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Diagnostic pathways of patients consulting at the infectious diseases ward for presumed Lyme disease : a qualitative descriptive study.

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Abstract

Objectives: Media coverage of Lyme disease (LD) has led to an increase in consultations for presumed LD in Europe. However, LD is confirmed in only 10-20% of patients, with a significant number remaining in a diagnostic dead-end. The objective of our study was to understand the genesis of the LD hypothesis in care pathways.

Methods: In spring 2019, we recruited for semi-structured interview the first 30 consecutive patients from a prospective cohort consulting in the infectious diseases department at University Hospital in Marseille for presumed LD. The inclusion criteria were: age ≥18 years, subjective symptoms for ≥ six months, no clinical or paraclinical argument suggesting current LD. The patients' medical trajectories were collected using a biographical approach by combining sequences from the interviewee's life and developing themes related to the medical history.

Results: The average duration of symptoms was 8.5 years. A majority of participants were convinced they had LD despite the lack of medical evidence and the scepticism of their referring GP. The diagnosis of Lyme disease was primarily triggered by identification with clinical stories circulating in the media. Most of participants had conducted the diagnostic investigation themselves.

Conclusions: Patient empowerment in the diagnostic process suggest a failure of modern medicine to propose solutions for medically unexplained symptoms. Clinicians should systematically explore patients' etiologic representations in a patient-centred care approach in order to create the conditions for a therapeutic alliance.

Key Words: Lyme borreliosis, post-Lyme disease syndrome, medically unexplained symptoms, social sciences, medical uncertainty.

Abbreviations:

LD: Lyme Disease

GP: General Practitioner

COREQ : COnsolidated Criteria for REporting Qualitative research

Introduction

Lyme disease (LD), which received little media coverage in France until the end of the 2000s, is now the subject of a controversy,[1] with sometimes virulent public debates. Doctors and patients represented by associations,[2] demand recognition of a chronic form of the disease associated with non-specific symptoms such as pain, asthenia, and concentration disorders.[3] In this context, long-term antibiotic treatments are often prescribed despite the absence of proven benefits and may cause serious adverse reactions and even death in some patients.[4–8]

To date, there is no evidence in humans pointing towards the diagnostic criteria of a possible chronic LD.[9] However, media coverage of this disease has led to an increase in consultations for presumed LD in France and in Europe.[10,11] In France, annual incidence is estimated at around 33,000 cases and presents strong regional disparities, with the incidence being very low around the Mediterranean area, where the vector is rare.[12]

Series of patients consulting with a suspicion of LD result in a confirmed diagnosis of LD for only 10 to 20%, while significant numbers of patients (6-26%) with non-specific symptoms (arthralgia, asthenia, myalgia, headaches) remain undiagnosed at the end of the etiological investigation.[13–15]

In a context of easier access to medical information, media coverage of many health issues, and official discourse promoting patient autonomy,[16] this paradigm has been begin to be reversed the last decades: physicians are now confronted with patients who produce diagnoses and seek to confirm them through the use of health professionals.[17]

Using a biographical approach, we sought to describe the diagnostic pathways of patients who initially consulted for a suspicion of LD and for whom this diagnosis had been rejected by an infectiologist at the time of their inclusion in the study. We wanted to better understand firstly

the influence of the social environment on the genesis of "LD hypothesis"; secondly the role of the patient in the diagnostic investigation.

Materials and methods

Design and Study Setting

This is a qualitative single-centre study. Between 1 May and 30 June 2019, we recruited the first 30 consecutive patients from a prospective cohort consulting in infectious diseases department for presumed LD as part of regional clinical research programme dedicated to tick-borne diseases led by University Hospital Institute (IHU) Méditerranée Infections in Marseille. This project was approved by an ethical committee. The reporting of this study follows the COREQ guidelines.

