apparent* social identity; this first impression incorporates some normative expectations of the person’s character and qualities. However, a person’s *actual* social identity is how the person actually is, and their essential personality (1959). Stigmatisation can occur when there is a discrepancy between a person’s actual identity and their apparent identity (1959). The participants feared that their *actual* identity as mentally healthy people would be lost if others defined them in the light of their diagnosis or on the basis of behavioural changes that accompanied the seizures. The discrepancy between self-definition and definition by others, constituted a threat to identity.

Participants’ strategies for preserving their actual identity may illuminate what was at stake. Participants made continuous trade-offs in social contexts in relation to how much of their condition they would permit to be seen. The basis for their deliberations provides an idea of

what they thought their environments could tolerate without them losing their actual identity. It was perceived as less threatening to allow some seizures to be seen, than to tell others about the diagnosis. Thus, some participants took part in social events, but said that their seizures were epileptic, whereas others isolated themselves completely. Conversely, the study also demonstrated that those participants who had an understanding of their disease in which their identity was not threatened (see Section 8.2) were able to partake in social events.

The attempts of our participants to isolate themselves socially and conceal their condition can thus be understood as a reflection of their need to preserve their *actual* identity as mentally healthy persons. However, this may be an inexpedient strategy, because social isolation may impact negatively on their sense of connectedness with others (7, 195). Furthermore, social isolation limits the possibilities for positive self-validation (7, 188), and may cause a loss of self-esteem (188) such that their identity is further affected.

An even more radical way for patients to protect their actual identity was to reject the diagnosis. Resistance against the diagnosis may be understood as a shield against a diagnostic label with which the patients could not identify. This indicates that obtaining diagnoses of medically unexplained symptoms involves identity-forming processes, and may help explain why many patients may initially reject a diagnosis that cannot be explained by organic findings.

Although at the PNES-group level there may be some aspects that makes it particularly difficult for these patients to find any meaning in their symptoms, patients with other forms of MUS describe many of the same challenges associated with being diagnosed with a disorder for which the underlying mechanisms are difficult to understand (6, 32, 33, 81, 126, 128, 178). Studies concerned with other forms of MUS have shown that patients and clinicians often have a different understanding of the condition (3, 7, 124, 164, 196) and that this may have a negative effect on the patients' identity (185). There is therefore much to suggest that both patients with PNES and those with other MUS experience that their sense of self could be threatened as a result of their diagnostic explanatory models not making sense and because of associations with a psychiatric diagnosis.

### **Lack of experienced legitimacy of a diagnosis**

In the Section above I have discussed how identity can be affected by diagnostic explanatory models that are not consistent with the patients' self-understanding. The analysis also showed that identity was affected as a result of a lack of experienced legitimacy of the diagnosis. The absence of organic findings led not only to the participants being afraid of being defined as "mentally ill", but it also led to a fear of being defined as "not credible people". Both of these conditions incorporate some doubt regarding whether their seizures were "real" or faked.

Both the participants themselves and the people in their environments perceived PNES as a socially illegitimate condition. The young adults described ongoing delegitimising experiences from colleagues, school teachers, and family members. Several studies on patients with other types of MUS have found, as we also found, that the patients tend to perceive bodily ailments that are unexplained by disease, as being "not real" or as being "all in the mind" (114-117, 120, 122, 176, 195) and that the patients' identity can be challenged because the symptoms do not result in classification as a "medically defined disease" (126, 128, 178, 184). A recent study from South Africa of 10 patients with PNES found that association with a psychiatric diagnosis was a major challenge, because then the condition was not considered to be a "real" disorder (113)

Although MUS, including PNES, is characterised precisely by the inability to explain the symptoms by the traditional biomedical concept of disease, it appears that patients' illness perceptions in our study were influenced by such a model. According to the strictly biomedical view of disease, those conditions that cannot be classified according to a medically defined disease are perceived as solely psychological / psychiatric, or as "non-real" or "all in the mind" (164). The consequences of such a view of disease are that only those symptoms that can be explained by organic findings should be considered as understandable and legitimate.

Categorisation of health problems into purely physical and purely non-physical disease is an inappropriately reductive approach (170). Nevertheless, both the patients' view of disease and also the view of our society seem to be characterised by such an understanding. Patients understand PNES to be purely psychological, while epilepsy is understood to be an unambiguous physical condition. There was a lack of emphasis on the possibility that seizures

that are epileptic in nature can be influenced by psychological factors, such as anxiety, stress, worry, expectations, and tension (197 p 98), while our understanding of biological factors on conversion conditions is increasing (68, 198). It could appear that it was difficult to incorporate possible biological or neurological explanations into the disease understanding of PNES, and to take into account that the condition contains “unexplained” aspects. Our results show that the aetiology (i.e., whether or not the condition was caused by organic findings) was essential for the participants’ categorisation of the symptoms as physical or mental.

This simplified and dualistic understanding of disease reinforces the hierarchy in social debates and everyday interpretations of illness and health. Concepts such as psychogenic and psychosomatic or “pseudo seizures” are negatively charged models in specific social and cultural contexts. Mental health-related issues tend to be discussed in a stigmatising way in the media, although a study from England indicates that this has improved somewhat in recent years (199). A review from 2004 reported that the term “psychosomatic” was used disparagingly in one of three newspaper articles (200). The diagnoses are ranked differently and have a different status (174, 175). Diseases that cannot be associated with any particular location in the body and conditions with unclear treatment models, ranked low (174, 175). There is, thus, also a moral aspect related to the diagnoses, and that had an impact on the patients’ identities in our study.

To summarise: the basis for the participants’ interpretations of their condition was a dualistic view of disease, in which disease is divided into two categories, physical or mental. The extent to which the aetiology could be asserted by organic findings was crucial. Symptoms that could not be explained by organic findings (i.e., PNES), were understood to be an expression of unilateral psychiatric illness, as “not real”, or as simulated. This triggered a series of stereotypical concepts associated with mental illness, and with which the participants could not identify. Their identity was threatened in two ways:

- 1) As fear of loss of self, because there was a discrepancy between their self-understanding as psychologically healthy and the explanatory model of the diagnosis as entirely psychological.
- 2) As fear of loss of social identity, because they feared that others would define them solely on the basis of their diagnosis.



## 8.2. Being believed – the turning point

The study identified some factors that contributed to successful coping. This section is based primarily on the population of patients who were admitted to the follow-up stay. We observed that most of the participants altered their understanding of their disease during the course of their stay; the diagnosis was altered from being perceived as a threat to their identity, to an understanding of the illness that gave greater meaning and was experienced as being more legitimate.

Our first sub-study clearly demonstrated that successful coping with the condition was strongly associated with understanding those factors that triggered the seizures. Other studies have described that patients with PNES who were assisted in integrating their personal life events into the diagnosis had greater abilities to cope (6, 32). One way of understanding this may be that the threat to identity diminished by integrating the causal relationship the patient's self-understanding. One purpose of the follow-up stay was that the diagnosis could be explained to the patients using a multifactorial model in which biological, social, and psychological factors may play a role in establishing the predisposing, precipitating, and maintaining conditions. An unexpected finding was that *being believed* constituted the turning point in the patients' coping with the condition. When participants felt believed, increased self-reflection was elicited, such that it was possible to change the previously stalled understanding of the illness.

The young adults used the terms to be believed and understood interchangeably. It could appear as though they were in search of a deeper recognition of themselves as people. Lind (201) writes that individuals with MUS have difficulties with self-recognition of bodily sensations. We found that the patients themselves had doubts about whether their seizures were produced voluntarily. This could be understood as being the doubts raised by others being “transferred” onto the patients. At the same time the patients themselves also thought that symptoms that could not be explained by organic findings were “not real”. They therefore needed confirmation that what they actually experienced in their own bodies made sense.

Thus, being believed did not primarily mean that the clinician had to agree with the patients regarding the *causes of the seizures*, it meant that they needed to be believed that the attacks

were not deliberately produced. It meant that they were seen and understood as the persons that they are, without being defined solely on the basis of their disorder. The analyses indicated that there were two particular factors that contributed to the participants feeling that they were believed.

*The first factor* was concerned with psychoeducation, in which it was explained that PNES was due to unconscious mechanisms. A deeper and more nuanced understanding of the mechanisms of the condition made it possible to believe that the attacks were not produced voluntarily. Through illness explanations in which the disease exists in the body, the participants were able to adjust their earlier understanding of illnesses in which only those symptoms that can be explained by organic findings are “real”. This was experienced as a turning point. One way to understand why this was so fundamental is on the basis of those consequences they experienced when they were *not* believed. In a simplified manner, this could be expressed as that when they felt that they were not trusted, their identity and self-understanding was hurt because their *actual* identity (see p 76) as mentally healthy and credible people was being threatened.

In contrast, the experience of being believed made it easier to preserve their actual identity as credible and mentally healthy people. In other words, being believed supported the patients’ self-definitions. How the patients defined themselves seems to be crucial for coping with the condition. This can be understood on the basis of that in terms of identification, what we think about ourselves is more important than what other people think about us, even though both these aspects are always of relevance (183). Kathy Charmaz writes that chronically ill people who move beyond loss and stigmatisation, define themselves as being much more than just their bodies, and as much more than just an illness (202). In being believed, the participants no longer simply defined themselves through their diagnosis. This was expressed as: *this is a condition; I am not like this*.

The participants described how feeling believed enabled a sense of security and increased self-reflection. This, in turn, allowed the participants to explore associations between stressful life events and bodily symptoms.

*The second factor* that contributed towards the patients feeling believed, was concerned with the relationship between the clinician and the patient. This meant that health professionals stated

that they believed that the attacks were real, but also that these professionals *saw* and confirmed the youngsters as being the persons that they believed that they were, without the “PNES diagnosis”.

