

## Definitely maybe: new governance of uncertainty and risk in patient group involvement with UK guidance on testing for Lyme disease

Article (Accepted Version)

Faulkner, Alex, Bloor, Kate and Hale, Vahsti (2021) Definitely maybe: new governance of uncertainty and risk in patient group involvement with UK guidance on testing for Lyme disease. *Science, Technology and Society*, 26 (1). pp. 116-134. ISSN 0971-7218

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Title & contacts

**Definitely maybe: new governance of uncertainty and risk in patient group involvement with UK  
guidance on testing for Lyme disease**

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**Abstract**

States that claim responsibility for citizens' healthcare try to deal with knowledge uncertainties while preserving a duty of care. Production of clinical guidelines in disputed medical conditions or where uncertainty is high, is difficult. Patient groups may advocate non-credentialed evidence, contribute to debates, and form alliances with established policy actors. In this context, Lyme disease, especially highly contested 'chronic' Lyme disease is a good case with which to examine how official governance institutions are managing diagnostic uncertainty and evidence for tests. The healthcare state has been provoked to develop extensive policy for Lyme disease. In the UK, national Health Technology Assessment agency, NICE, began a consultation process in 2016. NICE and other policy actors are moving toward more participatory modes of decision-making. The paper analyses NICE's recently published guidelines and consultation documents; patient groups' contributions; observations of consultations and of evidence review processes; and recent Department of Health systematic reviews including patient group participation. We draw on concepts of participatory governance, patient group activism and guideline involvement. We find an increased level of participation by patient groups in recent policy and evidence review processes, and hence legitimization of them as 'stakeholders', alongside a strengthened state position on pre-existing diagnostic and testing standards.

**Keywords:** Lyme disease diagnosis & testing; participatory governance; stakeholder engagement; patient organisations

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**Background**

The tick-borne bacterial infection of Lyme disease was identified in 1983. It has become increasingly common in the UK. Cases, defined by standard NHS laboratory tests, have increased 3.6-fold since 2001 and the true incidence is suggested to be close to 3000 per annum (Dubrey et al., 2014), although may be even higher due to omission of privately tested patients and clinical diagnosis figures. Many other countries experience similar or higher trends. Internationally, issues around Lyme disease diagnostic testing, processes and technology, disease identification, and their evidence bases are contentious (Tonks, 2007; Borgermans et al., 2015). Symptoms might include rashes (including the typical 'bull's-eye' rash) and flu-like symptoms including headaches, high temperature, and aching joints. Later symptoms, though contested medically, might include joint swelling, nervous system problems and heart inflammation. There are significant medically confusing parallels with disputed conditions such as fibromyalgia and chronic fatigue syndrome. The acute form is treated with antibiotics. A 'chronic' form of the disease is especially disputed, and patients often seek private tests, having not obtained a desired diagnosis through public healthcare (NHS). Since the late 1990s a 'two-tier' testing regime, involving two stages of laboratory antibody tests has been officially endorsed. Medical indications for testing (or not testing), interpretative procedures, and rules for disease identification affect access to and the nature of clinical advice and treatment available to patients.

Patient groups have proliferated in the UK. In 2015, a prominent businessman founded the Caudwell LymeCo charity, combining with other celebrities and patient groups. Whilst undertaking different roles and activities, these groups and charities challenge the existing official and professional guidance in the UK. At the same time, a range of new official policy initiatives has appeared, notably the development of clinical guidelines by the National Institute for Health and Care Excellence (NICE, 2018) and UK government Department of Health commissioned 'systematic reviews', one of which focuses on the

‘stakeholder experiences’ of Lyme disease diagnosis and testing (EPPI Centre, 2017). In the current culture of public policy development, statutory agencies have to deal with knowledge uncertainties while finding acceptable methods for involving interested parties.

Current UK (England) statutory sector policy has been led by Public Health England (PHE) from 2012 (formerly Health Protection Agency), and its reference laboratory for testing - the Rare and Imported Pathogens Laboratory (RIPL) (PHE, 2014). A discussion was held at the House of Lords in October 2015, and the UK Department of Health began its reviews in 2016. The British Infection Association (BIA), the primary professional body for infectious disease, had published a position statement somewhat earlier (BIA, 2011). However, medical opinion is not uniform. A recent internationally-authored editorial in the *British Medical Journal* argues for a broader debate including patient groups amongst the ‘stakeholders’ (Borgerman et al, 2015). Introduction of the ‘engagement’ language of ‘stakeholders’ in a high profile general medical journal may signal an emerging re-orientation of the modes and scope of state governance in Lyme disease.