Patients were eligible to participate if they were 18 years or older, French-speaking, and able to provide informed consent. They had all previously been seen in consultation with a senior infectiogist. Inclusion criteria were as follows: presenting non-specific symptoms such as fatigue, difficulty concentrating, joint, muscle or headache pain for at least six months; having a negative Lyme serology; and no evidence for an organic differential diagnosis.

Interview guide and biographical approach

The interview guide (**Appendix 1**) was developed and iteratively revised during pretesting with five older adults (not included in the study). The semi-structured interviews followed a biographical approach, using an interview guide covering the history of symptoms and referrals to different medical specialties, detailed diagnostic pathway, including the history of the differential diagnoses, genesis of the LD diagnosis, the patient’s prioritisation of the most likely diagnostic hypotheses, and the associated diagnostic degree of certainty (low, medium, high). The interview guide also covered relationships with health professionals, in particular

general practitioners (GPs), their role in conducting the diagnostic enquiry, the impact of symptoms on daily life, and finally patients' views of the disease. The biographical approach consisted of collecting patients' trajectories by combining sequences from the interviewee's life and developing themes related to the subject of study.[18] It uses a life-calendar (**Appendix 2**), a retrospective data collection tool highlighting the chronological order and proximity of events, important transitions in health pathwat and makes it possible to jointly analyse several aspects of the patient's life.

Data collection and analysis

One male investigator (RL) trained in qualitative methods (GP with a master's degree in social sciences) conducted all interviews in person. The investigator had no direct clinical relationship with any participant. Interviews occurred in a private meeting room at the IHU after a scheduled follow-up consultation. Interviews were systematically audio recorded with the patients' agreement. We also used personal documents spontaneously provided by the patient (medical files, illness diary etc) and investigator' field notes. Interviews were fully transcribed, coded and analysed using the NVivo qualitative data software. All the collected data were systematically cross-checked. Two investigators (RL and CE, the clinician who performed the medical consultation) independently coded all transcripts. Differences were reconciled by consensus until 100% agreement was reached.

The clinical profile category was defined according to the patient's prioritisation of symptoms (see **Table 2**), in decreasing order of their impact on their quality of life. The category "diagnostic survey coordinator" was coded from the intersection of the following elements: the person who asked for serology test, the patient's deliberate search for a "pro-Lyme doctor" to confirm the diagnosis, spontaneous consultation of specialists (without referral by the GP), particularly infectious disease consultations, presence/absence of a referring GP (or

other referring physician) and finally spontaneous statements during the qualitative interview (e.g. “I conducted the investigation”). The category “pro-Lyme caregiver” was chosen if the patient reported during their pathway at least one consultation with a “specialist in chronic LD”, whether they were a doctor or other caregiver providing non-conventional medicines. The category of “Lyme activist” was chosen if the patient was a member of an association or an active member of a forum dedicated to LD.

Results

Participants’ sociodemographic and clinical characteristics:

The 30 participants were mainly women with an average age of 47.3 years, with a high education level. Interviews lasted from 44 minutes to 85 minutes. The “pain” profile was predominant. The history of the disease was long with an average symptom duration of 8.5 years (**Table 1 and 2**).

Genesis of the Lyme Disease diagnostic hypothesis

A majority of patients (22) did not report a history of tick bites. For these patients, the main types of potential exposure to the Lyme vector were the presence of ticks in their environment (8), contact with traditionally tick-carrying animals (7), and having spent time in a region perceived to be endemic like forests in the north-east of France (7).

The hypothesis of “chronic LD” in the diagnostic pathway/trajectory of patients was most often triggered by their identification with other patients’ clinical narratives circulating on different media and social networks (14). Other circumstances triggered diagnostic investigations including presence of false positive Lyme serology during a medical check-up (6), family or close friends raising the question of LD (5) and finally, the hypothesis evoked by their doctors (5).