A recent study of patients with PNES (203) confirmed that health personnel play an important role in relation to patients’ subjective interpretation of their symptoms. The study showed that a good relationship with the health personnel is essential for patients’ ability to cope with the diagnosis (203). Health professionals have the power of definition because they possess expert knowledge of the patients’ suffering. The extent to which they indicate that they “believe in” a patient or not, may therefore have an impact on an individual patient’s self-image and understanding of illnesses. Most of the participants in our study had experiences of not being trusted by healthcare professionals. This became apparent by the health professionals giving the impression that the patients had more control over their seizures than the patients themselves expressed. A recent survey from USA (204), that was commissioned by American Epilepsy Society (AES), investigated how experts in epilepsy (N = 133) from level-4 epilepsy centres in USA communicated the “PNES diagnosis” to their patients. The study demonstrated that 86/133 health professionals told their patients that the seizures “are not real seizures”. That the seizures are not “real” can involve the consequent assumption that “then they are faked”. Similar attitudes have also previously been described in other studies (94, 111, 112) writes that the patients probably know whether the doctor believes them or not.

Patients with other MUS have also described that it is essential to feel believed (205). Although there may be some characteristics of people with PNES at the group level, that may influence their interpretation of their symptoms, it seems likely that people with other MUS have the same need to be believed, and also the same need to preserve their actual identity when faced with their diagnosis. A review of patients’ experiences with FM, reported that patients found it a relief when the doctors made their diagnosis from physical examinations, because they then understood that the disease existed in the body (173).

A biomedical concept of disease, in which disease is conceptualized as being due to abnormal functioning of some bodily system (154, 155), appears to underlie both health professionals’ and patients’ understanding of illness. From such a perspective it can be difficult to understand

that the seizures can really be beyond the patients' control, when the condition cannot be classified according to a specific medical diagnosis.

The young people who participated in our study were in a situation in which several things happened at once. They were provided with psychoeducation regarding there being unconscious processes involved in PNES, they received a diagnosis that was explained in the light of a multifactorial / biopsychosocial model, and they were in a specific setting over a prolonged period with a relation to health professionals and other patients with the same diagnosis. Furthermore, the time factor regarding the point at which they were first told about the diagnosis may also have affected the development of their disease understanding. Thus, there were probably several processes that interacted with each other and set in motion a positive cycle. Seen in the light of Jenkins's theory of social identity (183) we always identify ourselves in relation to others - as similar to, or different from, others. There is also a continuous movement between how an individual identifies him/herself, how an individual identifies others, and how others identify the individual (183). It may therefore be difficult to say anything definitive about how psychoeducation about PNES would, in itself, alter the participants' understanding of illness. Nevertheless, the empirical data clearly showed a step-by-step process, in which being believed was the basis for the forward-moving processes.

### 8.3. What about the biopsychosocial approach?

The intention behind the follow-up stay was to help the patients understand the "PNES diagnosis" in a more comprehensive manner. A biopsychosocial approach seems to support a more comprehensive understanding, more than a traditional biomedical-oriented medicine (157). Based on this, and on an article that suggested that the etiology of PNES could best be understood in light of a biopsychosocial model (35), it was decided at SSE that a biopsychosocial approach would be used during the follow-up stay.

The aim of the sub-study II was to explore *the impact on participants of using a biopsychosocial approach to explain their non-epileptic seizures*. In retrospect, it is clear that sub-study II has several methodological weaknesses. These diminish the possibility of making

assumptions about the importance of a biopsychosocial approach on participants' understanding of their illness. In this section, I discuss some of these methodological shortcomings, the practical aspects of a biopsychosocial approach, and the impacts that these may have had on the results.

The first limitation that I would like to highlight is associated with how when using the biopsychosocial approach to communicate the diagnosis, more details should have been available. Better guidelines would have ensured the quality of this communication.

Nevertheless, several efforts were made in advance of the study to ensure a common understanding of a biopsychosocial approach:

- The Department of Psychosomatics and consultants in child psychiatry at Oslo University Hospital transferred their knowledge about a biopsychosocial approach to staff at SSE. They have worked towards facilitating the use of a biopsychosocial approach for clinical use with children with medically unexplained symptoms, including children with PNES, for a long time. In 2011, consultant Helene Gjone gave a 90-minute lecture at the leader-group meeting at SSE on this topic, with doctors and psychologists from SSE invited. The same lecture has been given at the annual external courses arranged by SSE. Personnel from SSE (doctors, nurses, social workers, occupational therapists, physiotherapists and teachers) have attended these courses. (The PowerPoint presentation from Helene Gjone lecture is appended, see appendix 3).
- In 2011, a consensus document was developed for *Organization of services for PNES patients at AKE-SSE*, which states that «a biopsychosocial model is considered as the most appropriate model for providing the diagnosis for most patients».
- The year before the study began, the "PNES-teams" at the various wards at SSE were offered training in new patient care, founded on a biopsychosocial approach. The participants were primarily nurses, but other disciplines, such as social workers, physiotherapists, and doctors, also attended the training. The training consisted of ten

two-hour training sessions during the year. The content of these training sessions is outlined briefly below:

1. Education about how the aetiology of PNES can be understood in the light of a biopsychosocial approach (35).
2. Guidelines for the follow-up stay were reviewed (this is also described in Article II and in Section 4.3). According to these guidelines, initially there should be several discussions between the doctor and nurses with the children and young people and their families. These discussions should use an open approach, in which the basis is the patient's own perspective of their illness. During these initial conversations, an effort should be made by the healthcare providers to map the various different biological, social, and emotional stressors that could underlie the patient's seizures. The relevant stressors should then be written on a blackboard (in collaboration with the patient), and arranged according to predisposing, precipitating, and perpetuating factors. These lists should then form the basis for new discussions between the patient, nurses, doctor, and relatives. The intention of this approach is to create a greater meaning. At this stage, education about stress should be provided and how the mind and body can affect each other. The participants were also given a brochure about PNES (Appendix 4).
3. The training also included case reports. The staff presented patient cases, so that their experiences with the new approach could be shared with the training participants.

In retrospect, it is apparent that providing a precise manual with a strictly structured representation of the biopsychosocial approach, could have contributed to the communication of the diagnosis becoming more uniform.

Implementation of new approaches to patient care takes time. Although the training for health workers contributed to a common understanding, it must be assumed that in the empirical phase, the role and approach of the nurses, doctors, and physiotherapists was largely dependent upon the specific person. A further limitation was that the adult department (4-week hospitalization) had progressed significantly further than the paediatric ward in developing a structured content of the follow-up stay. The older participants were provided with a broader range of services, as described in Section 4.3 (The PowerPoint presentation that was used during the 4-week stay is appended, see Appendix 5).

A third limitation that made it difficult to fulfil the objectives of Article II, was that we did not have a systematic overview of everything that directly or indirectly affected the participants' understanding of their illness during the follow-up stay. As described in Section 8.2 above, several factors may have influenced the participants' understanding of the illness. The relationships that the participants had with health professionals and other patients, the group sessions, the teaching that they received, and the different amounts of time that they had to process the diagnosis, may all have had an impact.

A fourth limitation is related to the sample. When we stopped the inclusion of patients we had the impression that we had a rich and diverse material that was large and varied enough to address the purposes of the study. However, during the analysis it was clear that the large age span meant that there were few participants with respectively 2-week and 4-week admissions.

The analysis gave the impression that the youngest children already had formed an opinion regarding what could be the basis for the seizures before they attended the follow-up stay. Thus, these children may perhaps be less affected by the use of a biopsychosocial approach to explain the diagnosis. On the other hand, the wide age range was able to demonstrate different patterns among the older and younger patients. This underscores that children and youth with PNES are a very heterogeneous group, including with regard to the need for training and knowledge dissemination.

We chose to include participants consecutively, as they met the established inclusion criteria. A consecutive model means that participants that match the inclusion criteria are continuously included. As the participants had completed the follow-up stay, we hoped to obtain information that addressed the goals of the study. Participants in the 4-week stay had been through a prior screening process, because the admissions office had selected patients who they believed would benefit from participating in a group, in which the purpose was to share their experiences with others and reflect on their understanding of illness. It is possible that including a similar selection for the paediatric ward would have resulted in even the youngest participants providing more information about the biopsychosocial approach. A broader variation in the choice based on who showed themselves to be most information-rich, and a sampling strategy that resembled theoretical sampling, could have strengthened the study.

In summary, this shows that the study has several methodological limitations that make it difficult to explore the impact of using the biopsychosocial approach. Instead, in Study II we achieved greater insights into how the participants experienced their diagnosis subsequent to the follow-up stay. These experiences indicate topics that may be of relevance for future studies. This is particularly pertinent to the meaning of being believed, and the importance of obtaining an explanation that makes sense.



## 9.0 Conclusions and clinical implications

The purpose of the study was to expand our knowledge regarding patients' experiences of being diagnosed with psychogenic non-epileptic seizures (PNES). Sub-study I demonstrated that the "PNES-diagnosis" may constitute a threat to self-understanding. Sub-study II showed the importance of being believed and to get an explanation that makes sense. Sub-study III pointed out some connections between the participant's illness perceptions and social participation.

On the basis of these results, we were able to draw the following conclusions and practical implications.

### 9.1 Conclusions

- The immediate interpretation of receiving a "PNES –diagnosis" seems to be influenced by a dualism that separates bodily symptoms into two categories: physical and mental.
- On the basis of the PNES condition not having a physical explanation, the patients initially tended to interpret the condition as unilaterally mental.
- The association with a psychiatric diagnosis might lead to identity-threatening processes and contribute to lack of meaning and lack of experienced legitimacy of illness.
- Both the participants themselves and their close family members tended to perceive PNES as an illegitimate disorder, on the basis of the diagnosis not being a "medically defined disease".
- One strategy for tackling delegitimization associated with not having a "medically defined disease" was to conceal the diagnosis, or to become socially isolated.
- Successful coping with the condition was strongly associated with a personal conviction that the seizures were not willingly produced.
- Explanatory models that highlight that symptoms may have physical correlates, despite not being a part of a medically defined disease, might contribute to a feeling of being believed and understood.

