

Living with a Contested Diagnosis Lyme Disease

An interview study on the experiences of living with persistent
uncharacteristic complaints

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Abstract

Objective The diagnosis Lyme disease is contested due to a dissonance (disagreement) between healthcare providers and persons with uncharacteristic complaints attributed to Lyme disease. The purpose of this study was to explore the value of the diagnosis Lyme disease from the perspectives of these persons. In order to reach this goal, the reasons they ascribe their complaints to Lyme disease as well as the consequences of the diagnosis Lyme disease on their lives and primary-care provider relationships were explored.

Methods A qualitative interview study was conducted with 11 adult participants with persistent uncharacteristic complaints who believe they have Lyme disease. A combination of phenomenological and grounded theory methodology was chosen.

Results The process leading to a self-diagnosis Lyme disease was initiated by a third party. Subsequently, many became guided by confirmation bias, seeking clarity and treatment via the healthcare community. However, a lack of consensus concerning the diagnostic procedures and treatment approaches for Lyme among healthcare providers enhanced diagnostic ambiguity and reinforced the participants' self-diagnosis Lyme disease. Subsequently, Lyme disease became a contested illness with many consequences. It had significant negative impacts on the participants' lives. Themes that emerged included facing uncertainty, not getting social validation and lacking recognition. It also initiated the adoption of new roles including distancing from conventional medicine and becoming their own healthcare provider. In addition, it led to a shift towards a strained and instrumental patient-primary-care provider relationship. Nonetheless, participants desired a tailored partnership. Despite the contested nature of the diagnosis Lyme, it was a source of value for the participants. It gave their complaints a name, which enabled them to have access to treatment and served as a justification for themselves and their surroundings.

Conclusion The participants aim to improve the quality of life. They remain dependent on their primary-care providers as they are the gatekeepers in legitimising their complaints, hence giving access to the sick role and subsequently to the various functions of a diagnosis in society. Therefore, it is critical to foster good patient-primary-care provider relationships which require improvements in communication between patients and primary-care providers, and between specialists and primary-care providers. Furthermore, to improve patient satisfaction in the healthcare community there is a need for simultaneous actions including the standardisation of tests and the adoption of a combined diagnostic approach and the evaluation of the benefits and limitations of a more flexible prescription of short-term antibiotics against Lyme.

Keywords: Lyme disease, persistent uncharacteristic complaints, diagnosis, contested, primary-care provider

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Introduction

Lyme disease or Lyme Borreliosis (LB) is an infectious disease caused by a bacterial infection transmitted to humans by a bite of the sheep tick (*Ixodes ricinus*) infected with *Borrelia burgdorferi sensu lato* (CBO, 2013). One out of five ticks is infected with the bacterium and can cause Lyme disease (RIVM, 2016a). It is considered the most common tick-borne disease in North America, Europe, including the Netherlands, and temperate zones in Asia (Steere, Coburn & Glickstein, 2004; Coumou et al., 2015; RIVM, 2016b). Every year over one million people in the Netherlands are bitten by a tick, yet the probability of acquiring Lyme disease after a tick bite is estimated at 2% (RIVM, 2016a). Recent data indicate that in 2014 there were approximately 23 500 people diagnosed with an Erythema Migrans (EM) which is a slight increase from 22 000 in 2009 in the Netherlands. The number of general practitioner (GP) or primary-care provider consultations for tick bites has decreased from 93 000 patients in 2009 to 82 000 patients in 2014 (Zomer et al., 2014). It is presumed that people nowadays remove the tick themselves more often without consulting a GP. Nevertheless, the number of LB patients remain high. A possible explanation for the increase in Lyme disease in the Netherlands is the growing number of infected ticks. This is caused by many factors, including a rise in the tick questing season due to increasing temperatures (ticks become active when the temperature rises above 7°C), more suitable habitats due to the expansion of forest areas and the increase in feeding and reproductive hosts (Sprong et al., 2012).

Generally, Lyme disease is a condition that has a defined group of symptoms with a clear biological cause. The disease has several stages associated with symptoms that are characteristic of the disease. In the first stage (early localised infection) which occurs within a few days or weeks, a person may recognise a circular skin lesion or a uniformly coloured spot around the place of a tick bite, an EM. However, if the infection remains unnoticed or untreated, Lyme disease may disseminate. The *Borrelia* bacterium enters the bloodstream and spreads to organs and tissues, leading to the second stage (early disseminated LB) causing symptoms such as nervous system infection (Lyme neuroborreliosis), cardiac complaints (Lyme carditis) and inflamed joints (Lyme arthritis). The final stage (late disseminated LB), which may take months or years to develop, include discoloration of the skin (acrodermatitis chronica atrophicans) in combination with the symptoms found in early disseminated LB (Health Council the Netherlands, 2013). It is estimated that apart from 23 500 people diagnosed with EM, 1 400 people were diagnosed with the (late) disseminated form (Zomer et al., 2014).

The primary goal of a Lyme disease diagnosis is to assign an evidence-based treatment approach to treat the underlying cause, removing the LB infection. The diagnosis and treatment trajectory of Lyme disease is described in a national guideline for healthcare providers ¹(CBO, 2013). In order to reach a diagnosis, it is

¹ In this thesis, a healthcare provider refers to a GP or specialist working in mainstream/conventional medicine or alternative/complementary medicine

recommended to adopt a combination of diagnostic approaches. Firstly, carry out a clinical examination, secondly assess a person's case history, and finally carry out laboratory testing (RIVM, 2016c). The diagnosis of an early localised infection is fairly straightforward by carrying out a clinical examination, the identification of an EM. According to the CBO guideline (2013), healthcare providers are advised to estimate the probability of Lyme disease prior to conducting a laboratory test. The results of the test should be interpreted on the basis of pre-test probability. In the Netherlands, an indirect method is mostly used, known as the serological assay procedure which detects antibodies produced by the body. If Lyme disease is diagnosed, the infection is treated with antibiotic therapy. (Health Council of the Netherlands, 2013)

Most people recover completely after being treated with antibiotics, yet between 1 000 and 2 500 are reported to have chronic uncharacteristic symptoms which are attributed to LB (Zomer et al., 2014). Various uncharacteristic complaints of Lyme disease may arise which possibly resemble other diseases. Symptoms include fever, muscular pain, painful joints, headaches, a stiff neck, lethargy, fatigue and concentration problems. If these complaints are attributed to Lyme disease and last up to six months or more, persons may be known as having Persisting Symptoms attributed to Lyme Borreliosis (PSLB) or are also referred to as post-Lyme disease syndrome or chronic Lyme disease². More specifically, it includes persons who have had early or late disseminated LB and experience persistent uncharacteristic symptoms after antibiotic treatment which may indicate residual damage or treatment failure.

There is also a group of people who report multiple unexplained complaints but do not have objective symptoms supporting the diagnosis Lyme disease. These persons may be self-diagnosed and/or infection is perceived by a healthcare provider as unlikely based on case history, clinical examination and serology, or are misdiagnosed. However, these persons strongly believe that Lyme disease is the cause of their persistent symptoms (Berende et al., 2016; Hassett, Radvanski, Buyske, Savage & Sigal, 2009; Health Council of the Netherlands, 2013). This will be the target group of this study and will be referred to as 'persons/people with persistent uncharacteristic complaints'.

There is a growing social concern around Lyme disease in the Netherlands based on the wide range of initiatives in the public, political, medical and scientific domain. Firstly, this is caused by the growing incidence of people with early Lyme disease and persons with persistent symptoms attributed to LB (Berende et al., 2016). Secondly, LB has a significant disease burden mainly in persons with long-term symptoms that are attributed to LB (9.09 out of 10.55 DALYs per 100 000 population; i.e. 86%) (Van den Wijngaard et al., 2015). Thirdly, there are multiple issues in the Dutch healthcare system regarding the diagnosis and treatment of persons with persistent symptoms attributed to LB.

More specifically, there are issues concerning the inability to properly diagnose Lyme disease, hence treating these persons. Firstly diagnosing Lyme disease is problematic as a person may not recall a tick bite due to the size of a tick (1-3 mm) or the disguised location of the tick on the body.

² Post-Lyme disease syndrome and chronic Lyme disease are not acknowledged in the medical profession in the Netherlands due to the large amount of debate surrounding the term (Health Council of the Netherlands, 2013).

Secondly, the uncharacteristic manifestations may resemble other diseases or there is no pathological cause to be found. For example, persons may be officially diagnosed as having Medically Unexplained Physical Symptoms (MUPS or MUS)³ or translated into (SOLK Somatisch Onvoldoende verklaarde Lichamelijke Klachten). In contrast to Lyme or other disease diagnoses, the diagnosis MUPS is a *per exclusionem* diagnosis; a diagnosis made by excluding other diseases resembling the complaints yet there is insufficient evidence to establish an identifiable cause. It is an official diagnostic label used in the medical profession with a written diagnostic and treatment protocol (NHG, 2016; Trimbos-Institute, 2010). It adopts a differentiated treatment approach, a biopsychosocial approach to address the different dimensions through physical and mental treatment methods. This may include psychotherapy (cognitive behavioural therapy), physiotherapy and psychiatric therapy or treatment. It also is a customised approach whereby both the patient and primary-care provider work together to determine the problem and treatment approach, as opposed to many somatic pathological diseases. (NHG, 2016).

Finally, the serological tests are inconclusive as they may generate both false-positive and false-negative outcomes resulting from the window period, serological scar and cross-reactivity and stealth mechanisms of *Borrelia* (Health Council of the Netherlands, 2013). This sometimes may lead to problems in patient-provider interaction such as self-diagnosis through unreliable and conflicting information acquired from the internet or other media and personal surroundings (Broerse & den Oudendammer, 2013; Vernooij & Kobus, 2015; Health Council of the Netherlands, 2013). Consequently, persons visit healthcare providers for a confirmation, making it difficult to carry out a proper diagnosis and giving consecutive advice for treatment. It has been reported that persons seek care abroad for diagnosis, as (other) laboratory tests are used which confirm their belief they have LB (Stichting de Ombudsman & NVLP, 2011; NOS, 2015; Health Council of the Netherlands, 2013). Individuals confront the healthcare provider with the results but the outcomes are not acknowledged in the Netherlands (Broerse & den Oudendammer, 2013). This also has consequences on the treatment approaches. Without a diagnosis Lyme disease, (long-term) antibiotic treatment is not recommended (CBO, 2013; Health Council of the Netherlands, 2013). A recent study among people with PSLB showed that long-term antibiotic treatment does not have additional benefits on the health-related quality of life compared to short-term treatment (Berende et al., 2016).⁴

Subsequently, persons with persistent uncharacteristic complaints live with a so-called contested diagnosis. This means there is a significant dissonance in beliefs regarding the diagnosis Lyme disease between healthcare providers and their patients. The persons with persistent uncharacteristic complaints

³ “Physical complaints that persist for more than a few weeks and for which, following proper medical examination, no medical condition can be found that can adequately account for the complaint in question” (Health Council the Netherlands, 2013)

⁴ The study included patients with a low RAND SF-36 score. It also tested two types of antibiotics over a course of 3 months

strongly believe that Lyme disease is the cause of their complaints yet this cannot be (uniformly) confirmed by healthcare providers.

Having a contested diagnosis has several possible consequences for persons with persistent uncharacteristic complaints. Firstly, many remain untreated, significantly impacting their quality of life. Secondly, it has an economic and health burden on society and the individuals themselves. Thirdly, it creates tension between people and the healthcare providers. On the one hand, people feel abandoned and not taken seriously by the medical community (Health Council of the Netherlands, 2013; Rappaard et al., 2006; Broerse & den Oudendammer, 2013; Vernooij & Kobus, 2015; Stichting de Ombudsman & NVLP, 2011). On the other hand, healthcare providers sense their patients have a lack of confidence in their expertise (Broerse & den Oudendammer, 2013). To date, no studies have been found analysing the consequences of a contested diagnosis Lyme disease on people's lives and healthcare provider relationships. Only a few qualitative studies have been conducted, focusing on the insights of people's experiences with Lyme disease (Rebman et al., 2015; Ali, Vitulano, Lee, Weiss & Colson, 2014; Drew & Hewitt, 2006). Therefore, this study will examine these consequences on the lives of people with persistent uncharacteristic complaints and on primary-care provider relationship. This is because the GP acts as a gatekeeper and provides first-line care for people who encounter medical complaints which may or may not be associated with Lyme disease.

It is presumed that many persons with persistent uncharacteristic complaints are resolute to the diagnosis Lyme disease. This is because according to grey literature studies, they are in favour of prolonged antibiotic therapy and object when the healthcare provider sends a referral to a psychologist or psychiatrist (Health Council the Netherlands, 2013; Nasleep van een tekenbeet: Ziekte van Lyme, 2012; Broerse & Oudendammer, 2013). These persons strongly believe and continue searching for the confirmation of the diagnosis Lyme disease. However, without proper diagnostic evidence, LB is not confirmed by the healthcare provider, hence antibiotic treatment is not prescribed. In this situation, healthcare providers encounter persons who are unreceptive to alternative treatment options yet do not understand this reluctance (Broerse & den Oudendammer, 2013). Consequently, it has been reported that persons go abroad for diagnosis and treatment (Health Council of the Netherlands, 2013; Stichting de Ombudsman & NVLP, 2011; NOS, 2015).

Currently, it is unclear what the reasons are for persons with persistent uncharacteristic complaints to initially ascribe and retain the diagnostic label Lyme disease. Despite a considerable amount of studies on the issues surrounding Lyme disease in the Netherlands, the value of the diagnosis Lyme disease among this group has not been specifically addressed. Only one fragment in an article as part of a monograph states that the label Lyme disease legitimises pain, suffering and disability, provides access to a community and externalises the problem by blaming the environment instead of oneself (Sigal & Hassett, 2002).