All patients had previously undergone serological testing in a laboratory. For a majority of patients (16), the test was negative, for the others (14), the result was considered as a false positive by the clinician according to international and national guidelines.[19,20] Of the latter, half of them (7) had used laboratories whose techniques were not validated by international standards: private laboratories in Germany, “alternative” private French laboratories, or via a self-test kit obtained on the internet, all recommended by the websites of various Lyme associations. In addition, nearly half of participants (14) had received an antibiotic therapy for “chronic Lyme disease”, which was not justified on the basis of current recommendations.

Most of patients (24) stated that they believed the diagnosis of LD to be the main explanation for their symptoms, among which 13 considered this diagnosis to be highly certain (**Table 3**). Few patients had a “Lyme activist” profile (3), and had consulted a “pro-Lyme” caregiver (8). Serology was mainly prescribed at the request of patients (17), despite the fact that their referring doctors (23) were sceptical about the Lyme hypothesis. (**Table 3**).

Diagnostic pathway

During their diagnostic trajectory, patients had consulted 3.7 different specialists on average, half of them consulted a psychiatrist. 18 used complementary therapies (naturopathy, homeopathy, kinesiology, etc.) and 14 consulted a pain relief centre.

Regarding the history of differential diagnoses, 23 patients mentioned the diagnosis of fibromyalgia, among which 21 rejecting it because they considered it to be a “psychiatrisation” of their symptoms. In the end, we could conclude majority of participants coordinated the etiological investigation (23); for the others, the enquiry was conducted by their GP (4) or by their referring specialist (3) (**Table 4**).

Discussion

"When the patient makes the diagnosis"

A large majority of patients (with the participation of their entourage) were at the origin of the LD hypothesis. We found a significant role of narratives from other patients on social networks or in the media in the genesis of "diagnostic hypotheses". These stories are particularly valued by patients in situations marked by the absence of a satisfactory diagnostic proposal from doctors. Studies show the general public refers to this type of information source much more often, to the detriment of more "objective" and official sources. [16] This is in line with the results of a qualitative survey performed in Connecticut (USA), which reported that patients with LD placed greater trust in the experiences of close relatives who had contracted Lyme disease than in information disseminated by health professionals and health authorities.[21] Moreover, patients could find online tools to back up their hypotheses. A majority of patients managed to convince their GP to prescribe a Lyme serology test, illustrating that the medical decision is no longer monopolised by doctors. This reflects the contemporary role of patients claiming the legitimacy of a diagnosis based on their own experience.[22,23] The recognition of the patients' diagnostic proactivity has recently been the subject of studies in the field of social sciences of health.[17,24–26] Fainzang showed that diagnostic work was more particularly exercised by patients when physicians are unable to elucidate the causes of their disorders than when they are, with patients taking charge of the entire sequence from self-diagnosis to self-prescribing.[26]

"A diagnosis set in advance"

For the first time, our study explores the level of conviction associated with LD diagnosis. A large majority of patients did not report a tick bite, but often mentioned that they may have been bitten without noticing or remembering. A negative serology was not sufficient to

completely exclude the diagnosis of LD and, in the case of uncertain serologies, patients often gave more weight to the positivity of the ELISA test than to a negative Western Blot reference test. The “Lyme activist” profile, and/or an encounter with a “pro-Lyme” caregiver concerned a minority of the pathways described in this study and cannot by itself explain this high level of conviction observed among participants.

The attribution of symptoms to a well-identified external (environmental) cause is well described in the literature on LD and more generally on somatoform disorders.[27–30] The infectious origin is often guilt-reducing for patients who often refuse, as in our study, any psychological explanation of their symptoms, what they called “a psychiatrisation of their symptoms”. Moreover, the higher level of certainty about the LD hypothesis in patients leading their own diagnostic pathway than in the others suggests that patients had a pre-established etiological scenario and were seeking to put together the different elements of the medical puzzle to demonstrate this. In cognitive psychology, this phenomenon is known as “confirmation bias” and describes our tendency to seek, interpret, promote, and recall information that supports our previous personal beliefs.