- Feeling believed enabled the patients to interpret their symptoms as a legitimate part of their lives. This, in turn, made it easier to make sense of associations between biography and biology.
- A personally meaningful understanding of the condition, together with perceiving the condition as legitimate, may contribute to increased mastering of the condition and also increased social participation.

## 9.2 Clinical implications

The healthcare services continue to be characterized by structural and conceptual limitations that help to maintain the separation between psyche and soma. Initially in the discussion chapter (8.1.1), I clarified the problem of having a diagnosis of PNES made by the neurology healthcare services, whereas the treatment occurs in mental healthcare. The study confirms that it is a demanding process for patients to adapt to the diagnosis. A practice in which the external framework “forces” patients to accept their diagnosis as early as possible is therefore unfortunate. Patients need to have a period in which they have the opportunity to try out different ways of understanding for their seizures, along with healthcare personnel and without any pressure to identify causal factors.

For the general population, it is probably easier to think in dualistic terms, psyche and soma, than the more complicated understanding that is being increasingly used by health professionals for those conditions that are currently considered to lie at the interface between mind and body. However, with greater knowledge about these conditions, it is easier to understand how they may arise and the biological mechanisms that occur. This knowledge should be transferred to the patients.

The study has brought forward some aspects of patients’ perception of their condition, which, as far as we know, have not been previously described. These are concerned with the association between being believed and greater self-reflection, and the relationship between illness perception and social participation. This has implications for practice in terms of how clinicians can help patients with PNES experience their condition as being meaningful and legitimate. On

the basis of this, this study provides grounds for the following specific advice for clinical practice. The implications are directed both towards MUS in general and PNES specifically.

- Clinicians should be aware that the “PNES –diagnosis” can trigger identity-threatening processes, and take this into account.
- Clinicians should be aware that if the symptoms cannot be explained by organic findings, patients often interpret their symptoms as “not real” or as an expression of unilateral psychiatric illness. It is therefore important that a clinician relies on the patient’s subjective experience of symptoms as being real. Importantly, this does not mean that the clinician must agree with the patient regarding *causal explanations* for the symptoms.
- The explanations for MUS should be based on models that go beyond the concept of body-mind dualism.
  - Terminology and a conceptual system that divide symptoms into physical and mental categories (such as “physical cause” and “psychogenic seizures”) should be avoided
  - Clinicians must explain the mechanisms behind PNES / MUS as far as possible. In cases where the PNES are expressions of dissociation, psychoeducation about the mechanisms of dissociation should be provided. (Explanations showing there are unconscious processes involved in PNES, may contribute to better coping).
- Clinicians should explain, recognize, and legitimize an understanding of how a "lived life" leaves traces in the body as an introduction to the treatment.
- An attitude of what is “unexplained” today can be “explained” with time, should be fundamental when meeting patients with medically unexplained symptoms.
- The healthcare services must strive to develop more continuity in patient care for patients with PNES, and involve personnel from both neurology and psychiatry.

### 9.3 Recommendations for further research

Based on the study results the following themes and problems are recommended for further research:

- Qualitative studies that explore the impact a “disputed” diagnosis has for patient’s identity.
- How can the healthcare services assist with educating the general public to improve their understanding of MUS and prevent symptoms from being categorized as *either* physical *or* mental?
- How can the biopsychosocial model in this field be further developed? This implies particular research on the biological aspects of disease conditions that currently cannot be explained by organic findings, but also research that can help conceptualising and operationalizing the model even more to be an efficient tool in clinical practice.
- How can the organisation of the healthcare services be modified such that patients with MUS are increasingly offered a coherent patient pathway and a “multi-professional” environment?

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## **Appendix 1: Interview guide sub-study I**

- 1) Can you tell me about your symptoms and experiences that have resulted in you seeking assistance from the health services?
- 2) How have the health services interpreted your seizures up until you were admitted to the Norwegian national Epilepsy Centre?
- 3) How have you yourself understood your seizures before you were admitted to the Norwegian National epilepsy Centre?
- 4) Can you tell me what you thought when you heard that your seizures could be due to PNES?
- 5) Can you explain what you understand by the diagnosis PNES?
- 6) How do you understand your seizures based on the diagnosis you have now received?
- 7) Can you say something about how it feels for you to have received a diagnosis of PNES?
- 8) Do you think there is a difference between how you previously understood your seizures and how you understand them now?
- 9) Do you feel that some of the experiences associated with your current admission to the Norwegian national Epilepsy Centre have been positive or negative with respect to the diagnosis that you have received?
- 10) Can you say anything about your current thoughts on the future with respect to your seizures?
- 11) Have these thoughts changed from those that you had before you received a diagnosis of PNES?
- 12) Have you any thoughts on what the health services should do for you following your discharge from the Norwegian national Epilepsy Centre?

Now I have asked about the subjects that I was wondering about, is there anything else you would like to talk about? Is there anything that you think is important that I have not mentioned?





## **Appendix 2: Interview guide sub-studies II and III**

### *Understanding of the condition prior to the diagnosis being communicated:*

- Can you tell me about your first seizure?
- Do you have any ideas about why you had the first seizure; was there anything in particular that triggered it?
- What happened after you had this seizure? (where did you get help?)
- How have your seizures been since then? How often do you have a seizure, what are the circumstances in which they occur? How have the attacks developed?
- Can you recall what you thought about the seizures back then? (How did you understand them, what did you call them?)

### *Understanding of the condition after the diagnosis was communicated:*

- Now you've just had a chat or a conversation with a doctor and nurse about the seizures. Can you tell me what they said? How does this fit in with the way that you understand your seizures?
- How do you think the conversation went? Was it as you had expected?
- Can you remember what thoughts went through your mind during and after this conversation?
- How did you feel after the conversation?
- Have you talked to others about the conversation? Who and about what?
- What do you think about what others say about your seizures?
- Do you know what others think about your seizures (parents, teachers, friends)?
- How do you yourself understand the seizures now? (why do you think they come?)
- How did the doctor and nurse say that the seizures should be treated? What do you think about that?
- They call your seizures non-epileptic seizures (NES); what do you understand by NES?
- What do you yourself call your seizures now?
- How do you explain to others about NES if they ask?
- Do you know anyone else who has NES?

*Understanding of the condition in the light of a bio-psychosocial model:*

- Do you have any ideas about what happens in your body when you have a seizure?
- Can you think of anything that affects your seizures, such as school, home, friends or something else?
- Could how you are feeling physically have some effect, for example, if you are sleepy or tired?
- Does it matter how you are feeling emotionally? If you are feeling sad, scared, or angry, for example?
- Is there anything else in your life that you think may affect your seizures?
- Do you have, or have you previously had, seizures in some special situations?

*Something to live with:*

- Is there anything you find particularly difficult about having seizures?
- Is it the seizures in themselves that are the biggest challenge (or is it the reactions of other people, the social consequences of the seizures, the cause, the explanation problem for others, etc.)?
- Do you have any thoughts about what might be helpful for your progress?
- Is there anything you yourself can do to prevent the seizures?
- Is there something your family can do to be supportive?
- How do you think your life would be without seizures?

Now I have asked about the subjects that I was wondering about, is there anything else you would like to talk about? Is there anything that you think is important that I have not mentioned?

## Appendix 3: PowerPoint presentation from Helene Gjones lecture



## En helhetlig biopsykososial tilnærming ved behandling av PNES – hvordan bruke den i praksis?

Helene Gjone  
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Seksjon for psykosomatikk og CL-barnepsykiatri (BUP)  
Avdeling for nevrofag  
Kvinne og barneklivnikken  
OUS, Rikshospitalet

Helene Gjone, Seksjon for Psykosomatikk og CL-barnepsykiatri, OUS Rikshospitalet

## Ved seksjon for psykosomatikk og CL-barnepsykiatri, en integrert forståelse:

- et kart hvor det alltid må tas hensyn til både biologiske, psykologiske og sosiale... og kulturelle forhold
- forståelse > modell

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## Barne- og ungdomspsykiatrisk seksjon, RH

### HISTORIKK:

- 1884: Første Barneavdeling (RH)
- 1950: Første Barnepsykiatriske avdeling (BK, RH); sengeavd.
- 1963-68: Statens sentralteam og SSBU (Statens Senter for BUP)
- 1975: Statens sentralteam (1963) m/ tverrfaglig CL-team gitt RH
- 1979: BUP-seksjon, Barneklivnikken, RH opprettes

### Avdelingstilknnytning:

- ✓ Barneklivnikkens poster og poliklinikk
- ✓ Barnekirurgisk
- ✓ Barnereumatologisk seksjon
- ✓ Fortløpende henvisn fra alle andre avd som beh barn/unge

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## Seksjon for psykosomatikk og CL-barnepsykiatri, OUS-Rikshospitalet



### FUNKSJON:

*"Integrert del av RH's tilbud øremerket alle barn og ungdom (0-18 år) og deres familier, innlagt for somatisk lidelse eller somatisk symptomatologi"*

- Hovedvekt på lands-, flerregionale funksjoner

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- De fleste av våre psykosomatiske pasienter er sec.op.saker
  - et langvarig forløp før henvisning
  - viderføres til lokal oppfølging

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## Leting etter én årsak...et naturlig utgangspunkt:

- Barnet har anfall – hva er årsaken?