This study seeks to understand the value of the diagnosis Lyme disease from the perspective of people with persistent uncharacteristic complaints. This information may help clarify the reasons behind their resolution to the diagnosis Lyme disease. However, in order to reach this goal, it is important to understand the reasons they ascribe their complaints to Lyme disease as well as the consequences of the diagnosis Lyme

disease on their lives and primary-care provider relationships. As a result, this study attempts to contribute to the current literature by reporting on a qualitative study, answering the following research question:

What is the value of the diagnosis Lyme disease for persons with persistent uncharacteristic complaints attributed to Lyme disease?

The following sub-questions will be answered:

1. What are the reasons for persons with persistent uncharacteristic complaints to ascribe their complaints to Lyme disease?
2. What are the consequences of the diagnosis Lyme disease on the lives of persons with persistent uncharacteristic complaints?
3. What are the consequences of the diagnosis Lyme disease on the primary-care provider relationship of persons with persistent uncharacteristic complaints?

Theoretical Framework

The intent of this study is to examine the value of the diagnosis Lyme disease from the perspective of persons with persistent uncharacteristic complaints. Therefore, it requires a theoretical understanding of the concept diagnosis and its role in patient-provider communication. This is situated in the medical sociological literature on diagnosis, which will serve as a framework for this study.

A distinction can be made between diagnosis-as-a-category; a list of diseases and diagnosis-as-a-process, which is described as the act of reaching to a conclusion (Blaxter, 1978). Diagnosis is considered a central practice in medicine, bringing together and involving an interaction between a healthcare provider and a lay person. It is interpretive and organisational (Balint, 1964 as cited in Jutel, 2011). People seek an explanation when they self-interpret bodily irregularities from a medically authorised person. During the consultation with the healthcare provider, they share their perceptions, symptoms and experiences. The healthcare provider attempts to interpret this information and simultaneously carry out clinical and/or laboratory testing to interpret the condition. Diagnosis also organises. It validates and locates by “sorting out the real from the imagined, the valid from the feigned, the significant from the insignificant, the physical from the psychological” (Jutel, 2011). It distributes, by determining treatment options, prognosis and allocating resources. In other words, diagnosis plays a crucial role for both persons and healthcare providers.

Essentially, diagnosis is a classification tool that organises illness and disease (Jutel, 2009). These are two distinct terms often used interchangeably in the English language, and hence require clarification. Illness is subjective, a personal experience of what a specific individual perceives as an undesirable condition. Disease is objective, representing what professional medicine perceives as pathological. Therefore, disease is

diagnosed whereas illness is not. During the patient-provider encounter, the healthcare provider has the medical authority to transform an illness to a disease through diagnosing (Jutel, 2011).

Diagnosis has often been taken-for-granted (Madden & Sim, 2006) and mainly been viewed from a biomedical perspective. However, it may also be viewed from a sociological perspective. Usually, it has been embedded in the analysis of theories such as medicalisation, whereby human conditions become defined as medical conditions. Phil Brown was the 'founding father' of the term sociology of diagnosis, a discipline of study focusing on the diagnostic experience. He argued that "understanding diagnosis provides an important insight into how we understand disease, health and illness and the forces which shape our knowledge and practices" (Brown 1995 as cited in Jutel, 2009). Placing diagnosis central from a sociological perspective enables to understand topics such as illness experiences, health social movements and patient-provider interaction (Jutel, 2011).

Patient-provider interaction is considered a complex phenomenon, yet a requirement for optimal medical care. Research has shown there is a relationship between effective patient-provider communication and improved health outcomes (Stewart, 1995). However, the effectiveness of patient-provider communication is influenced by multiple variables including (i) background variables such as cultural variations, relationship preferences (i.e. paternalistic or egalitarian), personal characteristics and diseases; (ii) process variables such as communication behaviours (i.e. task-oriented or case-oriented) and; (iii) desired outcomes (Ong et al., 1995).

The healthcare provider is a gatekeeper for diagnosis, hence the subsequent actions, and interprets the information provided by their patient. This stresses the importance of the interaction and communication process between the patient and healthcare provider. The diagnostic moment, as Jutel (2011) calls it, is essential in the patient-provider relationship. It is a "site of contest and compromise because it is a relational process with different parties confronting illness with different explanations, understandings, values and beliefs" (Jutel, 2009). Therefore, this may create a strained relationship between patients and healthcare providers.

Tension may arise between patients and healthcare providers due to three reasons (Jutel, 2009). Firstly, there may be a thin line between illness and disease; symptoms are experienced by patients but there is an absence of sufficient clinical or medical signs to reach a diagnosis. Secondly, there is an unequal relationship in terms of knowledge and power between patients and healthcare providers as the latter holds the medical authority for a patient to give the sick role. This leads to the third reason. A diagnosis is needed as it gives an individual access to the medical system. Without a diagnosis, a patient may live in fear and uncertainty, may not receive social recognition in terms of support, have no/limited access to health services and feel stigmatised by having a psychological disorder that is perceived as in in the mind. These findings have been supported by empirical research. For example, patients with MUPS search for validation. Without a diagnosis, they acknowledge confusion and uncertainty about the future. Deprived of having the permission to be ill they lack access to services. Not having a diagnosis may make them question themselves that it is psychological. They also experienced anger by not being taken seriously, feeling marginalised by medicine.

Access to information and support groups was also difficult (Nettleton, Watt, Malley & Duffey, 2005; Nettleton, 2006; Kornelsen, Akins, Brownell & Wollard, 2016).

Another reason for possible tensions between the healthcare provider and their patient is due to differences in perspectives between the two groups. The medical viewpoint of the meaning of diagnosis focuses on clinical and laboratory evidence. In contrast, lay perspectives of illness is affected by issues of treatment and compensation, effects on professional and personal lives and the susceptibility and severity of their complaints (Zavestoski et al., 2004).

The sociology of diagnosis helps to understand the similar and different meanings diagnosis has for healthcare providers and patients. For both groups, a diagnosis has a medical role. It creates a clinical picture and determines the treatment options that will presumably benefit the patient. In addition, a diagnosis aids in prognosis, forecasting the future outcome (Brown, 1995; Jutel, 2009). However, diagnosis has various purposes in society and distinct psychological and sociological meanings for healthcare providers and patients.

In society, diagnosis gives access to research about the disease; studying causes, seeking treatment methods and exploring patterns (Jutel, 2011). However, it has other very important roles for society. It “represents the time and location where medical professionals and other parties determine the existence and legitimacy of a condition” (Brown, 1995). By providing legitimisation, diagnosis gives permission and socially validates a person’s claim to be ill as the complaints are officially labelled a disease by a healthcare provider. It gives the patient what Jutel (2009) states as “the allowance for, or tools to palliate and explains, what makes him or her different (deviant) from the norm”. It has a social/ cultural component as it distinguishes between normal and deviant behaviour, hence determining what conditions are entitled to be treated (Jutel, 2009). By certifying a medical complaint, diagnosis creates social order by opening doors in society: “once diagnosed, that bureaucratic and technically alienated disease-defined self now exists in bureaucratic space, a *simulacrum* thriving in a nurturing environment of aggregated data, software, bureaucratic procedures, and seemingly objective treatment plans” (Rosenburg, 2002). In short, diagnosis links an individual to the social system.

For healthcare providers, diagnosis is a key component in the practice of medicine, linking the healthcare provider and the patient. It enables healthcare providers to master the knowledge of the problem since they are confronted with an “unorganised illness”, consisting of a set of symptoms or complaints without an immediate clear origin. Their role is to apply their expertise and reach to an “organised illness” (Balint 1972 as cited in Zavestoski et al., 2004 and Brown, 1995). Constructing a diagnosis has a power and control role. It sets the medical professional apart from the lay person, confirming a healthcare providers’ expertise and status to allocate resources (Jutel, 2009). Moreover, within medicine, it also defines which healthcare providers are authorised and given responsibility for treating a particular condition (Jutel, 2009).

A diagnostic label has several consequences for a patient. Firstly, it enables them to be treated seriously and is given support by healthcare providers as well as their private and professional surroundings. Moreover, this label has a bureaucratic purpose as it is a gateway, providing legal access to privileges

including health services, insurance reimbursement, employment rights, etc. It also enables persons to seek networks of support by obtaining membership in support groups both physically and virtually through other communication channels, hence removing them from isolation (Jutel, 2009). To summarise, “the doctor certifies the medical nature of the complaint and ‘medical advice’ informs administrative and policy decisions” (Jutel, 2009). Secondly, diagnosis is an emotional ritual with psychological significance (Brown, 1995). It provides an explanation or “structured narrative” (Rosenburg, 2002), enabling people to make sense of their illness experience. Although it may not directly improve their condition, it may ease their fear of the unknown. At the same time, it may rearrange an individual’s identity whereby he/she may adopt different attitudes.

Various qualitative studies have been carried out studying the diagnostic experience among people with controversial diseases, showing that a disease-specific diagnostic label may be enabling. Apart from providing access to treatment, obtaining services and predicting prognosis (Sim & Madden, 2008; Daker-White, Sanders, Greenfield, Ealing & Payne, 2011), a diagnosis has many other meanings for a person. Firstly, diagnosis provides validation as complaints are confirmed by a healthcare provider, thus an individual achieves credibility by confirming that they were not imagining or faking their symptoms (Thompson, Isaac, Rowse, Tooth & Reuber, 2009; Sim & Madden, 2008; Daker-White et al., 2011). Secondly, diagnosis may offer reassurance that other serious conditions can be ruled out (Thompson et al., 2009; Sim & Madden, 2008; Undeland & Malterud, 2007). Thirdly, it may legitimise their symptoms as a disease (Undeland & Malterud, 2007; Thompson et al., 2009). Fourthly, diagnosis may give relief by removing uncertainty and by getting a name for their symptoms (Sim & Madden, 2008; Undeland & Malterud, 2007; Huibers & Wessely, 2006). Finally, diagnosis may remove individual blame and externalises the problem (Thompson et al., 2009).

However, a disease-specific diagnostic label may also be disabling. Firstly, it may create sadness and despair (Undeland & Malterud, 2007) due to the realisation of future uncertainty regarding upcoming progress (Sim & Madden, 2008), treatment options available, respect and understanding (Undeland & Malterud, 2007). Secondly, some people may reject the diagnosis as it may not reflect their understanding and meaning of the symptoms they experience (Sim & Madden, 2008). Thirdly, it may lead to victimisation and create an illness identity as well as exacerbate their symptoms leading to a self-fulfilling prophecy of non-recovery (Huibers & Wessely, 2006; Woodward, Broom & Legge 1995).

A study has also examined the diagnostic experience among people with MUPS. It shows that they sought a name for their symptoms. Particularly since patients experienced getting lost in the healthcare system, stigmatisation through psychologising the symptoms and being confronted with continuous certainty. However, for some, a diagnosis by exclusion was not considered helpful because it was still given with a sense of uncertainty. For others, any diagnosis was satisfying as it provided access to services or gave credibility. Nevertheless, some patients with longer unresolved symptoms began accepting MUPS by focusing on dealing with their symptoms to improve their quality of life and placing diagnosis at a lower priority level (Kornelsen et al., 2016).

In summary, diagnosis is a social process with various functions for people based on theory and qualitative studies. Using this theoretical background, this research will examine lay perspectives of the value of the diagnosis Lyme disease among persons with persistent uncharacteristic complaints.

Methods

A combination of phenomenological and grounded theory methodology was chosen (Creswell, 2013). In this study, the focus of inquiry was placed on describing the lived experiences of the participants and using this information to derive an explanatory framework concerning the value of the diagnosis Lyme disease grounded in the participants' lived experiences. A qualitative study was executed in which semi-structured interviews were conducted with persons with persistent uncharacteristic complaints.

Study Sample

The participants were recruited through purposive sampling from two Lyme organisations in the Netherlands; Stichting Tekenbeetziekten and Nederlandse Vereniging voor Lyme patiënten (NVLP).

The inclusion criteria were (a) Dutch-speaking adults, (b) who believe they have Lyme disease, (c) experiencing persisting long-term (> 6 months) uncharacteristic complaints (e.g. fever, muscular pain, painful joints, headaches, a stiff neck, lethargy, fatigue and concentration problems). Comorbidities may be included with similar clinical complaints and (d) the primary-care provider, the GP, in the Netherlands perceives active infection as unlikely. The sole exclusion criterion was participants with positive results following serological testing from the GP in the Netherlands. It is important to note that the use of antibiotics against Lyme distributed by a GP or specialist was not considered an exclusion or inclusion criterion.

The participants were asked by the Lyme organisations whether they were willing to participate in the study. Via email, the participants were informed about the study objectives and interview topics, asked permission for audio recording the interview and were informed that their anonymity would be guaranteed (Appendix I). Those who agreed to participate in the study were approached by telephone to make an appointment. Prior to the interview, a signed informed consent was obtained (Appendix II).

All the participants that were recruited by the Lyme organisations and were willing to take part in this study were interviewed. In total 11 people participated in this study; 7 participants were female and 4 participants were male. The age of the participants ranged between 21 and 76 years (mean: 50 years). The number of years living with complaints as reported by the participants ranged between 4 and 21 years (mean: 9 years).

Data Collection

The interviews took place at the participants' place of residence and lasted 45- 90 min. Semi-structured face-to-face interviews were conducted by means of an interview guide (Appendix III). The aim of

the interviews was to gain in-depth access to (i) the experiences and (ii) the meanings of the diagnosis Lyme disease. In order to break the ice, the participants were asked about their current health situation. Next, they were encouraged to recount their experience from the moment they were aware of a change in health status until their present health situation. Aspects that were important to explore included their past and current care trajectory including their complaints, Lyme tests, alternative diagnoses (including their perspective on MUPS) and treatments. Due to the scope of this study, the participants were asked to elaborate more on their experiences with their GP. The second half of the interview explored the consequences of (no) diagnosis Lyme on their lives. In order to allow the participants to relive their narratives, the interviews had an open character. This also enabled the researcher to ask probing questions, allowing further elaboration on the topic. Therefore, the topics were addressed in varying order.