Disappointment with science and scientific controversies

The current controversies over the chronic form of LD remind us of the strength of the population’s contemporary disenchantment with modern science, as highlighted by Ulrich Beck.[16,31] especially when modern science generates a multitude of highly specialised, fragmented, temporary, and often contradictory results, especially in the biomedical field. This is especially the case in the French context of the Lyme controversy. In 2018, French scientific societies and the National Academy of Medicine refused to approve the recommendations on LD published by the Haute Autorité de Santé, a French government agency.[32,33] Indeed, French scientific societies (including French College of General

Practitioners) did not recognise the new clinical entity called “symptom/polymorphic syndrome persisting after a possible tick bite” arguing that the term was not based on scientific evidence and opened the door to over-diagnosis and inappropriate antibiotic prescriptions.[33,34]

Strengths and limitations

The originality of this study lies in the population studied which consists of patients who have reached a diagnostic dead-end. In this frequent situation regarding LD, we showed that most of the patients were the driving force of the etiological investigations of their symptoms. Despite medical evidence, almost half of them were strongly convinced that they were suffering from LD.

To our knowledge, this study was also the first to apply a biographical approach to the analysis of the diagnostic trajectories of patients consulting in infectious disease wards for a suspected LD. This approach allowed for the joint analysis of contextualised self-reported data and clinical data from medical records. The interviews, by focusing on the overlap between life, medical and clinical events, highlighted the two dimensions at work in any care pathway: biology and biography.[35]

Our qualitative approach limits our ability to extrapolate but our sample size (n=30) was the largest to date among the international qualitative research published on the subject.[27,28,30,36,37]

Comparison with existing literature

A previous qualitative study involving 13 patients in the Savoy region of France reported the same results on the role of the internet and the media in the care pathways of these patients and in triggering their suspicion of chronic LD.[27] However, the method of recruitment

through a patient association led to an over-representation of patients who had activist attitudes, who were likely to support conspiracy theories, who were explicitly reported to be in conflict with the medical profession, and who had a relationship with “pro-Lyme caregivers”, than our study population. Other international qualitative studies on the subject focused on the experience and impact of the disease in the daily life of patients.[28,36,37]

Implications for practice

The patients in our study were the main actors in their diagnostic enquiry. The role of physicians appears paradoxical. Although patients frequently solicited them (number of referrals), physicians were powerless, or unwilling, to offer structured care pathways.
The clinician’s ability to listen to the patient’s disease history rarely includes consideration of the patient’s diagnostic experience.[17] Dissatisfaction with the medical diagnosis is the classic explanation for the use of alternative medicine.[38,39] By opposing the doctor as the sole custodian of the medical diagnosis,[40] to patients reduced to the subjectivity of their symptoms, run the risk of seeing the development of diagnostic dead-end or parallel diagnostic pathways. Finally, patient empowerment in the diagnostic process suggest a failure of mainstream medicine to propose solutions for symptoms that it cannot explain because of lack of evidence when the limits of medical knowledge are attained. In conclusion, clinicians should systematically explore the diagnostic work and etiologic representations of their patients in a patient-centred care approach in order to create the conditions for a therapeutic alliance.

Author Contributions:

Dr Lutaud had full access to all of the data in the study and takes responsibility for the integrity of the data and the accuracy of the data analysis.

Study concept and design: RL, PV, PW, CE.

Collection of data and data analyses: RL, CE.

Drafting of the manuscript: RL, CE.

Critical revision of the manuscript for important intellectual content: RL, PV, PW, CE.

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Table 1. Characteristics of the 30 patients

Characteristic	**No. (%)**
Age, mean (SD), y	47.3 (13.3)
Female sex	25 (83)
Living in a couple	23 (77)
Educational level	
<Secondary school education	7 (23)
Secondary school education	6 (20)
≥ Tertiary education	17 (57)
Professional situation	
Active employment	22 (73)
Unemployed	3 (10)
Retired	4 (13)
Disability	1 (3)
Currently on sick leave	17 (77)
Geographical origin[a]	
Provence-Alpes Côte d'Azur	28 (93)

[a] The 2 other participants were also from the Mediterranean region (Occitania).