*Kanskje det er mange forhold som innvirker?*

- Den bio-psyko-sosiale forståelsen i praksis – å organisere en forståelse mhp reorientering som grunnlag for bedring

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## Faglig fundament

### **Helhetlig, multifaktoriell sykdomsforståelse**

*Komplekse interaksjoner mellom ulike biologiske, psykologiske og sosiale faktorer i forståelsen av predisponerende, utløsende og vedlikeholdende faktorer ved all sykdom*

### **BIO-PSYKO-SOSIAL sykdomsforståelse;**

utredning, behandling og oppfølging

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## En bio-psyko-sosial forståelse:

- helhetlig;
  - Individet er udelelig
  - Ingen tanke uten transmittorsubstanser
  - Nevral plastisitet og erfaring
- ivaretar det multifaktorielle
  - ramme for utredning
- inkluderer ulike teoretiske og empiriske innfallsvinkler
- omfatter alle aspekter av sykdom
- utgangspunkt for
  - kommunikasjon med familie
  - valg av terapeutiske intervensjoner

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## 2. Det biopsykososiale perspektivet på psykosomatikk:

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## Somatiske symptomer uten organiske funn; kroppens språk

1. manglende bevisst integrering av livsbelastning/traumer - dissosiasjon
2. manglende språk for emosjoner og livsbelastning
3. (familie) systemet tillater ikke at emosjoner/belastning uttrykkes
4. sosial læring - barnet formidler familie-systemets språk
5. det ensidige kroppspråket gir sensitisering

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## Mål i fht psykogene/dissosiative anfall:

- Oppnå en forståelse hos pasient og familie som kan bidra til å bringe pasienten ut av anfalls-situasjonen gjennom;
  - endring av situasjon som har utløst og/eller opprettholder anfall
  - øket støtte/ tilrettelegging i fht anfallsdisponerende sårbarhetsfaktorer
  - utviklingen av/åpning for et mer direkte språk for vanskelige situasjoner og følelser

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## 3. PNES og dissosiasjon

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## F44 Dissosiative (konversjons) tilstander

Generelle kriterier:

- G1. Det må **ikke være holdepunkter for en fysisk tilstand som kan forklare** det denne tilstandens karakteristiske symptom (selv om fysiske tilstander som gir opphav til andre symptomer, kan være tilstede).
- G2. Det er **overbevisende sammenhenger i tid mellom start av symptomer på tilstanden og stressende/belastende** hendelser, problemer eller behov.

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## Hvor mye stress/traumatisk belastning skal til for å utløse en dissosiasjon

Genotype (A) konstitusjon (B)

+/x

Livserfaring, livsbelastning

Biologisk miljø pre- peri- og postnalt

Tilknytning (C) stimulering, utviklingsmessige rammer

Akutte traumer, kronisk stress (D)

=

Individuell variasjon/fenotype

A,B,C,D avgjørende for dissosiasjon

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## Hos oss

- F44.4. Dissosiative motoriske forstyrrelser
- F44.5. Dissosiative kramper:
- F 44.7 Blandet dissosiativ (konversjons) tilstand
  
- F44.4, F44.7 mer vanlig enn rene motoriske anfallstilstander (F44.5)
  
- Ser 44.0-F44.3
  - som del av blandingsproblematikk
  - som del av forsvar i fht belastninger relatert til somatisk sykdom og belastende prosedyrer
  - obs spedbarn

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## Hva sier vi til pas med konversjonssymptomer?

1. Innledningsvis undrer vi oss sammen...

– Dette er ikke så lett å forstå...

2. Så begynner vi å lete etter hva pas ellers opplever, for å bli kjent med ressurser og mestring og utfordringer..og litt etter litt sier vi at:

– *Noen ganger kan kroppen ha et språk som det er viktig for oss å høre på.. kan det være at kroppen din si fra om noe....?*

3. Og når vi har arbeidet sammen nok til å etablere tilstrekkelig tillit, kan vi ta et skritt til siden og si at

– *Noen ganger så kan følelsene "blokkere" sånn at hjernen ikke får de signalene som gjør at vi føler og beveger oss ;*

Da har vi etablert en trygghet og en forståelse og kan åpne for arbeid med underliggende problem(er)

uavhengig av dissosiasjonen

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## 4. I denne prosessen: praktisk bruk av en bio-psyko-sosial tenkning

- Terapi begynner med det første møtet med pasient og foreldre:
  - Romme angst og bekymring
  - Ta kroppen på alvor
  - Gi tid til å høre familiens bekymring og forståelse
- Felles møte med BUP-team og somatisk lege/team
  - Trygge helhetsspektivet; redegjøre for tilnærmingen
  - Sikre at vi snakker om det samme
  - Unngå demotiverende gjentakelser
  - Sikre at vi trekker i samme retning mhp å bidra til forståelse

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## Anvendelse av en bio-psyko-sosial forståelse

- for å organisere en multifaktoriell sykdomsforståelse i komplekse saker
- sjekkliste/mal for utredning
- ramme for tilbakegivning og utveksling av forståelse med pasient og familie – kan bidra terapeutisk til kognitiv og emosjonell restrukturering
- grunnlag for terapianbefalinger og planlegging av tiltak

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## En bio-psyko-sosial sykdomsforståelse

1. Predisponerende/sårbarhetsfaktorer
  2. Utløsende faktorer
  3. Opprettholdende/modererende faktorer
- Dvs multifaktoriell sykdomsforståelse hvor både 1,2 og 3 kan inkludere biologiske og psykososiale faktorer

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## En bio-psyko-sosial forståelse predisponerende faktorer:

- Biologisk
  - kjønn, alder
  - genetik: generell utvikling/temperament og spesifikk sårbarhet
  - pre/perinatale forhold
  - somatisk sykdom/historie
- psykologisk: relasjonelle faktorer med innvirkning på
  - mental helse
  - psykososial fungering
- Sosialt
  - familiens rammebetingelser; historie, sosioøk.forh, sykdom
  - barnets øvrige rammebetingelser; skole, fritid/venner

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## En bio-psyko-sosial forståelse - utløsende faktorer

- Biologisk
  - egen somatisk sykdom eller skade
  - Aldersrelaterte endringer
- psykososialt (det uoverkommelige stresset):
  - traumer/overgrep ex SO, mobbing
  - familiære belastninger ex konflikt, skilsmisse, sykdom
  - andre livshendelser ex flytting, skoleskifte
  - vanskelig skoletilpasning el. andre endringer i krav forventinger utover individuelle forutsetninger

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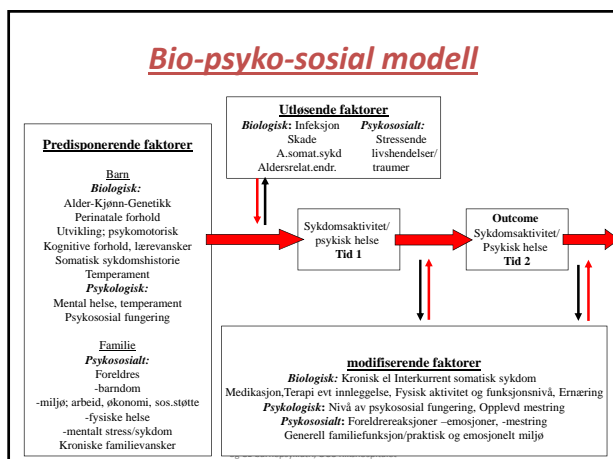
## En bio-psyko-sosial forståelse - modererende faktorer (=utg.pkt for behandl):

*Påpek selvforsterkende systemer eller onde sirkler med vekt på hvordan de kan brytes – avpass etter barnets/familiens fokus*

- Biologisk
  - selvforsterkende somatiske symptomer og symptomhåndtering
  - annen somatisk sykdom
- psykososialt
  - individuell og familiær sykdomsforståelse
  - familiesamspill relatert til sykdom
  - pågående traumer/overgrep
  - andre belastende livshendelser
  - sosiale vansker /evt sekundært til sykdom
  - skolevansker /evt sekundært til sykdom

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## Bio-psyko-sosial modell



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...kan noen fortelle meg hva som er psykisk og hva som er fysisk.....?

Vi er en helhet....  
Individet - udelelig



Likevel kan det være nyttig å sortere ..så vi lettere kan se sammenhenger....

	Predisponerende	Utløsende	Modererende/opprettholdende
Tid			
Kroppen			
Følelser og tanker			
Familie/ Skole/ venner			
Resurser			

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## å dele opp og sortere...i et helhetsperspektiv

### Å bruke tavle

- Å oppsummere kompleksitet der leting etter en årsak ikke har ført frem
- Å etablere et felles ytre fokus
- Å åpne et rom for samarbeide om å tenke prosesser/utvikle et felles språk
- Å utvikle en samlet formulering som grunnlag for videre tilnærming/behandling
- Å ramme inn utfordrende spørsmål

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## Å skape en forståelsesmessig og emosjonell reorientering

- Forutsetter god kartlegning av aktuell situasjon og bakgrunn
- Obs faktorer som virker med og mot reorientering?
- Tavlen fungerer oftest svært godt...

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## En bio-psyko-social sykdomsforståelse

Aktuelt problem ex 1: kramper uten påvist fysisk årsak ex 2: anfall med fjernhet/bevissthetstap

predisponerende/ sårbarhet	utløsende	modererende/ opprettholdende
tid		
biologisk	biologisk	biologisk
psykologisk	psykologisk	psykologisk
sosialt	sosialt	sosialt
kultur.....		

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## 5. Å møte familien å starte en terapeutisk prosess

- Helst i første møte med somatiker
- Hvorfor er BUP her?
  - presentere oss med vekt på at vi har møtt mange barn/unge som har strevet med sykdom hvor legene ikke har funnet ut av det: stressomt for barn og familie
- Hva er problemet?
  - Barnets opplevelse
  - Foreldres opplevelse
- Hvordan forstås problemet?
- Forventninger og behov i fht opphold og utredning?
- Avpasse etter hvor familien er hen i sin forståelse/erfaring når vi spør om erfaring med tidligere BUP-kontakt og tanker om hva vi kan bidra med??
  - Trygge på at det ikke er enten eller men en helhetlig bio-psyko-social forståelse som ligger bak. (På sykehuset tenker vi alltid .....). Å legge frem hva vi kan tilby og evt. høre hva de vil bruke?

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## Så...

- Felles plan for oppholdet
- Videre samtale med de ulike instanser hver for seg..