Data Analysis

All interviews were audio recorded and transcribed verbatim whereupon the transcripts were anonymised. Next, data from all the transcripts were analysed by assigning codes using ATLAS.ti software. The interviews were coded using a mix of inductive (raised by the participants) and deductive approaches (based on the interview guide). During the process, each transcript was analysed repeatedly in order to develop new codes and uncover thematic aspects. The themes identified have been organised in Table 1 and are further explained in the results section.

Results

What is the value of the diagnosis Lyme disease for persons with persistent uncharacteristic complaints attributed to Lyme disease?

People with persistent uncharacteristic complaints live with a contested diagnosis Lyme disease. There appear discrepancies between the beliefs of the participants and their healthcare providers but also among healthcare providers. The search for a diagnosis starts with framing a self-diagnosis through lay knowledge. However, a lack of consensus from the healthcare community enhances diagnostic ambiguity and reinforces participants' self-diagnosis, namely that Lyme disease is the underlying aetiology of their complaints.

The contested diagnosis has many consequences on participants' lives as well as the relationship with their primary-care provider. The results show that besides the impacts, it initiates a new role. A change is observed in respondents' personal lives. They distance themselves from conventional medicine and compels those afflicted to seek care from alternative medicine. Furthermore, they become their own healthcare provider, adopt coping strategies and become open to a holistic treatment approach. Moreover, a new role is also found concerning the relationship with their primary-care provider. There is a shifting balance of power

in the doctor-patient relationship. The participants no longer blindly trust the expertise of their GP but expect and desire a more mutual participation. The results have been summarised in Table 1.

The diagnosis Lyme disease can be summarised as being a ‘curse and a blessing’ for people with persistent uncharacteristic complaints. It is a curse because the diagnosis is contested causing participants to enter a no man’s land, experiencing daily limitations, lacking access to treatment and recognition. Consequently, it forces them to take action by becoming their own healthcare provider; they self-diagnose and search for treatment themselves for Lyme disease. Despite the adverse experiences, they continue ascribing their complaints to Lyme disease and are members of a Lyme organisation, hence it is also a blessing, a source of value for the participants. Even if the diagnosis is contested, it has, at times, enabled the participants to gain access to (short-term) treatments, which have partially been effective in tackling their complaints. In addition, there is possibly the emotional value of the diagnosis Lyme disease. Having a label, whether it is contested or not, validates the individuals’ claim that an illness exists in their body and may give some clarity to themselves and their surroundings, a possible justification of the complaints they have been experiencing. Currently, there is no plausible alternative diagnosis. Furthermore, perhaps it is also the ability to show themselves and possibly their surroundings, their unconditional effort, to reach their goal of controlling their complaints to improve their quality of life.

Table 1. Summary of the themes based on the interviews (n = 11)

	Main themes	Sub-themes
<p><i>Sub-Question 1:</i> What are the reasons for persons with persistent uncharacteristic complaints to ascribe their complaints to Lyme disease?</p>	<ol style="list-style-type: none"> 1. Suggestion by acquaintances 2. Guided by confirmation bias 3. Healthcare community reinforces unclarity 4. Access to healthcare: diagnosis a gate to treatment 	<ol style="list-style-type: none"> 2.1 Identifying complaints from online symptom lists 2.2 Participants' knowledge of unreliable Lyme tests 2.3 Searching for a positive Lyme test result 2.4 Believing Lyme disease causes other infections 3.1 Interphysician diagnostic discrepancies 3.2 (Partial) symptomatic relief from antibiotic treatment 3.3 Diagnostic tests leading physician decision-making 3.4 Misdiagnosis
<p><i>Sub-Question 2</i> What are the consequences of the diagnosis Lyme disease on the lives of persons with persistent uncharacteristic complaints?</p>	<ol style="list-style-type: none"> 1. Impacts 2. New Roles 	<ol style="list-style-type: none"> 1.1 Experiencing persistent untreated complaints 1.2 Enduring negative physical and social impacts of complaints 1.3 Facing daily and future uncertainty 1.4 Not getting social validation from surroundings 1.5 Lacking recognition from conventional medical community 2.1 Distancing from conventional medicine 2.2 Becoming their own healthcare provider 2.3 Learning to cope 2.4 Appearing receptive to a holistic treatment approach
<p><i>Sub-Question 3</i> What are the consequences of a contested Lyme disease on the primary-care provider relationship?</p>	<ol style="list-style-type: none"> 1. Impacts 2. New Roles 	<ol style="list-style-type: none"> 1.1 Ambivalence towards primary-care provider 2.1 Altering relationship 2.2 Desiring a partnership

What are the reasons for persons with persistent uncharacteristic complaints to ascribe their complaints to Lyme disease?

There are many reasons for persons with persistent uncharacteristic complaints to ascribe them to Lyme disease originally triggered by lay knowledge. In the beginning, there was an initial unawareness of a tick bite and the first complaints were not assigned to Lyme disease. In most cases, it was a third party that initiated the participants to conceptualise their complaints to Lyme disease. Subsequently, many became unconsciously guided by confirmation bias. To begin with through checking and identifying with the complaints found on online symptom lists for Lyme disease. This led to participants carrying out Lyme tests, yet became aware of the unreliability of the tests. They held on to the positive results rather than negative results. Over time, they developed additional infections related to a low immune system which they ascribed to Lyme disease.

The participants sought clarity and treatment via the healthcare community. However, the healthcare community created further unclarity and thus possibly reinforced the self-diagnosis Lyme disease. Firstly, due to discrepancies between healthcare providers regarding the diagnosis and treatment of the complaints. The belief that Lyme was a cause of the complaints was reinforced when participants noticed (partial) health improvements after antibiotic therapy. Secondly, it appeared that Lyme tests were predominantly used as definitive proof in confirming (no) diagnosis, whereby the clinical picture and case history was not taken into consideration. Thirdly, treatments were prescribed for alternative diagnoses, yet these were ineffective and so complaints persisted, suggesting a misdiagnosis.

In addition, participants possibly had a desire to ascribe their complaints to Lyme disease to gain access to healthcare; the diagnosis Lyme disease is a gate to treatment.

1. Suggestion by acquaintances

The diagnostic process started with all participants reporting an unexpected change in health status triggered by becoming aware of 'strange' complaints of an unknown origin, mainly fatigue without having noticed a tick bite. Three participants also reported a particular incidence such as a fall or injury that triggered their awareness. Most participants never instantly assigned their complaints to Lyme disease, even though some recall having a tick bite a few months before their complaints or many years ago (5/11). The majority, (6/11) did not recall having an EM. Some respondents did not visit a GP (4/11) whereas others visited a GP but did not receive antibiotics (3/11). Others could not recall ever being bitten by a tick throughout their life (2/11). However, as time passed and the variety and severity of the complaints intensified, the respondents gradually became to believe that Lyme was the cause of their complaints. Interestingly, in almost all cases it was suggested by a third party (8/11). This included an acquaintance such as a neighbour, a friend or family member (5/11) or a healthcare provider such as a neurologist or a dietician (3/11) but not a GP.

'At one point a friend said: you were a bitten by a tick, right? Yes, that's right. Then I think you have Lyme disease.' (Female, age 47)

2. Guided by confirmation bias

Participants may have ascribed their complaints to Lyme disease because they were, unconsciously, guided by confirmation bias. They seemed to have the tendency to identify with the complaints from online symptom lists for Lyme disease, show distrust towards Lyme tests, yet nevertheless, search for positive Lyme test results and assign Lyme disease as the cause of other infections. All which confirm their preconceptions that Lyme disease is the aetiology of their complaints.

2.1 Identifying complaints from online symptom lists

Many were not instantly convinced that Lyme disease could be the cause of their complaints and so sought information through the internet where they discovered lists of possible symptoms attributed to Lyme disease (5/11). The respondents identified themselves with the majority of the complaints listed which led to the self-diagnosis Lyme disease.

'Then you hear via acquaintances that they know someone with similar complaints and has Lyme. You don't immediately take it seriously. However, if you find a list of symptoms via Google of which 37/40 complaints apply to you, then you start believing that Lyme is a possible cause.' (Female, age 21)

'I found various checklists on the internet and I think out of the 40 complaints I had 25-30. I thought well, yes I have that, that I have for sure. Hey, that is weird that is a strange complaint I also have. After being bitten 80 times and the high scores on the list, it is almost inevitable that I have Lyme disease.' (Female, age 51)

2.2 Participants' knowledge of unreliable Lyme tests

Subsequently, once a plausible label for their illness experience was found, the process of seeking an 'official' diagnosis commenced. Participants conducted a Lyme test, first via a healthcare provider in conventional medicine (8/11), mainly their GP (6/11). In all these cases, the first test was negative. Three participants had a first positive test result. These tests were not conducted by a GP but by a naturopathic doctor, neurologist or at a commercial laboratory. Although the majority had a negative test result, the participants continued to believe that Lyme was the cause. This is because the respondents had immersed themselves in online information about Lyme disease and were aware of the shortcomings of the diagnostic tests (7/11), namely the perceived lack of reliability of the serological tests carried out in conventional medicine.

2.3 Searching for a positive Lyme test result

All participants had gone through multiple different Lyme tests by different specialists throughout the years in the Netherlands (and some abroad), yet these presented controversial results; some were negative and some were positive. Many tests performed by conventional medicine (GP and specialist) were serological tests (ELISA and Western Blot). These were almost in all cases negative. Some participants also did various other tests with the majority at a commercial laboratory in the Netherlands carrying out German tests (LTT) and American tests (PCR) (5/11). These outcomes were positive in all except one case. A few participants also did an LTT test (2/11) and NK56 (2/11), which were all positive. However, it was not clear where these tests were carried out. Three participants also carried out tests via alternative medicine (3/11). These were in all cases also positive.

As a result, it was possible that the participants discarded the first negative test and continued performing different diagnostic tests until a positive result was obtained. This result confirmed their belief that Lyme is the cause of their complaints which led to the participants discarding the possibility of a false-positive outcome.

2.4 Believing Lyme disease causes other infections

A few participants reported additional infections such as Pfeiffer and Bartonella (3/11), which they reported as co-infections. They consider it as a consequence of having Lyme disease. Furthermore, many stated being frequently susceptible to other, although not specified, common infections related to a poor immune system. The majority of the respondents stated that Lyme is a multisystem disease leading to reduced immune functioning hence exposed them to various infections (8/11). It appears that participants have the tendency to ascribe the majority of other infections to Lyme disease.

3. Healthcare community reinforces unclarity

The healthcare community also plays a role in reinforcing unclarity and so may sustain participants' existing beliefs that Lyme disease is the cause of their complaints. This is due to interphysician diagnostic discrepancies, (partial) symptomatic relief from antibiotic treatment, Lyme tests acting as conclusive evidence for Lyme disease and misdiagnosis due to ineffective treatments corresponding to alternative diagnoses.

3.1 Interphysician diagnostic discrepancies

Healthcare providers' discrepancies create unclarity and so may reinforce participants' existing beliefs that Lyme disease is the cause of their complaints. This is because the majority of the participants have had at least one positive test for Lyme and/or found a specialist in complementary and/or conventional medicine who was willing to treat them for Lyme. However, this was individually determined and often not acknowledged by other specialists. Therefore, it is uncertain whether these criteria resulted in clinicians diagnosing the participants with Lyme disease.

Participants have been through multiple diagnostic tests and treatments, offered by different healthcare providers during their illness trajectory. During the interviews, more than 20 different specialists were mentioned, for instance, a neurologist, internist, orthomolecular doctor and naturopathic doctor. These were also consulted in different health institutions such as hospitals, Lyme clinics, private clinics, etc. As previously mentioned, all participants have done multiple different Lyme tests among the different specialists and health institutions. Guided by confirmation bias, a positive Lyme test result may give the participants the potentially false impression they were diagnosed with Lyme disease.

Regarding treatment, throughout the years of living with the complaints, most participants reported receiving antibiotic regimens from a healthcare provider who was willing to prescribe antibiotic treatment for a period of time (9/11). Most received antibiotics through a specialist (8/11) and only three respondents received antibiotics from their GP. Two participants reported receiving treatment via a private clinic in the Netherlands. Two participants received treatment from a specialised Lyme clinic in the Netherlands. However, receiving antibiotic therapy was often for a short period of time either because the particular type of antibiotic treatment was not effective in reducing the complaints, only temporarily reduced complaints or due to other factors (i.e. doctor's pension, healthcare inspection, negative health effects of treatment, etc.) Therefore, receiving antibiotic treatment from one specialist did not guarantee that another specialist would also prescribe antibiotics. As a result, receiving antibiotic treatment against Lyme disease may have given participants the potentially false impression they were diagnosed with Lyme disease.

As a result, in this study, it is unclear whether the participants have been diagnosed with Lyme disease.

3.2 (Partial) symptomatic relief from antibiotic treatment

Another reason why the participants possibly ascribed their complaints to Lyme disease is because particular antibiotic treatments were (partially) effective in reducing or removing complaints as reported by the following participants.

'She prescribed doxycycline for 1 week, 100 milligrams per day. So I took it and I felt better again. I thought, damn, so it is Lyme.' (Male, age 76)

'During the 8 or 9 months, I used antibiotics the following disappeared: sleeping badly and frequent urination. Those were one of the first complaints that disappeared which had a significant impact on my energy level. Then joint pain decreased significantly and nerve pain.' (Male, age 42)

3.3 Diagnostic tests leading physician decision-making

Healthcare providers may create unclarity among participants as the (no) diagnosis Lyme disease appears predominantly based on the Lyme tests, not taking the clinical presentation and case history into account. This was also literally stated by one participant:

'[Healthcare providers] are too one-sided. They need proof from laboratory tests otherwise, they cannot make a diagnosis. The examination is very difficult. In the past, they did an abdominal tap. Now they do nothing.'
(Male, age 76)

According to the participant narratives, healthcare providers principally grounded their decision to (not) prescribe antibiotics based on the diagnostic tests carried out by a particular GP or specialist. If the test was positive, antibiotics were prescribed. Therefore, it appears that the diagnostic tests are conclusive, hence determine whether the participant had access to antibiotic therapy or not. It seems that the clinical aspects or persons' narratives are disregarded, yet are for the participant's clear proof for ascribing their complaints to Lyme disease. In only two cases antibiotics were prescribed purely on the clinical symptoms. Another reason for prescribing antibiotics was due to participant perseverance. Two respondents never received antibiotic treatment.