Clinical profile[a]	**No. (%)**
Chronic pain[b]	15 (50)
Neurological symptoms[c]	8 (27)
Chronic fatigue syndrome[d]	7 (23)
Average duration of symptoms [min-max], y	8.5 [0.5-54]
Had an average duration of symptoms ≥ 5 years	15 (50)

Table 2. Clinical characteristics of the 30 patients

[a] The clinical profile was defined according to patient's prioritisation of symptoms, in decreasing order of their impact on their quality of life.

[b] The "chronic pain" category included neuropathic, musculo-articular, diffuse, poorly characterised or headache-type pain.

[c] The "neurological profile" category included patients whose main complaint was vertigo or sensitive motor disorders or cognitive complaints.

[d] The "chronic fatigue syndrome" category included patients with predominant fatigue, often associated with concentration difficulties.

1

Table 3. Genesis of the Lyme Disease diagnostic hypothesis for the 30 participants

Characteristic features	**No. (%)**
Reported tick bite	8 (27)
Nature of exposure from the patient's perspective (other than a tick bite)	22 (73)
Observed presence of ticks in their environment	6 (27)
History of unidentified insects bites	4 (18)
Contact with traditionally tick-carrying animals	5 (23)
Tick-bite episode in the entourage	2 (9)
Endemic region	5 (23)
Confirmed history of erythema migrans	3 (10)
Origin of the "chronic Lyme" hypothesis	
Identification with clinical narratives (TV, media, internet)	14 (47)
Physician	5 (16)
Entourage	5 (16)
Medical check-up	6 (20)
Lyme serology performed in private laboratories	30 (100)
Results of Lyme serology test	
negative	16 (53)
false-positive[a]	14 (47)
Serology performed in a non-approved laboratory	8 (27)
Internet diagnostic self-questionnaire	15 (50)

Received “anti-chronic Lyme disease” antibiotic treatment	14 (47)
Pro-Lyme caregiver intervention during their diagnostic pathway	8 (27)
Including medical doctors	6 (20)
Members of a pro-Lyme association (“Lyme disease activists”)	3 (1)
Have requested and obtained a doctor’s prescription for a Lyme disease serological test	17 (57)
Referring physician’s position on the Lyme hypothesis	
Pro-active	5 (17)
Neutral	14 (47)
Sceptical	9 (30)
Absent	2 (6)
Patient’s diagnostic hypotheses ranking	
Lyme disease hypothesis rank 1st	24 (80)
The degree of certainty associated with the diagnoses among patients ranking Lyme hypothesis first	
High degree of certainty	13 (54)
Moderate-low degree of certainty	11 (46)

[a] False-positive: Lyme serology was negative in ELISA and/or WesternBlot. The presence of IgM over a long period of time without serological evolution with the appearance of IgG was considered as a false positive and therefore concluded as negative.

Table 4. Characteristic features of the diagnostic pathways for the 30 Participants

Characteristic features	**No. (%)**
Average no. of specialties used in relation to the history of symptoms (excluding infectiology and psychiatry)	3.7 [0-9]
Referral to > 5 medical specialists in relation to the history of symptoms (excluding infectiology and psychiatry)	10 (33)
Referral to a psychiatrist in relation to the history of symptoms	15 (50)
Use of alternative medicine in relation to their symptoms	18 (60)
Patients treated in a pain-treatment centre	14 (47)
Fibromyalgia: diagnosis evoked by a doctor	23 (77)
Refusal of "psychiatrisation[a] of their symptoms"	21(70)
Main diagnostic pathway coordinator (typological approach)	
Primary care physician	4 (13)
Referring physician (other specialties)	3 (10)
Patient	23 (77)

[a] The term "psychiatrisation" was used when the patient explicitly mentioned the refusal to be told that the cause of their pain was psychological (e.g. "it's not in my head").