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### 5.1. Familiemøtet: Ta det somatiske symptom på alvor og konkluder ikke for tidlig

Bruk magefølelsen og la roen senke seg i samtale(r)

- Bruk tid på symptomforståelse
- Sikre kontakt med somatiker
- Sikre at de får tid/undersøkelser
- Målbær deres spørsmål sammen med dem i fht somatiker
- Samtal om om kroppens reaksjoner på både fysisk og psykisk stress og hvordan dette kan sette seg i kroppen
- Noen kan nå spørres om barnets situasjon og om mulig stress-faktorer

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### 5.2. Familiemøtet : Familiehistorien

- Hvordan har sykdommen innvirket på barnet? Familien? Søsknen?
- Ha genogrammet i hodet eller bruk papiret: Å bli kjent med dere. (Hvor familien er avgjør om man starter ressursorientert eller sykdomsfokusert):
  - Tegn opp primærfamilien, evt utvid eller avvent: ta med både folk og fe..
  - Hva slags erfaring har familien med liknende sykdom? (hvem/hva/hvor/konsekvens)
  - Hva slags erfaring har familien med andre sykdommer?
  - Hva slags erfaringer har familien med tap?
  - Ovenstående som utgangspunkt for utvidet genogram
  - Andre belastninger i familien (spør evt om psykisk lidelse nå, evt avvent)
  - Hvem er familiens nærmeste?
  - Hva liker dere å drive med?

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### 5.3. Familiemøtet: familien og det syke barnet

psykosomatiske symptomer hos oss ofte langvarige/ kroniske:

- sykdommens mening; sykdom i et flergenerasjonsperspektiv
  - emosjonelle reaksjoner hos foreldre og søsken
- sosiale og praktiske konsekvenser for familien
- økonomiske og arbeidsmessige aspekter

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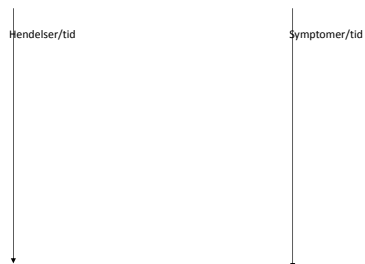
### 6. Utredning og terapeutisk prosess

- Foreldrearbeid kfr og suppler pkt 5 avhengig av hva foreldrene er åpne for;
  - Videre mhp stress/bio-psyko-sosial tenkning
  - Foreldreindividuelle samtaler bør tilbys
- Individuell utredning

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### Bruk av anamnesticke opplysninger inn i tidslinjer nyttig både m pas og foreldre

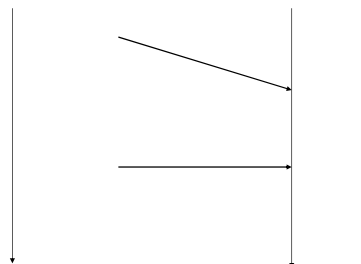
- Livshendelser
- Symptomutvikling



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### Bruk av anamnesticke opplysninger inn i tidslinjer nyttig både m pas og foreldre

- Livshendelser
- symptomutvikling



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## Individperspektivet: med barn/foreldre

- Mer symptomorientert kartlegning med foreldre
- Individuell barnepsyk. kartlegning av barnet
  - semistrukturert intervju
  - spørreskjema på dissosiative erfaringer
  - spesifikke metoder avhengig av indikasjon
- Spesialpedagogisk kartlegning oftest nødvendig

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## En bio-psyko-sosial sykdomsforståelse

predisponerende/ sårbarhet	utløsende	modererende/ opprettholdende
<b>tid</b>		
<b>biologisk</b> *for tidlig fødsel/ *tidligere sykdommer *tidl. utviklingsproblemer *funksjonshemninger *læreforutsetninger/lærevansker *temperament/rytmer	<b>biologisk</b> *skader *infeksjoner *pubertet/utvikling *a sykdom	<b>biologisk</b> *stress kan innvirke på kontroll med bevegelse og bevissthet *stress kan gjøre ns mer følsomt
<b>psykologisk</b> *læreforutsetninger/lærevansker *temperament *traumatiske erfaringer *vansker med tilpasning *store krav til seg selv *familieforventninger *familietilpasning	<b>psykologisk</b> *ulykker *tap *traumer inkl overgrep *signifikante hendelser	<b>psykologisk</b> *evne til læring og hukommelse er redusert *det kan være vanskelig å snakke om det som er stressende? Er det stress/belastning som fortsetter? *det er ingen som forstår
<b>sosialt</b> *læreforutsetninger/lærevansker *traumatiske erfaringer *vansker med tilpasning *store krav til seg selv *familieforventninger *familietilpasning	<b>sosialt</b> *familiebelastning *mobbing/erting *skoleskifte	<b>sosialt</b> *vedvarende familievansker *vanskelig å ta opp kontakten med venner etter langvarig sykdom, og hvordan forklare? *det er hardt å begynne på skolefagene når man har vært borte lenge *det er vanskelig å vite hva man har lyst til å drive med hvis man ikke orker så mye som før

## En bio-psyko-sosial sykdomsforståelse

predisponerende/ sårbarhet	utløsende	modererende/ opprettholdende
		<b>biologisk</b>
<u>Kroppen må gradvis trene seg til å leve vanlig igjen</u> <u>– relatere vanlig funksjon</u>		← *stress kan virke inn på kontroll med bevegelse og bevissthet ← *stress kan gjøre ns mer følsomt
		<b>psykologisk</b>
*Å frigjøre kapasitet krever stabilisering mhp stress		← *evne til læring og hukommelse er redusert
*Bearbeiding av traumatisk erfaring/livsbelastning krever også stabilisering		← *det kan være vanskelig å snakke om det som er stressende?
*Pågående livsbelastning må løses		← Er det stress/belastning som fortsetter?
*Hjelp til så formidle en forståelse		← *det er ingen som forstår
*viktig å arbeide parallellt med stabilisering		← *presset er for stort også i behandling
		<b>sosialt</b>
*familie støtte til å unngå stress/fokus		← *hvordan greier familien seg?
*hjelp til å formidle en forståelse		← *det er vanskelig å ta opp kontakten med venner etter langvarig sykdom, og hvordan forklare?
*gradvis opptrapping til skolefag skolen må få en forståelse		← *det er hardt å begynne på skolefagene når man har vært borte lenge
*støtte i gradvis aktivitetsøkning og opptrapping funksjon		← *det er vanskelig å vite hva man har lyst til å drive med hvis man ikke orker så mye som før

## Stress og forstyrrelser av sansning og bevegelse

Et forsøk på å forklare stress og konversjon i en ansvarsgruppe (gjennomgått med fam. først)

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## Hva er stress??

- Stress er alt som påvirker oss psykisk og fysisk slik at vi kommer ut av balanse
- Stress krever noe ekstra
- Stress er påkjenninger utover det vi greier å "rydde på plass" hver dag
- Eksempler
  - Å ha en virus-sykdom er stress
  - Å hele tiden føle at en må gjøre mer enn en greier
  - Traumatiske livshendelser er stress

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## Alle typer stress gir kroppslige utslag

1. Ex. eksamenstress og hjertebank og tørr munn...
2. Ex skolestress, vennestress, familiestress og hodepine, magesmerter
3. Ex stort akutt stress/traumer eller sammensatt kronisk og akutt stress og konversjons/dissosiasjonsproblemer

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## Hva er det som påvirker de kroppslige utslagene

1. **Mindre grad av akutt stress:** den energimobiliserende (sympatiske) del av det ikke viljestyrte nervesystemet aktiveres – alt settes inn på å mestre den akutte utfordringen (hjerterbank, tørr munn..)
2. **Mer kronisk stress** kan virke på både viljestyrt og ikke viljestyrt muskulatur og gi smerter (anspenhet, magen blir urolig... ”magefølelse”)

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## Hva er det som påvirker de kroppslige utslagene

3. **Kombinasjon alvorlig stress/traumer:** Stressfølsomme sentre i mellomhjernen som har med regulering av følelser og adferd å gjøre påvirker/blokkerer normal regulering av muskelbevegelser, sansning og hukommelse

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## gr.3 er dissociative lidelser inkludert konversjonslidelser

- Dette er forstyrrelser hvor en ikke lenger greier å samle eller integrere det en husker, sanseinntrykk, kontroll over kroppsbevegelser og følelser

eks ”lammet av skrekk”, ”ved siden av seg selv”.  
”ute av seg”

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## Hvordan kan vi behandle dette

- Det handler om å forstå kroppens språk
- For å forstå må vi tenke helhetlig
- Dette er ikke bevisste tankeprosesser men ligger på et nivå under de bevisste prosessene i hjernebarken

dvs at det nytter ikke å ”ta seg sammen”

Helene Gjone, Seksjon for Psykosomatikk og CL-barnepsykiatri, OUS Rikshospitalet

- En god referanse:  
Review. Psychogenic non-epileptic seizures: A model of their pathogenic mechanism.  
Gaston Baslet  
Seizure 20 (2011) 1–13

## 7. Videre behandling

Helene Gjone, Seksjon for Psykosomatikk og CL-barnepsykiatri, OUS Rikshospitalet

## Hvordan kan vi behandle dette

- Det handler om å forstå kroppens språk
  - For å  
TRYGGE; STABILISERE;  
SETTE ORD PÅ OG GI ROM FOR REAKSJONER PÅ DET VANSKELIGE
    - Stoppe traumatiske og/eller fastlåste situasjoner
    - Sette ord på det som ikke har vært uttalt
    - Lette stresset i hverdags situasjoner (skole, fritid, hjemme)
    - Legge til rette for gradvis rehabilitering (fysisk, skolemessig, sosialt) uten press
    - Tilrettelegge mhp funksjonsinnkrenkninger (sansemessig, bevegelsesmessig og når det gjelder forstyrrelser av konsentrasjon og hukommelse)
    - Bearbeide ledsagende psykiske plager
- Å bryte onde sirkler

Helene Gjone, Seksjon for Psykosomatikk og CL-barnepsykiatri, OUS Rikshospitalet

## Behandling - generelt 1

Utfordre ikke somatiske attribuering før familien er klare for det/ gjør det gradvis – vektlegg evt det som kommuniseres av belastning uten å vektlegge symptomer

### Kognitiv restrukturering sammen med pas/fam

- understrek helhetsforståelse
- drøft sammenheng belastning/symptom
- lær familien symptomforståelse; hva er normalt/ikke farlig
- unngå unødvendige us,

Helene Gjone, Seksjon for Psykosomatikk og CL-barnepsykiatri, OUS Rikshospitalet

## Behandling - generelt 2

### 2. Kontroll og mestring gjennom individual og familierterapi

- støtte til familiesamspill, defokuserer på smerter/ubehag/symptom
- familierterapi mhp til å forstå/uttrykke fam.og ind.belastning, løsning og reorientering
- individualterapi mhp symptomkontroll og andre løsningsstrategier med ulike terapeutiske tilnærminger avh. av problematikk; - safety first

Helene Gjone, Seksjon for Psykosomatikk og CL-barnepsykiatri, OUS Rikshospitalet

## Behandling-generelt 3:

3. Evt medikamentell behandling av komorbide psykiske vansker
4. skolemessig tilrettelegging
5. Fysioterapi?
6. Hva skal jeg si til de andre i klassen?