3.4 Misdiagnosis

During the search of a medical condition that adequately explained the complaints, various alternative diagnoses or possible presumptions were made by a healthcare provider. Examples included a burnout, being overstrained, menopause transition, syphilis, thyroid disease, fibromyalgia and personality disorder. In almost all cases, the participants initially relied on the expertise of their healthcare provider and followed the treatment based on the diagnosis. However, the participants noticed that the complaints persisted after the treatments. Therefore, this possibly indicates a misdiagnosis as the treatment of the alternative diagnoses was ineffective. Hence, the participants continue to ascribe their complaints to Lyme disease.

4. Access to healthcare: diagnosis as the gate to treatment

The final possible more implicit reason or, perhaps, better said, a desire, for participants to assign their complaints to Lyme disease is due to the general function of a diagnosis in the medical community. The participants were not explicitly asked what their current goal is, however, it seems that the majority are searching for a treatment. More specifically, searching for methods to control their complaints rather than seeking a cure for Lyme disease. This is because during the interviews the participants described their complaints in utmost precision and explained how the complaints had a significant negative impact on their lives, physically and socially. Another reason is because many participants stated they do not necessarily desire antibiotic treatment. They are receptive to other treatment approaches. Although the majority reported they are in a healthier state than before, they continue seeking methods to control their complaints. As many stated, their main aim is to *'to improve the quality of life'* (6/11).

'The quality of life has improved significantly. But I am not there yet. I am still searching for a further improved quality of life.' (Male, 42)

'It appears you cannot treat it (Lyme) to ensure it is gone but under control. You need to ensure that your immune system improves so that your body can control it'. (Female, age 47)

'My goal is to maintain what I have. Maintaining this level and accept that I am not always healthy. That's fine. Of course, I do not want to fall back and my goal now is to wait for the science.' (Female, age 51)

However, it appears that this goal can only be achieved if the participants obtain a diagnosis; a label for their complaints. Without a diagnosis, the label Lyme, the participants have no access to the treatment of their complaints. Based on various participant narratives, if no abnormalities were found in the tests by the healthcare provider, the participant was left unaided even though he/she had persisting (severe), possibly visible, complaints.

Lyme disease seemed to be the best match, even if (some) diagnostic tests were negative. This is because either other diagnoses were made but the treatment was ineffective or the healthcare provider did not provide an alternative diagnosis as no abnormal pathologies were found even though the participants had various persisting complaints. Therefore, the participants continued to ascribe their complaints to Lyme disease.

To conclude, persons with persistent uncharacteristic complaints ascribe them to Lyme disease for many reasons. As a result, they are left in limbo land, living with a contested diagnosis. The participants strongly believe that Lyme disease is the cause of their complaints yet this cannot be (uniformly) confirmed by healthcare providers. This has had several consequences on the participants' lives and the relationship with their primary-care provider.

What are the consequences of the diagnosis Lyme disease on the lives of persons with persistent uncharacteristic complaints?

A contested diagnosis Lyme disease had multiple consequences on the participants' lives. They suffered from various (severe) persisting complaints, which had a large physical and social impact on their lives. They faced daily and future uncertainty concerning their complaints, treatment availability and prognosis. Furthermore, participants sensed a lack of understanding of their surroundings and a lack of recognition from the medical community.

However, a contested diagnosis also led to the participants' adopting a new role. Unsatisfied with conventional medicine, participants sought care from complementary medicine. Many also became their own healthcare provider through self-treatment and learned to cope with their restrictions. Also, it appeared they were drawn to a more holistic treatment approach.

1. Impacts

1.1 Experiencing persistent untreated complaints

The participants reported that over time their complaints manifested into a wide variety of medical problems (46 different complaints were reported - Appendix IV). The main ones included fatigue, headaches, concentration loss, neuralgia, painful joints and forgetfulness. Some participants (4/11) also had more severe complaints with loss or immobility of arms and legs.

'I was more or less confined to my bed or bedridden. I moved around with a walker which my mother at one point bought.' (Male, age 46)

Throughout the many years of living with the complaints, on average 9 years, these fluctuated; they come and go. After various treatments, many complaints have disappeared. The majority reported being in a better health state, yet many appear to continue searching for a treatment as some complaints persist, predominantly fatigue and to a lesser extent headaches, dizziness and muscle ache.

'Currently it is limited to many headaches, fatigue, painful joints, hair loss, weak intestines and poor resistance.' (Female, age 51)

1.2 Enduring negative physical and social impacts of complaints

All participants described their complaints as having a substantial adverse effect on their lives, physically and socially that determined their lives to a great extent. This has to do with the nature and number of the complaints, as well as with the unpredictability of experiencing those complaints.

During their all-time low periods when their complaints were most severe, all participants reported limited physical functioning. This was mainly the loss or immobility of arms and legs and constant fatigue. However, it was mainly the various social consequences which were considered constraining. These included interfering their occupational and interpersonal lives such as the inability to work and the loss of jobs, being forced to live with their parents and unable to carry out activities with family and friends.

'While everything is gone. My job is gone, my house was gone, I lived in small rooms, of course, I lost my friends, my social contacts, my mobility' (Female, age 37)

'Being ill is not only about being tired or in pain but also not going to work, doing fun things with your partner and children. "Shall we go to an amusement park [name]?" No, it takes me 3 days to reload so I can only go for 2 hours. I am constantly busy [with the complaints]. When I have this appointment, I need to rest afterwards. I cannot cook or help my partner.' (Female, age 43)

'There were periods where I could not drive a car or go to the cinema since the sound was too loud which literally hurt my nerves.' (Female, age 47)

Although the majority currently consider their health status as *'reasonable'* (8/11), most respondents emphasised various remaining social and physical restrictions. The main restriction was fatigue as respondents need to take regular periods of rest after each activity. Some reported their functioning capacity ranged between 1-3 hours per day (3/11).

'I must do everything with breaks. My max is 2 hours. Being busy or awake. Then I need to rest again. One hour or half an hour in the form of sleeping or lying upstairs in bed. I have to recharge after each activity. This can be vacuuming, doing an interview with you, grocery shopping or cooking.' (Female, age 43)

In addition, throughout the years many faced increased out-of-pocket medical expenses due to multiple diagnostic tests and treatments, often not covered by their insurance. One participant decided to go to trial against the insurance company in order to reclaim the treatment costs she made.

'Then you need to go to a laboratory (name) which costs 250 euros. It costs you money and you have to figure out everything yourself while you are ill.' (Female, age 43)

1.3 Facing daily and future uncertainty

Another limiting factor was the continuous uncertainty the participants faced on a daily basis. This concerned the fluctuating nature of the complaints showing no clear pattern. These complaints were experienced as unpredictable and unstable, making it difficult to create structure in the daily lives and plan activities. As one respondent stated:

'That is the most disturbing of this disease; that you cannot plan. I cannot arrange an appointment now before dinner because I do not know how I am then.' (Female, age 43)

At first, two participants initially thought the diagnosis Lyme provided relief until they realised no treatment was available. Therefore, participants were also confronted with future treatment and prognostic uncertainty due to the absence of treatment and disease prognosis from the medical system.

'And when I had the diagnosis [Lyme], I was relieved and I thought now I could be treated but two years later it appears I cannot be treated and that is terrible.' (Female, age 62)

Based on the narratives, (antibiotic) treatment could not be guaranteed. Furthermore, the majority of the participants (6/11) stated that the diagnosis Lyme had no effect on treatment opportunities as there is

currently, in the Netherlands, no long-term treatment available against Lyme if the two-week antibiotic treatment was ineffective.

'Even if I am diagnosed with Lyme, I still don't have a treatment'. (Male, age 46)

'[The diagnosis Lyme] means nothing. Someone who is diagnosed with Lyme also only receives a two-week antibiotic treatment and that is not sufficient, only if you had a tick bite and then receive a two-week antibiotic treatment. However, there are also people who are diagnosed with Lyme and receive a two-week antibiotic treatment yet become ill years later. Then they [the medical community] say that you have received antibiotics. It is the protocol so you have something else. In any case, you are left unaided. So it actually doesn't matter.' (Female, age 47)

However, contradictorily, participants seemed to continuously seek care from different specialists who, first, confirmed they have Lyme, and second, were willing to provide long-term (antibiotic) treatment.

Participants also faced future uncertainty regarding their health prognosis. With or without the diagnosis Lyme, many (5/11) believed their health state is chronic and so is incurable.

1.4 Not getting social validation from surroundings

Most participants spoke of unconditional support from their surroundings (family, friends and/or colleagues) (8/11). For some, the diagnosis Lyme provided clarity for their surroundings, an explanation of the complaints they have been experiencing.

'When I got the [Lyme] label, even though it was not from the GP or hospital, it was accepted. If you have the [Lyme] label, it becomes clear for them [the surroundings].' (Male, age 42)

'I looked into chronic Lyme and it cannot be healed. It is what it is. It has its advantages as your surroundings like to know it is an animal. It has really bitten you. It is a real disease which can be proven.' (Male, age 59)

However, more than half also mentioned situations where they sensed a lack of understanding or empathy (8/11). Some participants explained they got the impression they needed to constantly justify themselves due to symptom invisibility – i.e. the lack of outward signs of illness. This can be illustrated through the following excerpts:

'The family was supportive but there were strange and irritating questions proving that they do not understand. [For example]: I am very ill and they say, with enthusiasm, that in 6 months we are going on vacation to South Africa with the entire family. I had to explode. I don't want to go all the way to South Africa. I cannot go to South Africa, it is a punishment for me. I do not feel well enough to go abroad. I would

feel worse. I would be away from my familiar environment, I have to travel, my bed might not be good, I have difficulty sleeping, etc. That empathy is missing. You need to be very angry in order to wake them up. If you're angry, you get emotional and then it all of a sudden becomes clear to them. People think there is nothing wrong with you. They don't see anything. That's what makes it hard.' (Male, age 42)

'They listen but you recognise that they don't get it, they don't understand. For example, I can tell my friend something and the next moment she says: shall we go away for the weekend and stay in a house somewhere (name place)? Or would you like to come for a day? And then you know they do not quite understand your situation.' (Female, age 37)

Four participants also mentioned that their surroundings attempt to cooperate by providing suggestions of possible diagnoses and/or treatment options. Three participants found this irritating.

'My experiences show that people lack understanding and say: could it be this, could it be that, have you tried swallowing silver. Then they read something in the newspaper. They come up with the strangest examples. I am not interested.' (Female, age 65)

However, one participant appreciated receiving suggestions.

'People come up with suggestions which I have always been grateful for. I save them on the computer. Sometimes things come together. For instance, based on the suggestions I decided to go to clinic [name].' (Female, age 51)

1.5 Lacking recognition from conventional medical community

Participants also reported a lack of recognition from the conventional medical community. Apart from receiving treatment, they also yearned for recognition from the regular medical community (6/11). However, in contrast to treatment, they do not seem to be actively searching for recognition. The impression is given that many are no longer searching for proof or the confirmation from conventional medicine. The youngest participant answered the question: are you still looking for the official diagnosis Lyme disease? As follows:

'No, not for myself. I hope it will come in the future that conventional medicine will help. You do not have to search by yourself. But for me, it is no longer necessary. I have lost hope there.' (Female, age 21)

It is unclear in which context recognition is meant. On the one hand, based on participants' accounts, a lack of recognition is associated with the participants sensing that the medical community does not take their complaints seriously; perceiving them as exaggerating their complaints. As two participants reported:

'It is 80% recognition I am not crazy and that what I have claimed was all true because now I do not feel recognised.' (Female, age 51)

'It would be nice if there would be recognition. That chronic Lyme exists. Then you have something [...]. If only I had cancer or multiple sclerosis because then I would be taken seriously.' (Female, age 47)

On the other hand, based on the participants' narratives in its totality, a lack of recognition may also be associated with an absence in the medical community of a coherent approach to diagnosing Lyme disease. This relates back to the previous section stating there are disparities concerning the diagnosis Lyme among healthcare providers.

Furthermore, a lack of recognition may also be associated with not receiving compensation for the financial costs made regarding diagnostic tests and treatments from the medical insurance companies. Besides, not receiving recognition from the UWV (Employee Insurance Agency) and so not eligible for employment benefits.

'Actually I want to see something on paper from the medical community that I can use against the UWV [...]. They want me to work for 20 hours per week [...]. I can't as all my energy is gone after being busy for 4 hours at home, including my regular activities and personal care' (Female, age 62)

Whatever the case may be, a few participants (4/11) reported that other diseases in which cancer was mainly used as an example, as more recognised in society. This illustrates that some participants explicitly stated they would have preferred a different diagnosis; a disease which is accepted in society.

