Article

Biographical Approach to Patients Consulting for Presumed Lyme Disease: a contribution to the understanding of patient pathways in symptom-based diseases.

Authors: Romain Lutaud*[1,2,3], Pierre Verger[3], Patrick Peretti-Watel[4], Carole Eldin[5].

Appendices/ Supplementary Materials

This supplementary material has been provided by the authors to give readers additional information about their work.

COREQ (COnsolidated criteria for REporting Qualitative research) Checklist

A checklist of items that should be included in reports of qualitative research. You must report the page number in your manuscript where you consider each of the items listed in this checklist. If you have not included this information, either revise your manuscript accordingly before submitting or note N/A.

Topic	Item No.	Guide Questions/Description	Reported on Page No.
Domain 1: Research team and reflexivity			
Personal characteristics			
Interviewer/facilitator	1	Which author/s conducted the interview or focus group?	8
Credentials	2	What were the researcher's credentials? E.g. PhD, MD	8
Occupation	3	What was their occupation at the time of the study?	8
Gender	4	Was the researcher male or female?	8
Experience and training	5	What experience or training did the researcher have?	8
Relationship with participants			
Relationship established	6	Was a relationship established prior to study commencement?	8
Participant knowledge of the interviewer	7	What did the participants know about the researcher? e.g. personal goals, reasons for doing the research	Supplements
Interviewer characteristics	8	What characteristics were reported about the inter viewer/facilitator? e.g. Bias, assumptions, reasons and interests in the research topic	N/A
Domain 2: Study design			
Theoretical framework			
Methodological orientation and Theory	9	What methodological orientation was stated to underpin the study? e.g. grounded theory, discourse analysis, ethnography, phenomenology, content analysis	7-8
Participant selection			
Sampling	10	How were participants selected? e.g. purposive, convenience, consecutive, snowball	7
Method of approach	11	How were participants approached? e.g. face-to-face, telephone, mail, email	7
Sample size	12	How many participants were in the study?	7
Non-participation	13	How many people refused to participate or dropped out? Reasons?	N/A
Setting			
Setting of data collection	14	Where was the data collected? e.g. home, clinic, workplace	8
Presence of non-participants	15	Was anyone else present besides the participants and researchers?	8
Description of sample	16	What are the important characteristics of the sample? e.g. demographic data, date	9
Data collection			
Interview guide	17	Were questions, prompts, guides provided by the authors? Was it pilot tested?	7
Repeat interviews	18	Were repeat inter views carried out? If yes, how many?	6
Audio/visual recording	19	Did the research use audio or visual recording to collect the data?	8
Field notes	20	Were field notes made during and/or after the inter view or focus group?	8
Duration	21	What was the duration of the inter views or focus group?	8
Data saturation	22	Was data saturation discussed?	N/A
Transcripts returned	23	Were transcripts returned to participants for comment and/or	N/A

Topic	Item No.	Guide Questions/Description	Reported on Page No.
		correction?	
Domain 3: analysis and findings			
Data analysis			
Number of data coders	24	How many data coders coded the data?	8
Description of the coding tree	25	Did authors provide a description of the coding tree?	8/9
Derivation of themes	26	Were themes identified in advance or derived from the data?	8
Software	27	What software, if applicable, was used to manage the data?	8
Participant checking	28	Did participants provide feedback on the findings?	N/A
Reporting			
Quotations presented	29	Were participant quotations presented to illustrate the themes/findings? Was each quotation identified? e.g. participant number	N/A
Data and findings consistent	30	Was there consistency between the data presented and the findings?	9-10-11
Clarity of major themes	31	Were major themes clearly presented in the findings?	9-10-11
Clarity of minor themes	32	Is there a description of diverse cases or discussion of minor themes?	9-10-11

Developed from: Tong A, Sainsbury P, Craig J. Consolidated criteria for reporting qualitative research (COREQ): a 32-item checklist for interviews and focus groups. *International Journal for Quality in Health Care*. 2007. Volume 19, Number 6: pp. 349 – 357

Once you have completed this checklist, please save a copy and upload it as part of your submission. DO NOT include this checklist as part of the main manuscript document. It must be uploaded as a separate file.

Figure A2. Interview Guide

We are interested in knowing your care history, particularly the history of diagnoses related to the symptoms that led you to consult at the IHU but also how you experienced this journey, and the role of the doctors who accompanied you. There is no right or wrong answer. Please do not feel like you have to answer in a certain way. The questions are also NOT specific about you, meaning all questions are hypothetical.