Helene Gjone, Seksjon for Psykosomatikk og CL-barnepsykiatri, OUS Rikshospitalet

## *Familien med det psykosomatisk syke barn vil ikke alltid ha psyk.behandling...*

- Obs motivasjon, erfaringer i fht BUP
- ta det somatiske symptom på alvor; kontrollert utredning
- foreldre som kompetente medspillere/samarbeid
- inntak/genogram
- individualvurdering av barnet
- kommunikasjon med somatiker avgjørende

Helene Gjone, Seksjon for Psykosomatikk og CL-barnepsykiatri, OUS Rikshospitalet

## Oppsummering: En bio-psyko-sosial sykdomsforståelse kan brukes som:

- Ramme for en bred utredning
- Sjekkliste for at vi har gjennomgått det vi har planlagt
- For å sortere våre komplekse saker
- Som grunnlag for å danne hypoteser
- For å etablere en forståelse sammen med pasient og familie og tverrfaglige samarbeidspartnere
- For å planlegge/gjennomføre behandlingstiltak

Helene Gjone, Seksjon for Psykosomatikk og CL-barnepsykiatri, OUS Rikshospitalet

- The practice of medicine is an art, not a trade; a calling, not a business; a calling in which your heart will be exercised equally with your head.

[Sir William Osler 1849-1919](#)

## Appendix 4: Brochure about PNES





Q

# Psykogene, ikke-epileptiske anfall (PNES)



norsk epilepsiforbund

## HAR DU SPØRSMÅL OM EPILEPSI?

### **RING EpiFon1: 22 00 88 00**

Mai: [epifon1@epilepsi.no](mailto:epifon1@epilepsi.no)

### **BETJENT**

Mandag og Tirsdag (1000-1400)

Torsdag (1700-2100)

Mange spørsmål dukker opp når man får epilepsi tett innpå livet. Kontakt EpiFon1 for å få noen å prate med. Her finner du treneide likemenn som selv har diagnosen eller er pårørende til noen med epilepsi.

Alle likemenn har tausehetsplikt.

### **FORFATTERE**

Hilde Nordahl Karterud, Avdeling for kompleks epilepsi, Klinikk for kirurgi og nevrofag, Oslo Universitetssykehus

ISBN 978-82-93215-10-3

### **REVIDERT**

Februar 2014



## **Psykogene, ikke-epileptiske anfall**

Psykogene, ikke - epileptiske anfall kan defineres som en type anfall som ligner på epileptiske anfall, men uten epileptisk aktivitet i hjernen. Psykogen betyr at fysiske eller psykiske plager har psykiske eller psykososiale årsaker. Psykogene, ikke - epileptiske anfall forkortes til PNES som står for Psychogenic, Non - Epileptic Seizures.

### **Hvor vanlig er PNES?**

Det er ingen pålitelig informasjon om forekomsten av PNES i befolkningen, men den ansås å være i størrelsesorden 2-33 pr.100 000. Ca. 20 % av pasienter som er innlagt ved epilepsisentra får "PNES - diagnosen", tilstaden er hyppigst blant unge kvinner. Det er ikke uvanlig at pasienter har en kombinasjon av både epileptiske og psykogene, ikke-epileptiske anfall.

### **Hva skyldes PNES?**

Det er mangelfull kunnskap om hvordan psykiske forhold kan føre til epilepsilignende anfall. Det man vet, er at det ikke er en enkel årsaksforklaring. Det er heller ikke alltid man finner frem til hvorfor en person har utviklet psykogene anfall. Samspill mellom ulike biologiske, psykologiske og sosiale faktorer vil være av betydning for om en person utvikler psykogene anfall.

Pasienter som får "PNES - diagnosen" er en svært heterogen gruppe. For enkelte kan anfallene være forårsaket av enkle følelsesmessige problemer som går over av seg selv, når problemet blir erkjent og akseptert. For andre kan anfallene være et symptom på en mer kompleks og sammensatt psykisk lidelse, som trenger langvarig behandling, ofte med bakgrunn i traumatiske opplevelse eller en kronisk vanskelig livssituasjon. Anfallene kan også være en reaksjon

på akutt stress hos personer som ikke har hatt psykiske problemer tidligere, eller det kan være en reaksjon på uheldige samspillsrelasjoner. Dette er bare noen få eksempler på forhold som kan bidra til en person utvikler psykogene, ikke - epileptiske anfall.

Det er også beskrevet økt forekomst av lærevansker og mindre hodeskader hos personer som har psykogene, ikke-epileptiske anfall. Man regner at ca  $\frac{3}{4}$  av dem som får "PNES - diagnosen" har kjente psykiske problemer, mest vanlig er angst og depresjon.

## **Behandling**

PNES skal behandles som en psykisk lidelse, vanligvis innen psykisk helsevern. Fordi årsakene til PNES er så mangfoldige, er det heller ikke en bestemt behandlingsform som er riktig. Dersom man vet hva anfallene skyldes, må behandlingen rettes mot årsaken (e). Dersom man ikke vet det, må terapeut og pasient sammen forsøke å finne ut om det er forhold i livet som kan forklare anfallene. For noen går anfallene over av seg selv uten at man finner frem til en bestemt årsak.

Fordi PNES som regel diagnostiseres i nevrologien og behandles i psykisk helsevern, kan det oppstå uenighet om diagnosen og pasientene kan bli kasterball mellom de ulike behandlingsmiljøene. Det er derfor en fordel om behandleren har erfaring med både epilepsi og PNES.

Siden det kan være alvorlige forhold som forårsaker anfallene, er det alltid viktig å søke profesjonell hjelp. Jo tidligere man kommer gang i med behandling, jo større er sjansen for å bli kvitt anfallene.

### **Kan man bli anfallsfri?**

Kunnskapen om langtidsprognosen hos pasienter med PNES er



foreløpig begrenset. Det man vet er at prognosen er avhengig av den (de) bakenforliggende årsaken (e). De fleste studier konkluderer med at vel en tredjedel blir anfallsfrie, mens to tredjedeler fortsatt har noe anfall 3- 4 år etter diagnosen er satt.

Det er flere forhold som kan påvirke forløpet. Prognosen er bedre dersom PNES blir oppdaget tidlig, slik at behandling kan iverksettes tidlig. Barn har også bedre prognose enn voksne. Når det gjelder anfallsutformingen og anfalletenes varighet, er det ikke enighet om hvorvidt dette har betydning for prognosen.

## Diagnostisering

Det kan være vanskelig å diagnostisere PNES. Psykogene, ikke – epileptiske anfall kan ligne på flere tilstander, for eksempel besvimelser, hjertesykdom, diabetes, nattskrekk, migrene, raserianfall, hyperventilasjonssyndrom eller tics. Mest vanlig er det at man tar feil mellom epilepsi og PNES. På tross av mye forskning på diagnostisering av PNES, finnes det i dag ikke diagnostikk som 100% sikkert kan skille epileptiske og psykogene, ikke - epileptiske anfall fra hverandre.

Når man skal stille ”PNES – diagnosen”, må man først utelukke mulige differensial diagnoser. Den sikreste måten å utelukke epilepsi på, er hvis pasienter får et typisk anfall under video-overvåket EEG-registrering (langtidsmonitorering), og man ikke ser epileptiske forstyrrelser på EEG under anfallet. I praksis kan dette være vanskelig, fordi pasientene ikke alltid får anfall under telemetri-registrering.

Klinisk kan PNES ligne på alle typer epileptiske anfall. Selv spesialister i epilepsi kan ta feil når de skal vurdere om et anfall er epileptisk eller et psykogent, ikke- epileptisk anfall, utlukkende ut fra observasjon av

anfallet. Observasjon av anfallsutforming er derfor kun et hjelpemiddel i den diagnostiske prosessen. Dette er et komplisert fagfelt. PNES-diagnosen stilles på bakgrunn av en totalvurdering - der anfallsutforming, anfallssetting, respons på antiepileptisk medisin, psykiatrisk intervju, EEG og bildediagnostikk av hjernen er viktige elementer.

Faren for feildiagnostisering mellom epilepsi og PNES er stor. Feildiagnostiseringen går begge veier, men mest vanlig er det at pasienter først blir feildiagnostisert med epilepsi, mens de egentlig har PNES. Ca 3/4 av pasienter med PNES har blitt behandlet for epilepsi først. Det hender også at noen får ”PNES – diagnosen”, når anfalletene egentlig er epileptiske. Konsekvensene ved feildiagnostisering kan være alvorlige, blant annet med bivirkninger av antiepileptika. Feildiagnostisering fører også til at de psykiske forholdene som ligger til grunn for PNES, forblir ubehandlet.