'At times I have considered. Maybe a wicked thought. If I had cancer, then I would have been taken seriously by hospitals, my surroundings and it was less unexplained. This is because cancer is more accepted in society, there are more treatments available and it is acknowledged by the UWV. If you have MUPS or Lyme, you constantly have to fight and defend yourself.' (Female, age 43)

2. New Roles

2.1 Distancing from conventional medicine

All sought therapy first through conventional medicine, via their primary-care provider often followed by visiting specialists (i.e. neurologist, etc.). However, at a certain moment, all participants except one, reported seeking treatment via complementary/alternative medicine. They appear to be unsatisfied with conventional medicine in the Netherlands. This was triggered by negative encounters with conventional medicine including not receiving treatment, insufficient alleviation of the complaints from antibiotics or the participants' realisation of the damaging effects of antibiotics. A wide range of alternative treatments was

mentioned including homeopathy, bioresonance, herbalism, Ayurveda, naturopathy and acupuncture. Almost all participants stated that the treatments were effective in reducing their complaints. Currently, (4/11) are being treated through alternative medicine. Some (3/11) are being treated with antibiotics and the rest is not being treated.

2.2 Becoming their own healthcare provider

Many participants have taken a healthcare provider's role, becoming their own advocates opting for an independent approach by distancing from the conventional medical. This can be concluded as the majority of the participants have sought out to complementary medicine, going abroad or embarked on a self-treatment approach based on their own initiative. Some participants decided to be treated abroad, in Germany (5/11). Some participants engaged in self-treatment. This included two participants who took the matter into their own hands by importing antibiotics from abroad. One participant also purchased antibiotics through the internet. Additionally, the majority have adjusted their lifestyle by adapting their diet, carrying out physical exercise and/or consuming supplements which help alleviate complaints (7/11). Few respondents even explicitly stated that they have become their own healthcare provider (5/11).

'I became, in fact, my own GP because I am forced to.' (Female, age 47)

'I am my own account manager. I know when. I know the symptoms and I set my own diagnosis.' (Female, age 65)

'I have ensured that I got better. I deserve an honorary medal as I do not have a diagnosis, no doctor, made financial losses and here I am now. Nobody who helped me.' (Female, age 51)

2.3 Learning to cope

Many participants have implemented coping strategies. Some reported they have learned to accept/deal with their limitations/current health situation (5/11). Acceptance is perceived as an essential coping strategy as illustrated by the following quote:

'I have always said you need to accept your situation. You have to accept that you are ill, you must accept that you had bad days. You should not be angry or sad, although it will occur. But the basic principle is always continue seeking for quality of life.' (Male, age 42)

However, it appears that all participants have not given up the search for treatments that help to alleviate their complaints. Another coping strategy is the acquisition of knowledge. Many have become lay experts in Lyme disease by reading scientific literature and books, some have developed their own medical

file; keeping track of all relevant information concerning their health. One participant also writes down treatment suggestions from her surroundings.

'I have an entire file where I keep track of my complaints, when I had them, what I have done, how extreme these were. A list of about 6 pages. I also keep a list of all the medications I received. It used to be a minor list. Later it became more extensive. And what I have done, my contacts, the results of the tests [...].' (Female, age 51)

Some participants visited Lyme gatherings, expressing dismay upon seeing the physical restrictions of their peers. The participants put their situation into perspective adopting selective comparisons to others who are sicker. One participant noted:

'I am grateful that I can go outside and ride my bicycle as some Lyme patients are in a wheelchair or bedridden and cannot do anything' (Female, age 62)

2.4 Appearing receptive to a holistic treatment approach

Participants appear to be drawn to a holistic treatment approach. This consists of combining multiple specialisations in complementary (i.e. herbalism) and mainstream medicine (i.e. antibiotics, physiotherapy) as well as lifestyle adaptations (i.e. physical exercise and nutrition) (8/11). This is perhaps because of their past experiences with these three different treatment approaches. Firstly, many respondents reported symptomatic relief while consuming antibiotics (6/11) yet a few also mentioned that (some) antibiotic treatments were ineffective or only partially reduced their complaints (5/11). Nonetheless, it is difficult to ascertain whether the participants are proponents or opponents of (long-term) antibiotics. On the one hand, many participants stated they preferred not (only) using (long-term) antibiotics (7/11). On the other hand, many have taken or currently are still using antibiotics. Only one participant was persistent to receiving antibiotic treatment for Lyme.

Secondly, almost all participants stated that complementary medicine was effective in reducing their complaints.

Thirdly, besides antibiotic treatment, participants also received other treatments offered in conventional medicine. Four participants stated receiving treatment by a physiotherapist which was reported to be effective. Some participants had also been to a psychologist or psychiatrist in the past (5/11) as suggested by their healthcare provider. In all cases, the participants stated they benefitted from psychological treatment. They often decided to follow the treatment for different reasons. In the beginning of the complaints, some received the diagnosis burn-out and so psychological treatment was a logical approach. Others were willing to do anything to improve their health situation. All reported that these sessions were helpful either as processing personal issues not directly related to their complaints (i.e. childhood issues) or dealing with and accepting their complaints. However, the complaints still persisted.

A broader, holistic treatment approach may also consist of psychological guidance. Many stated that in general psychological treatment can be helpful under certain conditions. They were aware that both the physical and mental wellbeing are interconnected, particularly in a state of ill health. Whenever obstacles are encountered due to sickness, psychological guidance can be useful to help with managing and adapting to the new health situation. It is obvious from the participant's narratives that psychological help should act as a supportive therapy and not considered as the main treatment. However, many did explicitly say that currently, they have no interest in psychological treatment although this question was not asked to all participants. As one participant summarised:

'Psychological support if you're ill is not wrong. You have to deal with a lot of sorrow and you carry a psychologically heavy burden. Of course being ill has a significant effect mentally as you cannot work, socialise or go on vacation. So support concerning finding a daily rhythm is a good idea'. (Female, age 43)

During the interviews, questions were asked about their perceptions of Medically Unexplained Physical Symptoms. Most participants have heard of the term MUPS (8/11). However, they admitted they barely know what it entails. They associated MUPS with a place where complaints will be perceived hence treated as psychological (7/11). Other perceptions by individual participants included: no room for antibiotic treatment, being hospitalised, a type of revalidation centre, uncertified doctors and receiving cognitive behavioural therapy. The majority have not been offered to go to MUPS clinic. Only one participant was advised yet declined and another took the initiative to get treatment from a MUPS clinic. However, only negative experiences were reported as it enhanced the label that their complaints were psychological. It appears that the participants are not against psychological guidance unless the healthcare provider implies the complaints have a psychological cause.

Currently, all respondents are uninterested in receiving MUPS treatment or as some state it would not provide any 'added value' (4/11). Numerous reasons were given. Firstly, many participants were satisfied with the way they cope with their complaints, either through lifestyle changes, alternative medicine or the use of antibiotics (7/11). Secondly, some participants stated they already have taken a similar treatment approach by receiving therapy from various disciplines (5/11). Thirdly, many also believed that MUPS lacks the physical component (5/11) and so may not acknowledge the physical complaints (4/11). The fourth reason for being unreceptive is because they are convinced that the complaints are accountable and caused by Lyme (4/11). Fifthly, a few stated they perceive themselves as psychologically healthy (4/11). Finally, a few participants viewed MUPS as a so-called 'waste pit' (3/11).

What are the consequences of a contested Lyme disease on the primary-care provider relationship?

A contested diagnosis had multiple consequences on the primary-care provider relationship. Generally, the participants reported negative experiences with their primary-care provider. This prompted the

participants to adopt a new role; a transformation in the relationship as well as the respondents' desire for a two-way relationship.

1. Impacts

1.1 Ambivalence towards primary-care provider

Overall, GP experiences were perceived as negative and some respondents had switched GP (4/11). In particular, participants felt they were not taken seriously by their GP's. The participants felt abandoned as they missed their cooperation. GP's did not think proactively or gave leads. Many participants felt that their complaints were disbelieved and GP's did not acknowledge participants' worriedness. This was possibly exacerbated by inconsistent diagnoses between healthcare providers; when a Lyme test carried out by another healthcare provider gave a positive result yet the GP decided not to prescribe antibiotics. Also, some participants criticised GP's for believed their complaints had a psychological cause. This impression arose when a healthcare provider discarded Lyme disease, when no physical cause could be found or when the provider referred the participant to a neurologist or psychologist.

Nevertheless, the participants do not condemn them (6/11) for two reasons. Firstly, many participants reported that their primary-care provider lacked knowledge about Lyme disease (6/11). The respondents believe GP's have received too little information about Lyme disease throughout their education (3/11). A respondent stated that their lack of knowledge is due to GP's prioritisation of becoming specialists for a specific disease caused by a lack of time and wide-ranging diseases that exist. Secondly, two other respondents also accept that their primary-care provider needs to adhere to the medical protocol for diagnosing and treating Lyme.

'I have never blamed my GP because they get far insufficient information [about Lyme] in their study.'
(Female, age 47)

2. New Roles

2.1 Altering relationship

It appears the relationship between the participants and their GP has altered. The relationship currently appears to be instrumental (i.e. a functional relationship aimed at achieving goals) and the expressive (i.e. a social relationship based on emotional support) component is reduced. At the start of the complaints, the participants relied on their GP's medical know-how, expecting them to provide a well-defined clarification of their complaints. However, the absence of a diagnosis and the negative encounters participants underwent possibly led to a transformation in the GP-participant relationship. Many respondents reported they no longer visit their GP for Lyme (6/11), which links with the theme that participants become their own healthcare provider and distance themselves from conventional medicine.

'The GP is useless. I go there for other things. For Lyme, you don't go the GP.' (Female, age 65)

The relationship some participants currently have with their GP is functional, based on the GP carrying out specific participant requests or for medical check-ups (3/11).

'I explained that every once in a while I want to carry out blood tests. He accepted and ever since the relationship has improved.' (Male, age 42)

'I basically do not have to come. She simply prepares the referral. I say I want to go to this laboratory as I want to test this. Then it's done.' (Female, age 47)

2.2 Desiring a partnership

The participants' experiences with their GP also have preceded to a transformation in participant-GP expectations. The impression is given that participants desire a committed GP who adopts a tailored approach involving a patient-provider partnership. However, there appears to be a paradox. The participants lack trust in the expertise of their GP, expect them to meet their desires and also are distancing from them. However, at the same time the participants are dependent on their GP, desiring an in-depth investigation and perhaps realising their GP is a gatekeeper for treatment in conventional medicine.

The respondents explicitly reported what they would have desired from their primary-care provider. Firstly, the majority wanted their GP to have acted proactively by providing leads including carrying out more medical research to provide clarity of their complaints (5/11) but also thinking what next steps can be taken (3/11). In the following quotes, three participants expressed their desires.

'That she would cooperate. Okay, so you are tired, which diseases cause fatigue? Let's look at your health in the past. What have you had? Then we can test Hepatitis since you have had that or Pfeiffer which you have also had. That is what I expected. Not that I need to nag and ask perhaps we need to look at parasites in my intestines. Invent something.' (Male, age 59)

'I think it is your (the GP's) job to give me clues where I have to go. If you say that you lack knowledge (of Lyme), I don't want anything to do with it. Give me suggestions where I need to be.' (Male, age 42)

'I appreciate if you can brainstorm together. That he explains how the medical world is put together. But also give ideas I have not suggested. Even if I come up with an idea, he does not have to agree as I do not always agree with him either. But what I do want is cooperation.' (Female, age 37)

Secondly, some participants also mentioned they would have liked their GP's to have a flexible attitude towards the protocol of treating Lyme disease and make exceptions whenever necessary (4/11). In other words, being willing to subscribe long-term antibiotic treatment (at least more than 2 weeks as is stated

in the protocol). Thirdly, a few wanted their GP to at least make an attempt to broaden their knowledge about Lyme disease (3/11).

It is important to mention that some participants also reported positive experiences with individual healthcare providers (specialists and GP's). These were healthcare providers who had experience with Lyme or patients with chronic complaints (4/11), were willing to carry out in-depth medical research (2/11), prepared to deviate from the protocol and provide treatment (2/11) and who were receptive to complementary medicine (1/11).

Discussion

This study aimed to understand the value of the diagnosis Lyme through analysing the illness experiences of people with persistent uncharacteristic complaints who self-identify as having Lyme disease. The results show that the diagnosis Lyme disease is contested; there are conflicting illness beliefs between participants and their healthcare providers. However, this is further intensified due to discrepancies among the healthcare community. This lack of consensus between physicians creates unclarity, hence facilitates self-diagnosis by reinforcing participants' beliefs that Lyme is the cause of their complaints. This has various consequences. Apart from the negative impacts on their lives, it forces participants to adopt a new role, including engaging searching for alternative treatments and becoming their own healthcare provider as well as altering the relationship with their primary-care provider. This illustrates the emergence of 'patient power', whereby the participants attempt to gain control over their complaints, creating a power struggle between themselves and their primary-care provider.

The diagnosis Lyme disease is valuable for participants with persistent uncharacteristic complaints because they continue to believe their complaints can be attributed to Lyme disease. On the one hand, even if the diagnosis is contested, the participants have had the ability to gain access to (short-term) treatments which have partially reduced the complaints. Furthermore, it provided some clarity for themselves and their surroundings. In addition, the participants had the implicit desire to recount their experiences in detail. Their narratives show their continuous involvement, struggle and perseverance for a confirmation of the diagnosis Lyme disease. On the other hand, it is considered predominantly disabling as the diagnosis is contested which, based on the narratives, had many negative impacts on their lives and primary-care provider relationships. On the whole, the participants appeared to desire a diagnosis. Their preference is the diagnosis Lyme disease, which would validate their belief. However, the participants appear open to an alternative diagnosis provided it is an uncontested illness that ultimately clarifies their complaints and coupled with treatments to help improve their quality of life.