Do you have any questions before to start?

Clinical history and impact
1) What do you do in life? (career, family, education)
2) Symptom onset, clinical history / specific dates
3) if you had to prioritize symptoms in the way they most impact your daily life What would be the first? the following in the order?
What impact have these symptoms had on your professional life? your entourage? are you currently on sick leave?
4) Are there any particular life events that you would like to talk about that may have had an impact on your symptoms?
5) If you had to prioritize the probable diagnoses that best explain all of your symptoms, which would you place first? which ones would you place next?
6) For this diagnosis that you placed first: how confident/certainty do you have in your answer? low (I am not sure)/ medium (50-50%), high (I am convinced)
7) For what reasons do you think of this diagnosis mentioned first? (let speak freely ++)

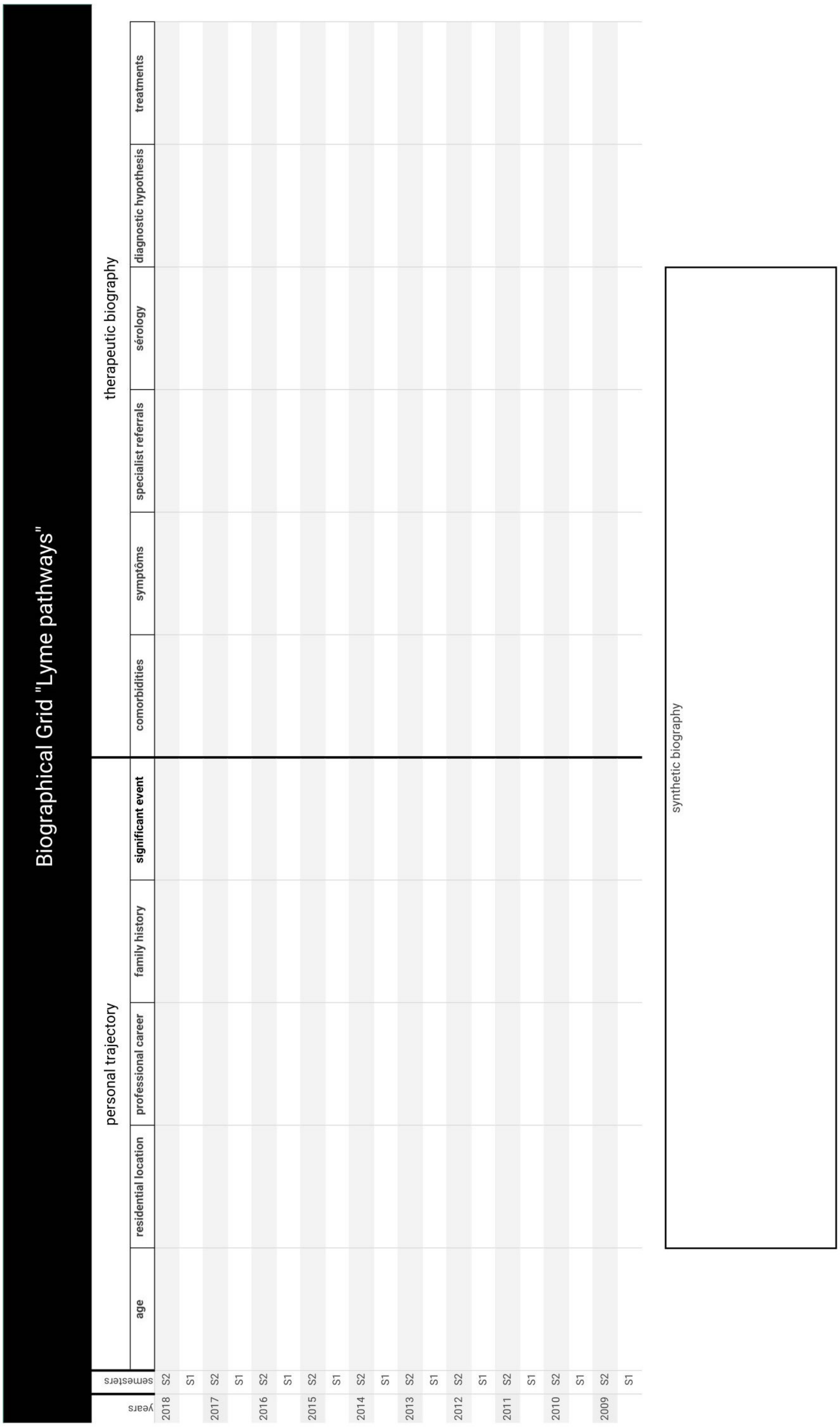
Genesis of the Lyme Hypothesis
1) Have you been exposed or even bitten by a tick? If so, can you tell us about the treatment/diagnosis that took place? Erythema Migrans?
2) When did you first hear about Lyme disease?
3) In what situations do you think you have been possibly exposed to the disease?
3) Under what circumstances has the hypothesis of Lyme disease been raised to explain your health problems? Who first brought it up or thought of it? (let the person speak freely if it comes up spontaneously)
4) Do you have an attending physician, or specialist doctor who regularly follows you for these symptoms? Have you discussed it with him/her? What did your doctor think about it? Has he or she encouraged you in this diagnostic process?
5) Have you used a Lyme diagnostic questionnaire on the internet? What was the result?
6) Did you do the serology? at the request of the doctor?
7) In which laboratory did you perform it? What was the result?
8) Have you received prolonged antibiotic therapy for chronic Lyme? Who prescribed it to you? Have you felt any improvement?