Fordi det kan være vanskelig å skille epilepsi og PNES fra hverandre, kan det ta lang tid før man finner frem til hva anfalletene skyldes. Dette innebærer at mange pasienter må leve med en usikker diagnose. En slik uavklart situasjon kan være vanskelig og frustrerende både for pasienten og legen.

PNES finnes ikke som egen diagnose i ICD-10 (International Classification of Disorders), eller i den amerikanske psykiatriske selskaps, DSM-IV. Tilstanden kan enten plasseres under F 44 Dissosiative lidelser eller under F 45 Somatiserings lidelser. Hvis man ikke vet hva anfalletene kommer av, kan man bruke R 56 Andre uspesifiserte kramper.

I Norge har man blant annet kalt denne type anfall for funksjonelle anfall. Internasjonale betegnelser som fremdeles er i bruk er for eksempel Pseudo-epileptic seizures, Pseudoepileptics, Non-epileptic

Seizures, Psychogenic seizures, Non-epileptic attack disorder (NEAD). Mest vanlig er det å kalle tilstanden for PNES etter den engelske betegnelsen "Psychogenic, Non-epileptic Seizures".

### ***Kan det være PNES når man slår seg under anfall?***

Det er ikke uvanlig at pasienter som har psykogene, ikke- epileptiske anfall skader seg under anfallene. En måte å forstå dette på, er at ulike psykologiske mekanismer kan påvirke "bevisstheden" slik at man mister kontrollen over kroppens reaksjonsmønstre.

### ***Kan man selv kontrollere anfallene når det er PNES?***

Mange er redd de skal bli tatt for å "spille" anfall når det blir kjent at anfallene ikke skyldes epilepsi. Det er viktig å være klar over at det her ikke er snakk om spill. Hvis man selv iscenesetter anfallene, kalles det simulering/anfall, og det er noe annet enn PNES. På samme måte som ved epileptiske anfall, kan noen pasienter med PNES finne frem til teknikker som reduserer eller stopper anfallene. For noen kan det for eksempel hjelpe med spesielle pusteteknikker.

### ***Skal psykogene, ikke- epileptiske anfall stoppes med anfallsdempende medisiner?***

Det skal ikke gis Vival/Stesolid eller andre anfallsdempende medisiner for å stoppe PNES. Disse medisinene stopper sjelden anfallene. Fordi Vival og Stesolid kan ha en beroligende effekt, kan det likevel hjelpe til å dempe anfallene i noen tilfeller. Mange som har vært feildiagnostisert med epilepsi har feilaktig fått mye Stesolid. Dette kan skape avhengighet og gi bivirkninger. Antiepileptika skal trappes ned av lege når det er slått fast at anfallene ikke er epileptiske.

### ***Kan man kjøre bil når man har PNES?***

Når det gjelder epilepsi må man ha vært anfallsfri i ett år før man kan

kjøre bil. Ved psykogene anfall er det ingen slike regler. Hvert enkelt tilfelle må vurderes av behandlende lege.

## **Å leve med PNES**

Noen synes det er en lettelse å få høre at anfallene er psykogene. Når det ikke er noe organisk som utøser anfallene, er det lettere å tenke seg at man selv kan påvirke sykdomsforløpet. Spesielt hvis man får hjelp hos psykolog eller andre innen psykisk helsevern. Man slipper angsten for leve med en vanskelig regulerbar epilepsi og man slipper bivirkninger av antiepileptika. Dessuten kan det få noen positive praktiske konsekvenser for førerkort og arbeid.

Andre opplever at PNES er en vanskelig diagnose å få, og ville heller foretrukket at anfallene var epileptiske. En grunn til dette er at "PNES – diagnosen" kan være en vanskelig diagnose å forstå. Mange bruker mye krefter på å lete etter en bestemt årsak, mens det ofte er flere forhold som spiller inn. Dersom man ikke vet hva anfallene kommer av, kan det også være vanskelig å forklare andre om diagnosen. Epilepsi er lettere å forklare, fordi det kan forklares ut fra epileptisk aktivitet i hjernen. Fordi det er mangel på kunnskap om PNES i helsevesenet, hender det at pasientene føler seg dårlig ivaretatt. Mange opplever stigmatisering både innad i helsevesenet og ellers i samfunnet. Psykiatriske diagnoser gir dessverre ofte en lavere status enn somatiske diagnoser.

Fordi mange har vært feildiagnostisert med epilepsi først, må de gjennom en prosess hvor de går fra neurologiske til psykologiske forklaringsmodeller på anfallene. Dette kan være en krevende prosess. Hvilken diagnose man identifiserer seg med, er av betydning for en persons identitet.

Samspillet mellom kropp og sjel er komplisert, og det er mye man i dag ikke har nok kunnskaper om. Den som har fått "PNES-diagnosen" befinner seg midt i dette kompliserte landskapet. Mange beskriver at de føler seg alene og uten støtte. Psykogene, ikke-epileptiske anfall kan også virke skremmende, fordi den kroppslige reaksjonen er så kraftig. Da kan det være en hjelp å forstå PNES som "kroppens måte å si ifra på". Kroppen uttrykker følelsemessige påkjenninger på forskjellige måter, noen får hodepine, andre får vondt i magen og noen utvikler psykogene anfall. Noen synes PNES er en dårlig betegnelse som gir liten mening, og velger derfor å kalle det "stress anfall", eller rett og sett bare "hendelser".

Norsk Epilepsiforbund er en interesseorganisasjon for mennesker med epilepsi, deres pårørende og andre interesserte.

I og med at svært mange mennesker med epilepsi i tillegg har psykogene anfall, eller at man ofte først diagnostiseres med epilepsi, er PNES-diagnosen viktig for NEF å ta tak i. De siste årene har NEF avholdt kurs for mennesker med PNES, samt kjørt kampanje for å øke kunnskapen om PNES innad i organisasjonen og utad.





## VÅRE BROSJYRER:

- A** Norsk Epilepsiforbund
- B** Kort om epilepsi
- C** Epileptiske anfall og epilepsisyndromer
- D** Epilepsi hos barnehage og skolebarn
- E** Epilepsi og utdanning
- F** Epilepsi og sosiale rettigheter
- G** Epilepsi og fysisk aktivitet
- H** Epilepsi og autisme
- I** Epilepsi og utviklingshemming
- J** Epilepsi og førerkort
- K** Epilepsi og alkohol
- L** Epilepsi og graviditet
- M** Plutselig uventet død ved epilepsi
- N** Epilepsi hos eldre
- O** Epilepsi hos menn
- P** Epilepsi hos kvinner
- Q** Psykogene, ikke-epileptiske anfall (PNES)
- R** Epilepsi og ansettelse
- S** Epilepsi og tannhelse
- T** For deg som er taxi- eller buss-sjåfør
- U** Epilepsi og samliv
- V** Epilepsi og svømming
- W** Barn med epilepsi
- W** Epilepsi og medisiner



## Psykogene, ikke-epileptiske anfall (PNES)



Avsender: Norsk Epilepsiforbund  
Karl Johans gate 7, 0154 Oslo  
22 47 66 00, [www.epilepsi.no](http://www.epilepsi.no)





Appendix 5:

PowerPoint presentation used by the staff in the 4-week inpatients stay,



# PNES

## *Psykogene non-epileptiske anfall*



# PNES

Anfall som likner på epileptiske anfall, men som oppstår gjennom psykogene mekanismer i hjernen og ikke epileptisk aktivitet



# PNES

Episoder med tap av kontroll som respons på stressende situasjoner, sensasjoner, følelser, konflikter eller minner, når alternative mestringsstrategier ikke er adekvate eller har blitt overveldet  
*(Reuber 2008)*



# Hvor vanlig er PNES?

- Forskning anslår at det er omtrent
  - 1500 personer i Norge som har diagnosen PNES
  - 150 person som får PNES-diagnosen hvert år
- 20% av alle pasienter som kommer til SSE har PNES



# Kjønn og alder

- PNES er noe vanligere hos kvinner enn menn:
  - Kvinner (75-80%)
  - Menn (20-25%)
- PNES kan komme i alle aldre, men de fleste er mellom 15 og 35 år gamle.



# PNES og epilepsi

- Det er ikke uvanlig å ha både PNES og epilepsi samtidig
- 10-30% av de som har PNES har også epilepsi
- Nesten alltid er det epilepsien som kommer først og PNES etterpå
- Hos de som har både PNES og epilepsi er det viktig å lære seg å skille mellom de ulike anfallstypene.



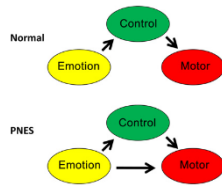
## Mekanismene bak PNES

- PNES oppstår gjennom ubevisste prosesser i hjernen

- Utviklingen av PNES skjer utenfor personens egen kontroll, slik at anfallet er ubevisste

- Belastningene på hjernen gjennom ubevisste prosesser blir for høye og hjernen reagerer med et anfall

- En beskyttelsesmekanisme for hjernen
- "Hjernen sier i fra"



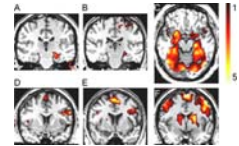
Fra: Functional connectivity of dissociation in patients with psychogenic non-epileptic seizures. Sylvie J M van der Kruis, et al. 2012



## Mekanismene bak PNES

- Nevrofysiologiske forandringer i hjernen

- Veier for informasjon og emosjon er annerledes ved PNES pasienter sammenlignet med kontroller



Fra: Functional connectivity of dissociation in patients with psychogenic non-epileptic seizures. Sylvie J M van der Kruis, et al. 2012



## Mekanismene bak PNES

- Ulike hypoteser om bakforliggende mekanismer:

- Konversjon av angst til kroppslige symptomer
- Dissosiativ reaksjon til overarousal eller traumatiske hendelser (dissosiasjon: oppløsning, spaltning, mangel på evnen til å integrere belastende erfaringer)
- Dysfunksjonell strategi for å takle stress
- En måte å kommunisere på



## Dissosiasjon

- Vi har ulike måter å takle stress eller skremmende situasjoner på
- Vi kan reagere med fysiske symptomer, som for eksempel hjertebank eller svetting
- Når vi opplever noe som er veldig belastende kan det bli overveldende. Da blir det vanskelig å takle følelsene på en bevist måte
- Noen ganger vil minnene av disse belastninger bli ubevisst "gjemt". Minnene kan være der, men man vil ikke huske det som har skjedd
- For noen kan minner av smertefulle hendelser plutselig dukke opp og trenge seg på i tankene og bevisstheten. Dette kan skje i stressfulle situasjoner eller når noe (ubevisst) trigger minnene
- Dissosiative anfall kan forstås som "cut-off mekanismer" for å unngå at vonde minner dukker opp. Personen spalter (eller dissosierer) følelsene fordi de kan være for vanskelige å takle.
- Anfallet er en ubevisst reaksjon og skjer utenom personens kontroll



## Mekanismene bak PNES

- Vi har ulike måter å takle stress på
- Hvis en person blir utsatt for veldig mye stress (for eksempel en person som jobber mye, bekymrer seg for ikke å strekke til, sover lite o.s.v.) kan kroppen reagere med:
  - Hodepine
  - Smerte i musklene i nakke og rygg
  - Magesmerter og kvalme
  - Utmattelse
  - Brystsmerter
  - Magesår
  - PNES

- Det varierer alt etter hva man er predisponert for å få.