The myriad of consequences of a contested diagnosis Lyme disease found in this study share many similarities to themes previously identified in qualitative studies in the USA concerning people with Chronic Lyme disease (CLD). A small phenomenological study by Drew & Hewitt (2006) identified six themes in

which comparable results were found. These included delayed diagnosis whereby participants underwent multiple diagnostic tests and visited many healthcare specialists, dealing with financial stress and becoming their own advocates in terms of self-diagnosis through identifying with the symptoms listed on the internet and discussing the possibility of Lyme disease with their healthcare provider. Another study using the dimensions of the Health Belief Model revealed that CLD affected people's social and physical functioning, created future uncertainty, diverging encounters with healthcare providers and the respondents seeking unconventional treatment approaches such as alternative medicine as well as being drawn to a holistic treatment approach (Ali et al., 2014). A more recent study revealed similar findings including physical and social limitations leading to a 'new normal' state by adopting coping strategies as well as future doubt, which was related to 'interphysician subjectivity' meaning participants receiving different diagnoses and treatments from different physicians. Finally, the study showed how people endured from multiple invisible complaints such as fatigue, pain and headaches. This invisibility was perceived as affecting social support (Rebman et al., 2015).

Some findings of this thesis also relate to the grey literature studies mentioned in the introduction. Participants in a focus group study carried out by the Health Council of the Netherlands (2013), reported the need for a clear diagnosis. As quoted, 'they all too often involve uncertainties about the results and hence the diagnosis' (p. 56). This illustrates what the present report calls a contested diagnosis Lyme disease and the consequences on the participants. Additional findings congruent with other studies include participants experiencing healthcare providers failing to take them seriously and their lack of knowledge, undergoing a lack of recognition in society, seeking care abroad, switching to alternative medicine and desiring a multidisciplinary approach. Moreover, the participants being open towards involving a psychologist but only as a part of a combined treatment approach (Health Council of the Netherlands, 2013)

However, this study also identified additional themes and dissimilar findings. These include a lack of social validation from their surroundings, and becoming their own healthcare provider in terms of seeking treatment. Furthermore, it highlights the consequences on the relationship with their primary-care provider. In addition, the previous studies also revealed the value of the diagnosis Lyme disease for people with CLD. These included relief, validation and optimism for recovery (Drew & Hewitt, 2006; Ali et al., 2014). However, this study does not completely support these findings. For some participants, the diagnosis provided relief, however only temporarily. Also, the diagnosis did provide clarity for themselves and their surroundings, yet participants still sensed a lack of validation. Furthermore, the grey literature studies indicate that people expressed a desire for prolonged antibiotic treatment. However, this thesis cannot accurately deduce whether this is the case as the on the one hand many participants stated they preferred not (only) using (long-term antibiotics). On the other hand, many have taken or currently are still using antibiotics.

Based on Frank's typology of illness narratives (1995), the stories of the participants have features of the *restitution*, *chaos* and *quest* narratives regarding the content of the interviews. The *restitution* narrative is not often found among chronically ill people as the story precedes as follows: determining the cause, seeking help and treating the condition in order to return to health. Nonetheless, in the beginning, most participants in

this study started with a *restitution* narrative. At the onset of their complaints, most attributed their symptoms to a possible minor acute illness, expecting to recover.

However, as the participants continued explaining their past illness journeys, these shifted to *chaos* narratives, which typically lacks structure whereby the story misses a clear beginning, middle and ending. A possible explanation is that symptoms became more severe resulting in physical and social consequences, yet the origin remained ambiguous. The participants visited countless healthcare providers, had done many tests and received different treatments over a long period of time all which created unclarity, thus sustaining the *chaos* narrative. Furthermore, retrospectively reflecting on the interview process, the *chaos* narratives were evident as the narratives were often disconnected with sudden recollections not related to the interview questions and often recounted without a clear sequence. Many participants were open-hearted and described their illness journey in detail. Sometimes the participants were so immersed in their own story, the interview question was unanswered or needed to be repeated. Also, it seemed that they longed for someone to listen and show interest in their story.

As the interview progressed to focus on their current health situation, nuanced *quest* narratives were observed, whereby the person achieved personal gain from the illness experience, such as self-awareness. The majority currently consider themselves in a reasonable health state. They attempted to make sense of their illness experience by searching for coping mechanisms to manage their complaints. These included, inter alia, seeking treatment from alternative medicine, becoming their own healthcare provider by adapting their lifestyle, keeping a medical file, expanding their knowledge on Lyme disease and putting their situation into perspective by comparing to others.

When reflecting on the sociology of diagnosis theory, it highlights that a diagnosis has several roles for people. These include the access to treatment, predicting prognosis and obtaining benefits. It also enables a person to make sense of their illness experience, provide relief by reducing uncertainty and being taken seriously by their surroundings (Jutel, 2009; Rosenburg, 2002; Brown, 1995). Congruent with the theory, the participants attempted to pursue these roles, yet were unsuccessful. Even though the diagnosis Lyme provided an explanation and rearranged their roles, the participants did not enjoy the benefits of a diagnostic label. In general, the diagnosis Lyme disease had a predominantly disabling role. Most participants did not receive treatment from conventional medicine and had no access to health and employment benefits. Furthermore, they faced daily and future uncertainty, lacked social validation from their personal surroundings and recognition from the medical community.

In other words, the participants did not gain access to the 'sick role'. This concept developed by Parsons in the 1950s consists of two rights and responsibilities. The rights include being exempted from particular duties over a period of time and not regarded accountable for their sickness. However, the person is responsible for cooperating with medical help and desiring to recover (Segall, 1976). Access to the sick role can only be achieved if a diagnosis is given and so it becomes legitimised. The gatekeeper is the healthcare provider in conventional medicine, which often begins at the GP. As this study shows, a contested diagnosis Lyme disease does not legitimise illness, hence prevents access to the sick role. Consequently, the participants

were excluded from receiving treatment and benefits, are held personally responsible for their health state and do not gain social support, hence illustrates the power of the healthcare provider. This was also shown in another study on chronic back pain sufferers striving for the sick role (Glenton, 2003). Although the participants in this thesis adopted a new role, they were aware that they remained dependent on their primary-care provider.

The possible underlying explanation why participants do not gain access to the sick role and so not obtain access to the functions of a diagnosis, is that Lyme disease is a medically unexplained or contested illness. A contested illness shares the following features: i) the aetiology of the symptoms is uncertain. The causality is disputed whether it is physical or psychological ii) the illness is linked to other diagnoses and comorbid conditions which exhibit similar symptoms, iii) there is a lack of available fitting treatment regimens and iv) the legal classification is questioned, affecting access to healthcare benefits including insurance reimbursement (Dumit, 2006). In other words, contested illnesses have a controversial status, also known as ‘illnesses you have to fight to get’ (Dumit, 2006). As summarised by Conrad & Barker (2010), ‘they are illnesses where sufferers claim to have a specific disease that many physicians do not recognise or acknowledge as distinctly medical’. Consequently, this results in a clash between the medical view and individuals’ illness experiences living with ongoing medically unexplained symptoms.

A significant amount of literature is available on illness experiences of people with medically unexplained symptoms and contested illnesses such as chronic fatigue syndrome (CFS), Myalgic Encephalomyelitis (ME) and fibromyalgia. Nettleton (2006) identified three impacts of living with MUS from previous research: i) living with uncertainty, ii) issues of receiving legitimacy and iii) resistance to a psychological association of their complaints. A meta-synthesis on the subjective experience of fibromyalgia showed that informants suffered from the invisibility of their symptoms, namely pain and fatigue. They sensed future uncertainty, sensing illegitimacy, experiencing changes in their life situation (i.e. employment, social responsibilities, etc.) and employing various coping strategies (Sim & Madden, 2008). A meta-synthesis of qualitative studies on myalgic encephalomyelitis and chronic fatigue syndrome showed a change in identity, issues of legitimacy, reductions across physical, social and economic domain and the adoption of coping mechanisms (Larun & Malterud, 2007; Anderson, Jason, Hlavaty, Porter & Cudia, 2012).

The experiences of the participants in this study overlap significantly with the illness experiences of people with contested illnesses. However, the diagnosis Lyme disease is also unique as it can be both an uncontested and contested illness. On the one hand, Lyme disease can be identified and treated quickly. In this case, it is uncontested as i) people may develop symptoms that are characteristic of the disease, such as the occurrence of an erythema migrans ii) the cause is known, due to a bite of a tick and iii) there is a treatment regimen available, namely antibiotic therapy. On the other hand, Lyme disease can be chronic if the infection remains unnoticed/ untreated or complaints persist even after antibiotic treatment. It becomes contested since an accurate diagnosis cannot be established as i) still cause significant false-positive and false-negative results and ii) clinical symptoms resemble other physical or psychological causes. Currently, there is no clear

treatment approach as long-term antibiotic treatment is not recommended. Furthermore, there appears to be a lack of access to welfare benefits such as employment and insurance coverage.

Practical Recommendations

What this study shows is that Lyme disease is generally an uncontested illness but can become a contested illness, with significant negative consequences on participant lives and the primary-care provider relationship. It reveals that Lyme disease becomes an empty diagnosis as the anticipated role of a diagnosis which includes access to treatment, obtaining benefits and being taken seriously are unaccomplished. The lack of consensus between physicians means that persons with persistent uncharacteristic complaints do not receive permission to be ill with all the associated consequences on their lives and primary-care provider relationship.

One of the main issues is the lack of clarity and inconsistent diagnoses among the healthcare community. This has consequences on persons with persistent uncharacteristic complaints as they bear the brunt. They may not be treated for Lyme disease and therefore develop chronic symptoms or the actual cause of the symptoms are not treated and the person continues living with complaints. Also, society as a whole is affected. It leads to inefficient healthcare; the loss of time and money through persons seeking care from different specialists and carrying out different tests.

In order to lessen these consequences, specific suggestions can be made concerning communication between i) specialists and primary-care providers and ii) patients and primary-care providers. Specialists and primary-care providers may communicate more systematically to discuss individual cases to determine a diagnosis and subsequently treatment, whereby a patient receives a consistent and credible explanation. This may give the patient the impression they are taken seriously and increases the integrity of the outcome.

Also, improvements can be made regarding patient-primary-care provider communication. GP's have a crucial role as they are the gatekeepers in legitimising their patients' complaints hence giving access to the sick role and subsequently to treatment, etc. Within the scope of this study, improvements can be made regarding primary-care provider communication to people with persistent uncharacteristic complaints. Firstly, a GP should communicate clearly how the diagnosis Lyme is established (listening to their complaints and taking into consideration their case history, clinical picture and tests) and being transparent in why this approach is chosen (i.e. unreliability of the tests). Secondly, being as clear as possible if the GP believes the complaints are caused by Lyme (or not) and provide a clear explanation why the probability is low or high. Thirdly, state that psychological counselling does not indicate that the GP interprets the patients' complaints as 'in the mind' or that the cause is psychological but possibly as a source of guidance. All in all, a communication approach that conveys their patients are taken seriously, yet the primary-care provider remains in control of the diagnosis and treatment. These recommendations aim to reduce the probability that patients feel forced to seek treatment on their own as well as have confidence in their primary-care provider and healthcare community.

In the long-term, it is recommended that the healthcare community improves patient-provider communication by providing a more accurate and consistent diagnosis. As the RIVM and the Health Council

of the Netherlands previously announced, and which this study once again emphasises, is the following. Firstly, there is a need for the standardisation of Lyme tests used in the Netherlands in order to prevent different test results coming from different healthcare providers (Health Council of the Netherlands 2013). Reliable tests are crucial for people as the findings show that they decide whether a person obtains the label Lyme, thus are entitled to treatment. Secondly, due to the lack of reliability of serological tests, these should not be decisive in diagnosing Lyme disease. The RIVM stresses a more prompt recognition. A combined diagnostic approach should be adopted which includes, chronologically, first clinical examination followed by case history and ultimately the blood values from the laboratory test (RIVM, 2016c). Lastly, the following question arises: if Lyme disease is suspected and verified through a persons' narrative or clinical symptoms but not according the Lyme test, should healthcare providers prescribe short-term antibiotic treatment? This is because as this study shows, proceeding to a chronic state that may be caused by Lyme disease has multiple consequences on people's lives and their primary-care provider relationship.

Future Research

Future studies directly related to this thesis can be carried out. This includes recruiting a larger sample size and analysing more specific themes related to patient-primary-care provider relationships. Furthermore, in order to validate the communication recommendations given in the previous section, it is suggested to carry out a study which includes the views of GP's in order to compare and contrast the perspectives between patients and primary-care providers to develop more concrete communication recommendations.

In addition, more biomedical studies can be conducted. This includes determining the laboratory tests that are most reliable (produces the least false-positive and false-negative results) in determining the antibodies the body produces against Lyme. Related to this, a study or a sociological debate may be initiated involving the general function of Lyme tests in the medical community. Whether laboratory tests in general act as conclusive evidence for getting a diagnosis and if so, whether this is medically and socially desirable. This is debatable since, in the case of Lyme disease, a test measures the body's response to the bacterium. Therefore, a positive result means the body produces antibodies against the bacterium yet this does not imply a person has Lyme disease. This is because antibodies can remain detectable for many years after the bacterium has been removed. Furthermore, in approximately 5% of healthy people have antibodies against the bacterium in their blood (RIVM, 2016c).

Finally, a study is needed to look into the consequences of primary-care providers being more flexible in subscribing a short-term antibiotic treatment for Lyme disease. This involves finding a balance between the consequences of risking delayed treatment and developing chronic complaints which may or may not be attributed to Lyme disease and the consequences of possible health risks (i.e. side-effects) and antimicrobial resistance.

Strengths and Limitations

This study attempts to fill the knowledge gap concerning the impact of Lyme disease on people's lives (Broerse & den Oudendammer, 2013). It strives to shed light on the processes by which persons with persistent uncharacteristic complaints conceptualise these to Lyme disease and how a contested diagnosis Lyme disease shape participants' illness experience and subsequent quality of life. Many participants were open-hearted and described their illness journey in detail. Nevertheless, there are a number of methodological limitations to this study that needs to be addressed. Firstly, the small number of eligible participants who were purposefully recruited from two Lyme organisations in the Netherlands. The small sample size, homogeneous characteristics and participants who were willing and able to partake in the study limits the generalisability of the findings to more diverse groups of participants who may have experienced different illness trajectories than those reported in this study.