Care pathway
1) Let's go back over the history of the symptoms, can you give a precise account (chronology) of the doctors you have used in this context?
2) Who referred you to the IHU?
3) Did you consult a psychiatrist, for example? pain-centre?
4) Have you been hospitalized for these health problems?
5) Can we list all the diagnoses that have been mentioned by the doctors?
6) Did you have recourse to alternative medicine?
(to be explained)

7) Finally, have you met with professionals who are "specialists" in Lyme disease?
8) Do you regularly visit forums dedicated to Lyme disease? or are you a member of a patient association?
9) Generally speaking, have you felt that your doctors have listened enough to you about these health problems? How would you characterize the relationship with your GP?

In conclusion, is there anything else I haven’t asked you about this topic that you would like to share?

Figure A3. Life calendar (image)



Biographical Grid "Lyme pathways"

		personal trajectory					therapeutic biography					
years	semesters	age	residential location	professional career	family history	significant event	comorbidities	symptôms	specialist referrals	sérology	diagnostic hypothesis	treatments
2018	S2											
	S1											
2017	S2											
	S1											
2016	S2											
	S1											
2015	S2											
	S1											
2014	S2											
	S1											
2013	S2											
	S1											
2012	S2											
	S1											
2011	S2											
	S1											
2010	S2											
	S1											
2009	S2											
	S1											

synthetic biography

Table A1. Social characteristics

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

Table A2. Clinical characteristics of the 30 patients

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

[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.

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

Characteristic features	N=30
Reported tick bite	8
Nature of exposure from the patient's perspective (other than a tick bite)	22
Observed presence of ticks in their environment	6
History of unidentified insects bites	4
Contact with traditionally tick-carrying animals	5
Tick-bite episode in the entourage	2
Endemic region	5
Confirmed history of erythema migrans	3
Origin of the "chronic Lyme" hypothesis	
Identification with clinical narratives (TV, media, internet)	14
Physician	5
Entourage	5
Medical check-up	6
Lyme serology performed in private laboratories	30
Results of Lyme serology test	
negative	16
false-positive[a]	14
Serology performed in a non-approved laboratory	8
Internet diagnostic self-questionnaire	15
Received "anti-chronic Lyme disease" antibiotic treatment	14
Pro-Lyme caregiver intervention during their diagnostic pathway	8
Including medical doctors	6

Members of a pro-Lyme association ("Lyme disease activists")	3
Have requested and obtained a doctor's prescription for a Lyme disease serological test	17
Referring physician's position on the Lyme hypothesis	
Pro-active	5
Neutral	14
Sceptical	9
Absent	2
Patient's diagnostic hypotheses ranking	
Lyme disease hypothesis rank 1st	24
The degree of certainty associated with the diagnoses among patients ranking Lyme hypothesis first	
High degree of certainty	13
Moderate-low degree of certainty	11

[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
Referral to > 5 medical specialists in relation to the history of symptoms (excluding infectiology and psychiatry)	10
Referral to a psychiatrist in relation to the history of symptoms	15
Use of alternative medicine in relation to their symptoms	18
Patients treated in a pain-treatment centre	14
Fibromyalgia: diagnosis evoked by a doctor	23
Refusal of "psychiatrisation of their symptoms"	21
Main diagnostic pathway coordinator (typological approach)	
Primary care physician	4
Referring physician (other specialties)	3
Patient	23