## Hva er årsakene bak PNES?

- Som oftest er det ikke én årsak, men mange faktorer som bidrar til utvikling av PNES

- Disse faktorene kan være både
  - **Psykologiske** (f.eks. Tendens til å bekymre seg mye)
  - **Biologiske** (f.eks. Annen kroppslig sykdom)
  - **Sosiale** (f.eks. Høyt forventningspress)

- Faktorene kan deles inn i
  - **Predisponerende faktorer**
  - **Utløsende faktorer**
  - **Vedlikeholdende faktorer**



## Predisponerende faktorer for PNES

- Arvelige forhold
- Lærevansker
- Mindre hodetraumer
- Psykiske traumer
  - Vonde opplevelser som en ikke har fått bearbeidet
    - Mobbing
    - Overgrep
  - En del psykiske traumer kan være "blokkert" for hukommelsen
- Psykisk sykdom som angst, depresjon
- Annen kroppslig sykdom
- Kjennskap til epilepsi/anfall
- Uheldige strategier for å takle stress og konflikter
- Personlighet med stort forventningspress til seg selv
- Vanskelige familieforhold



## Utløsende faktorer

- Stor arbeidsbelastning på skole, jobb eller liknende
- Uløselige problemer
- Tap av en nær person
- Kirurgiske inngrep
  - Operasjoner
  - Tannlegebehandling
- Andre traumatiske opplevelser
- Andre former for økt stress



## Vedlikeholdende faktorer

- Manglende diagnose
- Opplevelse av manglende støtte
- Isolasjon
- Depresjon
- Vanskelig livssituasjon



## Årsaker til PNES

- De eksakte bakenforliggende årsakene til PNES vil altså være forskjellige fra person til person

### Eksempler:

- PNES kan være en reaksjon på akutt stress hos en person som ikke tidligere har hatt noen psykiske problemer
- PNES kan være en reaksjon på enkle følelsesmessige problemer, der anfallene forsvinner når problemet blir erkjent og akseptert
- PNES kan være et symptom på en mer kompleks og sammensatt psykisk lidelse



## Kroppslige mekanismer under PNES

- Ofte inneholder PNES et element av hyperventilasjon
- Når en hyperventilerer puster en ut ekstra mye CO<sub>2</sub>, slik at det blir mindre CO<sub>2</sub> i blodet. Dette gjør flere ting med kroppen:
  - Mengden fritt kalsium i blodet faller:
    - Du får prikkinger og nummenhet i huden spesielt i fingre og rundt munnen
    - Det kan oppstå sitringer i muskulatur og ristninger oppstår lettere
  - Blodårene i hjernen trekker seg litt sammen:
    - Du blir svimmel
    - Til slutt kan en besvime
  - Muskene i mellom ribbena jobber ekstra hardt
    - Du kjenner at det snører seg i brystet
    - Det virker tungt å puste
  - Disse følelsene kan være skremmende og bidra til å øke hyperventilasjonen.



## Kroppslige mekanismer under PNES

- Kroppen har instinktive reaksjonsmønstre som den iverksetter ved truende situasjoner
  - "Fright – Fight – Flight – respons"
  - Utslipp av adrenalin og noradrenalin
  - Effekter på kroppen:
    - Høyere hjertefrekvens → Hjertebank
    - Blodårene i muskulaturen utvides
      - Dersom en står stille blir mye av blodet i bena → Nærbesvimelse
    - Påvirkning av blodkarene i huden → Blekhet eller rødme
    - Pupillene utvider seg
    - Økt svette → Klamme hender
    - Økt spyttsekresjon
    - Fordøyelsen bremses → Kvalme, oppkast, manglende matlyst
    - Ristninger i muskulaturen



## Kroppslige mekanismer ved PNES

- PNES oppstår altså gjennom underbevisste og i utgangspunktet «normale» kroppslige reaksjoner
- Utformingen vil kunne være individuell ut fra hva en er predisponert for av symptomer.
  - Noen personer vil for eksempel uttrykk rykninger på én side mer enn den andre.
  - Noen vil ha mer uttalt brystsmerte og hjertebank
  - Noen vil ha mer uttalt kvalme og oppkast



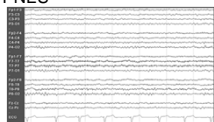
## Kroppslige mekanismer ved PNES

- **Bevissthetsreduksjon er veldig vanlig ved PNES**
  - Noen er helt borte og husker ingenting av det som skjer under store deler av anfall
  - Andre kjenner at de er våkne, men klarer ikke reagere, klarer ikke snakke eller bevege seg
- Dette skyldes at den normale funksjonen til hjernen (bevisstheten og evnen til å kontrollere kroppen) blir blokkert av de ubevisste prosessene i kroppen.
- Det er ingen skade på hjernen, men funksjonen er forstyrret.
  - Derfor kalles PNES også funksjonelle anfall
  - En sammenlikning:
    - Epilepsi er som en hardware-feil
    - PNES er en software-feil



## Hvordan stilles diagnosen?

- Mistanke om PNES ut ifra sykehistorien, komparentopplysninger
- Observasjon av anfall
- Registrering av anfall under video- og EEG-registrering (telemetri)
  - Ingen epileptisk aktivitet under anfall
  - Utformingen av anfallene passer med PNES
- Utelukke andre årsaker:
  - Besvimelser
  - Hjertesykdom
  - Diabetes
  - Migrene
  - Tics



## Hva skiller PNES-anfall fra epileptiske anfall?

- PNES likner på epileptiske anfall, og kan noen ganger være svært like som epileptiske anfall
  - Anfallsrelaterte skader, tungebitt og vannavgang forekommer også ved PNES
  - Mange får en usikker epilepsidiagnose og blir behandlet med antiepileptika, før en finner ut at det er PNES.
- Men som regel er det en del forskjeller i utformingen av anfall:
  - PNES kan vare mye lengre enn epileptiske anfall
  - Bevegelsene og ristingene ved PNES er ofte annerledes enn ved et epileptisk krampeanfall
  - Anfallene varierer oftere fra gang til gang, eller endrer seg over tid
  - Antiepileptika og anfallskuperende behandling hjelper som regel ikke ved PNES.



## Hva skiller PNES-anfall fra epileptiske anfall?

- Det er ikke farlig
- Du slipper antiepileptika (unntatt de ca 10-30% som har epilepsi i tillegg)
- Du kan lære å få kontroll over anfallene selv



## Behandling

- Ettersom årsakene bak PNES er så forskjellige, er det ikke én behandling for PNES.
- Behandlingen må skreddersys til hver enkelt
- Hos noen blir anfallene bedre/borte bare ved at de får diagnosen og forstår at det dreier seg om psykiske mekanismer.



## Behandling

- Kartlegge hvilke faktorer som kan ha bidratt til utviklingen av anfallene
- Deretter må en forsøke å bedre disse
  - Samtaler med psykolog, psykiater eller psykiatrisk sykepleier kan hjelpe
- Psykomotorisk fysioterapi er bra
  - Fordi en lærer å kjenne hvordan kroppen reagerer på stress og finne alternative måter å håndtere det på.



## Behandling

- Selv om prosessene i hjernen som gir opphav til PNES i utgangspunktet er helt ubevisste, kan en lære seg teknikker for å kontrollere anfallene
- Pusteteknikker kan være effektivt
- Når en forstår de prosessene som skjer i kroppen under anfall, kan en begynne å ta bevisst kontroll for å stoppe disse.



## Blir anfallene borte?

- Mange blir helt anfallsfrie
  - Med riktig behandling blir 60-70% av voksne anfallsfrie
  - Sjansen for å bli anfallsfri er enda bedre for barn og ungdom
- Sannsynligheten for å bli anfallsfri er høyere dersom diagnosen stilles tidlig, slik at behandling kan starte tidlig



## Hvordan forklare PNES til andre?

- Anfallene ser ofte dramatiske ut, og veldig mange som ikke kjenner til PNES og epilepsi godt, vil tro at PNES er epileptiske anfall
- Dessverre er kunnskapen om PNES i befolkningen liten (også beklageligvis blant mange leger)
  - Det kan gi opphav til misforståelser
- Noen ganger kan det være greit å bruke et annet navn:
  - Funksjonelle anfall
  - Epilepsi-liknende anfall
  - Stress-anfall
- En enkel forklaring er "en stressreaksjon fra hjernen".



## Hva med førerkort?

- Hvis man har plutselig, uventede bevissthetstap kan man ikke kjøre bil.
  - Det gjelder uansett om årsaken er epilepsi eller PNES



## Kjente personer med PNES