Secondly, it cannot be fully excluded that some of the participants in this study may be grouped as PSLB or post-Lyme disease persons; who have had early or late disseminated LB (not identified or diagnosed by their healthcare provider) and experience persistent uncharacteristic complaints after antibiotic treatment which may indicate residual damage.

Thirdly, semi-structured interviews were carried out which may have some limitations. Participants did not describe their illness experience in a comparable way, making it challenging to explain their journey chronologically. Also, there were individual differences concerning probing questions asked, which resulted in some themes being discussed in more depth than others. Another limitation from the semi-structured interviews is that a broad range of themes was discussed. On the one hand, it provided a wide range of insights of participants' illness experience. The study identified various consequences of a contested diagnosis and the value of the diagnosis Lyme disease. On the other hand, more in-depth research would be needed to explore specific themes.

Fourthly, concerning the interview questions, there is room for improvement. Questions that were missing which were potentially valuable and could make the analysis more convincing include 'why are you a member of a Lyme organisation 'and' what are your current goals'? A pilot study may have facilitated in enhancing the validity of the study.

Fifthly, another limitation is that the study was patient-centred, focusing on their perceptions and experiences with their primary-care provider. As a result, the information is one-sided and the voice of the healthcare providers is not included. It would have been interesting to compare the results with the narratives of the primary-care providers. This would also help further determine how communication between the patient and primary-care provider as well as specialists can be improved.

Lastly, the study focused on participant retrospective experiences. This may lead to memory bias as the participants may not recall particular experiences or currently assign a different meaning to their past experiences.

Conclusion

In this study, persons with persistent uncharacteristic complaints ascribe them to Lyme disease. However, there are discrepancies between the beliefs of the individuals and their healthcare providers. Furthermore, a lack of consensus concerning the diagnostic procedures and treatment approaches for Lyme among healthcare providers enhances diagnostic ambiguity and reinforces the participants' self-diagnosis Lyme disease. Subsequently, Lyme disease becomes a curse, a contested illness with many consequences. It has significant negative impacts on the participants' lives and initiates the adoption of new roles including seeking alternative treatment approaches. It also leads to a shift towards a strained and instrumental patient-primary-care provider relationship. Nonetheless, participants desire a tailored partnership. Despite the contested nature of the diagnosis Lyme, it is a blessing, a source of value for the participants. It gives their complaints a name which has enabled them to gain treatment and serves as a justification for themselves and their surroundings. Participants desire a diagnosis. They remain dependent on their primary-care providers as they are the gatekeepers in legitimising their complaints, hence giving access to the sick role and subsequently to the various functions of a diagnosis in society. Therefore, it is critical to foster good patient-primary-care provider relationships which require improvements in communication between patients and primary-care providers and specialists and primary-care providers. Furthermore, to improve patient satisfaction in the healthcare community there is a need for simultaneous actions including the standardisation of tests, the adoption of a combined diagnostic approach and the evaluation of the benefits and limitations of a more flexible prescription of short-term antibiotics against Lyme. As a result, to help to provide better care and help their patients improve their quality of life.

References

- Academisch Medisch Centrum (AMC). Lyme en het AMC. Retrieved from <https://www.amc.nl/web/Het-AMC/Afdelingen/Medische-afdelingen/Amsterdam-Multidisciplinair-Lyme-Centrum-AMLC/Amsterdam-Multidisciplinair-Lyme-Centrum-AMLC/Lyme-en-het-AMC/Lyme-en-het-AMC.htm> on 04 April 2016.
- Ali, A., Vitulano, L., Lee, R., Weiss, T. R., & Colson, E. R. (2014). Experiences of patients identifying with chronic Lyme disease in the healthcare system: a qualitative study. *BMC family practice*, *15*(1), 1.
- Anderson, V. R., Jason, L. A., Hlavaty, L. E., Porter, N., & Cudia, J. (2012). A review and meta-synthesis of qualitative studies on myalgic encephalomyelitis/chronic fatigue syndrome. *Patient education and counseling*, *86*(2), 147-155.
- Berende, A., ter Hofstede, H. J., Vos, F. J., van Middendorp, H., Vogelaar, M. L., Tromp, M., ... & Kullberg, B. J. (2016). Randomized Trial of Longer-Term Therapy for Symptoms Attributed to Lyme Disease. *New England Journal of Medicine*, *374*(13), 1209-1220.
- Blaxter, M. (1978). Diagnosis as category and process: the case of alcoholism. *Social Science & Medicine. Part A: Medical Psychology & Medical Sociology*, *12*, 9-17.
- Broerse, J.E.W & den Oudendam, W.M. (2013). Onderzoeksagenda ziekte van Lyme Behoeften van patiënten, hoogrisicogroepen en zorgverleners. Athena Instituut, Vrije Universiteit Amsterdam. Retrieved from https://www.gezondheidsraad.nl/sites/default/files/achtergrond_Lyme_onder_loep_A1301_1.pdf
- Brown, P. (1995). Naming and framing: the social construction of diagnosis and illness. *Journal of Health and Social Behavior*, 34-52.
- Centraal BegeleidingsOrgaan (CBO). (2013). Richtlijn Lymeziekte. Retrieved from <http://www.diliguide.nl/document/1314>
- Conrad, P., & Barker, K. K. (2010). The social construction of illness key insights and policy implications. *Journal of health and social behavior*, *51*(1 suppl), S67-S79.
- Coumou, J., Herkes, E. A., Brouwer, M. C., van de Beek, D., Tas, S. W., Casteelen, G., van Vugt, M., Starink, M.V., de Vries, H.J.C., de Wever, B., Spanjaard, L & Hovius, J.W.R. (2015). Ticking the right boxes: classification of patients suspected of Lyme borreliosis at an academic referral center in the Netherlands. *Clinical Microbiology and Infection*, *21*(4), 368-e11.
- Creswell, J. W. (2013). *Research design: Qualitative, quantitative, and mixed methods approaches*. Sage publications.
- Daker-White, G., Sanders, C., Greenfield, J., Ealing, J., & Payne, K. (2011). Getting a diagnosis v. learning to live with it? The case of the progressive ataxias. *Chronic illness*, *7*(2), 120-133.
- Drew, D., & Hewitt, H. (2006). A qualitative approach to understanding patients' diagnosis of Lyme disease. *Public Health Nursing*, *23*(1), 20-26.

- Dumit, J. (2006). Illnesses you have to fight to get: Facts as forces in uncertain, emergent illnesses. *Social science & medicine*, 62(3), 577-590.
- Frank, A. W. (2013). *The wounded storyteller: Body, illness, and ethics*. University of Chicago Press.
- Glenton, C. (2003). Chronic back pain sufferers—striving for the sick role. *Social science & medicine*, 57(11), 2243-2252.
- Hassett, A. L., Radvanski, D. C., Buyske, S., Savage, S. V., & Sigal, L. H. (2009). Psychiatric comorbidity and other psychological factors in patients with “chronic Lyme disease”. *The American journal of medicine*, 122(9), 843-850.
- Health Council of the Netherlands. A closer look at Lyme disease. The Hague. Health Council of the Netherlands, 2013; publication no. 2013/12E.
- Huibers, M. J., & Wessely, S. (2006). The act of diagnosis: pros and cons of labelling chronic fatigue syndrome. *Psychological medicine*, 36(07), 895-900.
- Jutel, A. (2009). Sociology of diagnosis: a preliminary review. *Sociology of health & illness*, 31(2), 278-299.
- Jutel, A. G., & Conrad, P. (2011). *Putting a name to it: Diagnosis in contemporary society*. JHU Press.
- Kornelsen, J., Atkins, C., Brownell, K., & Woollard, R. (2016). The meaning of patient experiences of medically unexplained physical symptoms. *Qualitative health research*, 26(3), 367-376.
- Larun, L., & Malterud, K. (2007). Identity and coping experiences in Chronic Fatigue Syndrome: A synthesis of qualitative studies. *Patient education and counseling*, 69(1), 20-28.
- Madden, S., & Sim, J. (2006). Creating meaning in fibromyalgia syndrome. *Social science & medicine*, 63(11), 2962-2973.
- Nasleep van een tekenbeet: Ziekte van Lyme. (2012, March). *Bio-Wetenschappen en Maatschappij*. Retrieved from <http://bwm.trefcon.nl/wp-content/uploads/2014/07/Ziekte-van-Lyme.pdf>
- Nettleton, S. (2006). ‘I just want permission to be ill’: towards a sociology of medically unexplained symptoms. *Social science & medicine*, 62(5), 1167-1178.
- Nettleton, S., Watt, I., O’Malley, L., & Duffey, P. (2005). Understanding the narratives of people who live with medically unexplained illness. *Patient education and counseling*, 56(2), 205-210.
- Nederlands Huisartsen Genootschap (NHG). NHG-Standaard Somatisch Onvoldoende verklaarde Lichamelijke Klachten (SOLK). Retrieved from <https://www.nhg.org/standaarden/volledig/nhg-standaard-somatisch-onvoldoende-verklaarde-lichamelijke-klachten-solk#idp164816> on 04 April 2016
- NOS. (09 July 2015). Artsen zijn het oneens: heb je Lyme of niet? Retrieved from <http://nos.nl/op3/artikel/2046056-artsen-zijn-het-oneens-heb-je-lyme-of-niet.html>
- Ogden, J., Branson, R., Bryett, A., Campbell, A., Febles, A., Ferguson, I., ... & Tayler, M. (2003). What’s in a name? An experimental study of patients’ views of the impact and function of a diagnosis. *Family Practice*, 20(3), 248-253.
- Ong, L. M., De Haes, J. C., Hoos, A. M., & Lammes, F. B. (1995). Doctor-patient communication: a review of the literature. *Social science & medicine*, 40(7), 903-918.

- Rappaard, B., Luske, B., de Boer, M., Gijsen, M., Hintzen, N. T., van Tilburg, V., & Schraa, G. (2006). *Be aware of the cick: problemen in de diagnose en behandeling van Lyme-borreliose in Nederland* (p. 38). Wageningen University.
- Rebman, A. W., Aucott, J. N., Weinstein, E. R., Bechtold, K. T., Smith, K. C., & Leonard, L. (2015). Living in Limbo Contested Narratives of Patients With Chronic Symptoms Following Lyme Disease. *Qualitative health research*, 1049732315619380.
- Rosenberg, C. E. (2002). The tyranny of diagnosis: specific entities and individual experience. *Milbank Quarterly*, 80(2), 237-260.
- RIVM (19 April 2016a). Vragen en antwoorden tekenbeten en Lyme. Retrieved from http://www.rivm.nl/Documenten_en_publicaties/Algemeen_Actueel/Veelgestelde_vragen/Infectieziekten/ISI_standaarden/ISI_Ziekte_van_Lyme
- RIVM. (11 April 2016b). Ziekte van Lyme-bijeenkomst. Retrieved from http://www.rivm.nl/Documenten_en_publicaties/Algemeen_Actueel/Agenda_Items/Agenda_2016/Ziekte_van_Lyme_bijeenkomst
- RIVM. (25 March 2016c). Labtest alleen niet doorslaggevend bij diagnose Lyme. Retrieved from http://rivm.nl/Documenten_en_publicaties/Algemeen_Actueel/Nieuwsberichten/2016/Labtest_alleen_niet_doorslaggevend_bij_diagnose_Lyme
- Segall, A. (1976). The sick role concept: Understanding illness behavior. *Journal of Health and Social Behavior*, 162-169.
- Sigal, L. H., & Hassett, A. L. (2002). Contributions of societal and geographical environments to " chronic Lyme disease": the psychopathogenesis and aporology of a new " medically unexplained symptoms" syndrome. *Environmental health perspectives*, 110(Suppl 4), 607.
- Sim, J., & Madden, S. (2008). Illness experience in fibromyalgia syndrome: A metasynthesis of qualitative studies. *Social science & medicine*, 67(1), 57-67.
- Sprong, H., Hofhuis, A., Gassner, F., Takken, W., Jacobs, F., Van Vliet, A., Van Ballegooijen, M., Van der Giessen, J & Takumi, K. (2012). Circumstantial evidence for an increase in the total number and activity of Borrelia-infected Ixodes ricinus in the Netherlands. *Parasit Vectors*, 5, 294.
- Steere, A. C., Coburn, J., & Glickstein, L. (2004). The emergence of Lyme disease. *The Journal of clinical investigation*, 113(8), 1093-1101.
- Stichting de Ombudsman & Nederlandse Vereniging voor Lyme patiënten (NVLP). De ziekte van Lyme Een onderschat probleem. Onderzoek naar de gevolgen voor patiënten en maatschappij. (2011). Retrieved from <http://www.tekenbeetziekten.nl/wp-content/uploads/2013/11/Lyme-onderschat-probleem-van-NVLP-en-SdO.pdf>
- Stichting Tekentbeetziekten (STB). Reactie op het Gezondheidsraadadvies. (2013). Retrieved from <http://www.tekenbeetziekten.nl/wp-content/uploads/2013/11/Rapport-Reactie-op-het-Gezondheidsraadadvies-naar-aanleiding-van-het-Burgerinitiatief.pdf>

- Stewart, M. A. (1995). Effective physician-patient communication and health outcomes: a review. *CMAJ: Canadian Medical Association Journal*, 152(9), 1423.
- Street, R. L., Makoul, G., Arora, N. K., & Epstein, R. M. (2009). How does communication heal? Pathways linking clinician-patient communication to health outcomes. *Patient education and counseling*, 74(3), 295-301.
- Thompson, R., Isaac, C. L., Rowse, G., Tooth, C. L., & Reuber, M. (2009). What is it like to receive a diagnosis of nonepileptic seizures?. *Epilepsy & Behavior*, 14(3), 508-515.
- Tijssse-Klasen, E., Jacobs, J. J., Swart, A., Fonville, M., Reimerink, J. H., Brandenburg, A. H., van der Giessen, J.W.B., Hofhuis, A & Sprong, H. (2011). Small risk of developing symptomatic tick-borne diseases following a tick bite in The Netherlands. *Parasit Vectors*, 4(1), 17.
- Trimbos-Institute. (2010). Somatisch Onvoldoende Verklaarde Lichamelijke Klachten en Somatoforme Stoornissen. Retrieved from <http://www.diliguide.nl/document/2833/solk-en-somatoforme-stoornissen.html>
- Undeland, M., & Malterud, K. (2007). The fibromyalgia diagnosis—Hardly helpful for the patients? A qualitative focus group study. *Scandinavian journal of primary health care*, 25(4), 250-255.
- Van den Wijngaard, C. C., Hofhuis, A., Harms, M. G., Haagsma, J. A., Wong, A., de Wit, G. A., Havelaar, A.H., Lugner, A.K., Suijkerbuijk, A.W.M & van Pelt, W. (2015). The burden of Lyme borreliosis expressed in disability-adjusted life years. *The European Journal of Public Health*, 25(6), 1071-1078.
- Vernooij, F & Kobus, Z. (2015). “Pak de Teek, Voordat de Teek Jou Pakt”. Profielwerkstuk over de ziekte van Lyme.
- Woodward, R. V., Broom, D. H., & Legge, D. G. (1995). Diagnosis in chronic illness: disabling or enabling—the case of chronic fatigue syndrome. *Journal of the Royal Society of Medicine*, 88(6), 325-329.
- Zavestoski, S., Brown, P., McCormick, S., Mayer, B., D'Ottavi, M., & Lucove, J. C. (2004). Patient activism and the struggle for diagnosis: Gulf War illnesses and other medically unexplained physical symptoms in the US. *Social Science & Medicine*, 58(1), 161-175.
- Zomer, T. P., et al. "Staat van Zoönosen 2014." *RIVM rapport 2015-0151*. (2014).

Appendix I – Cover letter

Geachte heer/ mevrouw X,

Allereerst wil ik u bedanken voor uw interesse om deel te nemen aan mijn scriptie onderzoek over de ziekte van Lyme. Via (naam) heb ik uw contactgegevens ontvangen. In deze brief wil ik u verder een toelichting geven over dit onderzoek.

Wie ben ik?

Allereerst kort iets over mijzelf. Ik ben Valérie Eijrond, een masterstudente Gezondheid en Maatschappij aan de Wageningen Universiteit. Op dit moment ben ik in mijn laatste fase van mijn studie waarbij ik een masteronderzoek doe onder begeleiding van de Wageningen Universiteit en het RIVM.

Waar gaat mijn onderzoek over?

Zoals u misschien weet is de ziekte van Lyme een veelbesproken onderwerp in Nederland, vooral m.b.t patiënten met langdurige niet-kenmerkende klachten. Het doel van mijn onderzoek is om in kaart te brengen de betekenis van de diagnose Lyme vanuit uw perspectief.

Hoe ga ik dit onderzoeken?

De methode die ik gekozen heb voor de uitvoering van het onderzoek zijn diepte interviews. Ik zal u vragen stellen betreffende Lyme. Onderwerpen die aan bod zullen komen zijn uw ervaringen rondom klachten, het proces dat u heeft doorlopen, de communicatie met uw huisarts en de betekenis van de diagnose Lyme en alternatieve diagnoses.

Wanneer vinden de interviews plaats?

Ik zal het op prijs stellen wanneer het interview zal plaatsvinden in juni 2016. Het interview zal ongeveer 45 tot 60 min duren. Ik kom graag bij u (thuis) langs om het interview af te nemen. Hebt u voorkeur voor een specifiek datum of andere plaats? Dan hoor ik dat graag. Ik zal u deze week bellen om een afspraak te maken. Vanwege de zorgvuldigheid van het onderzoek zal het interview worden opgenomen. Hierbij wil ik benadrukken dat de anonimiteit volledig gewaarborgd zal worden.

Wilt u deelnemen aan het onderzoek?

Ik zal deze week telefonisch contact met u opnemen om een afspraak te maken. Als u nog verdere vragen heeft, meer informatie wilt ontvangen of toch liever niet wilt deelnemen, neem dan gerust contact op via mail (valerie.eijrond@wur.nl) of telefonisch (06 30 36 63 63).

Ik hoop u hiermee voldoende te hebben geïnformeerd en dank u bij voorbaat hartelijk voor uw deelname aan dit onderzoek.

Met vriendelijke groet,

Valérie Eijrond

Studente Wageningen Universiteit

Appendix II – Informed Consent

Toestemmingsformulier:

Onderzoek over de diagnose Lyme door Valérie Eijrond

Dit onderzoek gaat over de betekenis van de diagnose Lyme. U bent benaderd voor een interview. Deelname duurt ongeveer 45-60 minuten.

Het onderzoek is uitsluitend voor academische doeleinden. Daarbij zullen alle gegevens anoniem behandeld worden en dus niet naar de deelnemers te herleiden zijn.

Door middel van het tekenen van deze verklaring geeft u aan voldoende geïnformeerd te zijn over dit onderzoek, stemt u geheel vrijwillig in met deelname aan dit onderzoek en geeft u toestemming voor het anoniem gebruiken van de gegevens uit dit interview.

U kunt zonder opgave van redenen weigeren mee te doen aan het onderzoek of uw deelname voortijdig afbreken. Ook kunt u achteraf (binnen 24 uur na afloop van het interview) uw toestemming intrekken voor het gebruik van uw antwoorden of gegevens voor het onderzoek.

Indien u verdere vragen heeft, kan de onderzoeker (Valérie Eijrond) meer informatie geven.

Naam:

Woonplaats:

Email adres:

Geboortedatum (D/M/J):

Geslacht:

Datum:

Handtekening:

Appendix III – Interview Guide

Introductie (noteer: datum, tijd, locatie, respondent informatie)	
Welkom	<p>Voorstellen</p> <p>Heel fijn dat u mij kan helpen met mijn masteronderzoek</p>
Studie	<p>Doel van het onderzoek: Ik ben een masterstudente Gezondheid en Maatschappij aan de Wageningen Universiteit. Op dit moment ben ik in mijn laatste fase van mijn studie waarbij ik een masteronderzoek doe onder begeleiding van de Wageningen Universiteit en het RIVM.</p> <p>Mijn onderzoek gaat over de ziekte van Lyme en daarom wil graag eens met u praten over uw ervaringen met deze ziekte. Onderwerpen die aan bod zullen komen zijn uw ervaringen rondom klachten, het proces dat u heeft doorlopen, de communicatie met uw huisarts en de betekenis van de diagnose Lyme en alternatieve diagnoses.</p> <p>Het is echt een open gesprek dus u mag zo uitgebreid mogelijk de vragen kan beantwoorden. Er zijn geen goede of foute antwoorden.</p> <p>Het interview zal ongeveer drie kwartier duren.</p>
Privacy	<p>Bedankt voor het tekenen van het toestemmingsformulier (informed consent). Ik wil nog graag benadrukken dat alle antwoorden worden vertrouwelijk behandeld. Ook kunt u te allen tijde stoppen met het interview, zonder dat u hoeft uit te leggen waarom. Heeft u verder nog vragen hierover?</p> <p>Vanwege de zorgvuldigheid van het onderzoek zal ik graag de interview willen opnemen. Vind u dat goed?</p>

(START AUDIOTAPE)**Het ijs breken**

1. Hoe gaat het nu met u?
2. In hoeverre bent u in staat uw normale dagelijkse bezigheden te verrichten?
3. Hoe komt dat volgens u?

Laten wij beginnen over de ziekte van Lyme.

Ervaring & proces

4. Kunt u mij vertellen over uw ervaringen en het proces die u heeft doorlopen vanaf het moment dat u klachten kreeg?

(De afgelegde zorgtraject: persisterende klachten, vanaf wanneer last van de klachten, diagnoses, (huidige) behandeling, hoe gaat hij/zij nu om met de klachten; doorverwezen naar welke specialisten, buitenland (test & behandeling & heeft het gewerkt), Lyme test (hoe vaak, door wie, positief/negatief, welke test)

Doctor-patiënt interactie

5. Kunt u mij vertellen over uw ervaringen met uw huisarts vanaf het moment dat u klachten kreeg?

(o.a. antwoorden op: Vanaf wanneer naar de huisarts?; Hoe reageerde uw huisarts toen u aangaf dat u denkt dat u de ziekte van Lyme heeft? Wat vond u daarvan?;

Huisarts diagnose en behandeling)

6. Een officiële diagnose Lyme in de reguliere zorg in Nederland is niet gesteld. Hoe voelt u zich hierover? En over de huisarts?
7. Wat verwacht/ wat zou u graag willen van uw huisarts (houding van de arts, welke behandeling)?

Betekenis niet reguliere diagnose Lyme

Klopt het dat er geen officiële diagnose Lyme gesteld in de reguliere zorg in Nederland? Dus arts heeft niet gezegd: 'U heeft de ziekte van Lyme'? Maar u blijft erbij dat u de ziekte van Lyme heeft.

8. Vanaf wanneer dacht u dat u Lyme had?
9. Waarom denkt u dat u de ziekte van Lyme heeft?

10. Hoe reageert uw omgeving hierop? (werk, familie, vrienden, etc.) Wat doet dat met u?
11. Heeft u weleens overwogen dat het iets anders zou kunnen zijn?
12. Wat zijn de gevolgen van de niet reguliere diagnose Lyme op u dagelijks leven? (wat zijn de voordelen en nadelen)

Betekenis officiële diagnose Lyme

13. Bent u nog op zoek naar de officiële bevestiging van de ziekte van Lyme? Hoe uit zich dat?
14. Wat zou de officiële diagnose Lyme door de medische wereld voor u betekenen/ wat zouden de gevolgen kunnen zijn (wat zijn de voordelen en nadelen)? (Waarom bent u op zoek naar die bevestiging?)

Alternatieve diagnoses

15. Welke diagnoses heeft de huisarts gesteld en welke behandel traject heeft hij/zij aan u voorgesteld? Hoe reageerde u daarop en waarom?
16. Krijgt u weleens van anderen te horen dat het tussen de oren zit? Raad men een psychologische begeleiding aan? Wat vindt u hiervan?
17. Heeft uw huisarts weleens voorgesteld om naar een polikliniek te gaan waar mensen met onvoldoende verklaarde lichamelijke klachten worden behandeld? (Ja: Kunt u mij vertellen hoe dat ging en wat uw reactie was? Bent u wel/niet doorgedaan? Waarom wel/niet?; Nee: vraag 18)

Uitleg: Zo 'n polikliniek wordt ook wel een SOLK (Somatisch Onvoldoende verklaarde Lichamelijke Klachten) poli genoemd. De huisarts begint met een exploratie van de klachten om te kijken naar de ernst van de klachten. Er wordt gekeken naar de somatische, cognitieve, emotionele, gedragsmatige en sociaal dimensies. Het doel van de behandeling is vermindering van en/of meer greep krijgen op de klachten. Dit wordt gedaan vanuit een meersporen beleid. De klachten worden dan vanuit verschillende invalshoeken (lichamelijk, psychisch en sociaal) benaderd. Dit is maatwerk en wordt gedaan samen met de patiënt. Er worden een combinatie van verschillende therapieën aangeboden. Bijvoorbeeld: gesprekken met psychologen en/of psychiaters, gezinstherapie, groepstherapie, oefen- of fysiotherapie, cognitieve gedragstherapie. Medicatie kan soms wel een ondersteunende rol spelen bij het verminderen van verschijnselen zoals pijn,, spanningen, angst of bij stemmingsklachten. SOLK blijft een werkhypothese. Dat

	<p><i>betekent dat in de loop der tijd alsnog somatische pathologie aan de klachten ten grondslag kan blijken te liggen.</i></p> <p>18. Zou u daarvoor open staan? Waarom wel of niet? Onder welke voorwaarden zou u wel gaan?</p> <p>19. Hoe zou volgens u een SOLK behandeling gericht op chronische Lyme er idealiter vorm gegeven kunnen worden (behandeling, uren, reisafstanden, soorten therapie, groep/individueel)?</p> <p>20. Welke gevolgen zou deze behandeling hebben op uw dagelijks leven (wat zijn de voordelen en nadelen)?</p>
<p>Einde</p>	<p>Ik heb veel aan de informatie die u gegeven heeft en bedankt voor uw openhartigheid</p> <p>Zijn er nog aspecten die voor u en voor dit onderzoek belangrijk zijn, niet aan bod gekomen in dit gesprek?</p> <p>Heeft u er behoefte aan het letterlijk uitgeschreven interview na te lezen voor eventuele aanvullingen?</p> <p>U ontvangt via email nog mijn contact gegevens indien u nog vragen, opmerkingen heeft of interesse in de resultaten van het onderzoek.</p> <p>Heeft u nog vragen of opmerkingen?</p>

Appendix IV – List of reported complaints

1. A cold
2. Brain fog
3. Chills
4. Concentration loss
5. Confusion
6. Depression
7. Diarrhoea
8. Digestion problems
9. Dizziness
10. Eyesight
11. Fatigue
12. Fear/panic attacks
13. Forgetfulness
14. General pain
15. Headache
16. Hormonal problems
17. Lung problems
18. Muscle ache
19. Neuralgia
20. Nightmares
21. Obstipation
22. Overstrung
23. Painful bones
24. Burning eyes
25. Flu
26. Painful joints
27. Power loss arms and legs
28. Sensitive to light and sound
29. Sensations
30. Thyroid problems
31. Throwing up
32. Frequent urination
33. Hair loss
34. Hot flushes
35. Incontinence

36. Insomnia
37. Intestinal problems
38. Stiffness
39. Tinnitus
40. Personality changes
41. Jactation
42. Night sweat
43. Palpitations
44. Pneumonia
45. Stomach ache
46. Weight loss