Against this background, this paper explores how patient groups have been able to participate in and contribute to new guidelines and policy development on Lyme disease testing through the two examples of the NICE guidelines process and the Department of Health based systematic review (EPPI Centre, 2017). We examine how this interaction happened, profiles of its participants, and the power dynamics between actors - and how evidence is constituted and deployed in the process. Using this approach, we contribute to debates about patient group participation in health policy, and governance related to clinical guidelines development, showing the central importance of medical testing. We analyse these cases in detail to consider whether, and in what ways, these may be understood as moving in the direction of participatory governance, and we comment critically on the effectiveness, and otherwise, of stakeholders’ participation and how it might be developed. This is of general significance to understanding the role of patient and other stakeholders in contemporary health governance.

The structure of the paper is as follows. We introduce relevant concepts from recent literature on participatory governance in general, and then patient organisations and activism and their forms of knowledge, and patient participation in guideline development. Having described our methodology, we then present our analysis of the process and content of the NICE and EPPI Centre reviews. We conclude by discussing this analysis in terms of its significance for forms of governance relationships in this contested public health area, locating it in the literature, and summarising our interpretation and suggestions for improvement to such processes.

### **Theory of public policy governance and its emergence in health policy**

The last three decades have seen a broad societal reconfiguration and re-legitimation of public governance of science, well-described as developing ‘social robustness’ through extended societal participation (Nowotny, Scott & Gibbons, 2001). Key development of the concept of ‘participatory’ governance has been produced by Frank Fischer (2006, 2009), who highlights that ‘the most important actors in this new world of governance have been social movements and nongovernmental organizations’ (Fischer, 2006, p. 19). He notes the rhetorical nature of much of this trend: ‘In the world of politics it is more or less de rigour (*sic*) for policymakers to at minimum pay lip service to the importance of participation... some writers have now begun to describe... a new bureaucratic instrument for political-administrative manipulation.’ (Fischer, 2009: Ch.3, p. 2)

These developments have become evident in the medicine/healthcare sector. ‘Stakeholder’ governance involves an increasing participation of patients and patient organisations in healthcare policy and biomedical science, which has been advocated for public health research since the 1990’s (Popay & Williams, 1996). The World Health Organization has noted: ‘... policy-making through highly networked, multilevel, multi-stakeholder governance is not recent...’ (WHO, 2012, p. 27), and it is notable that the major health technology assessment international (HTAi) network has its ‘HTAi Interest Group for Patient and Citizen Involvement in HTA’, which ‘brings together patients and citizens, patient advocates, patient engagement consultants, clinicians, researchers... HTA agencies, government, industry and others...’ (HTAi, 2019). We also note that NICE has its own ‘Citizens’ Council’.

Patient participation in governance can obviously vary in breadth, depth, stage and volume of involvement. As de Freitas has written: 'Public and patient participation can take multiple forms and enable different degrees of power sharing .... At one end of the continuum are one-off participatory spaces, such as listening exercises or health consultation events, in which citizens are asked to contribute their views without further commitment to follow-up action. At the other end of the continuum are more durable participatory spaces, including health councils and national health bodies, in which citizens can engage in shared leadership' (de Freitas, 2017, p. 31). This framework is useful for positioning the Lyme disease governance case.

### **Conceptualising patient organisations, activism and guideline development**

Alongside the evolution of governance forms has been an increase in organised patient activism. As Epstein has noted:

'the sheer quantitative increase in the formation of patient groups and health movements... as well as their enhanced social visibility. This upsurge...reflects... more skeptical attitudes toward doctors, scientists, and other experts, trends also manifested in new conceptions of patients' rights.' (Epstein, 2011, p. 260)

In the context of these developments in governance, activism and participation, there is also a growing specific literature about patient involvement in Health Technology Assessment (HTA), which we also draw on to situate the case of Lyme disease testing.

In sociological studies, echoing de Freitas, it has been suggested that 'policy makers should be attentive to the well-demonstrated possibility that the inclusion of organized patient voices can improve health care by making it both more effective and more equitable, but they should not assume that there is any single ideal model' (Epstein, 2011, p. 271). It has been pointed out that the level of theoretical understanding regarding different forms of knowledge and expertise brought by patient organisations to HTA processes, is generally low (Moreira, 2015). In principle, inclusion of 'patient experience' could include the incorporation of key social or cultural factors into policy, which might impact on the effectiveness of implementation (e.g. compliance, application) (Moreira, 2015).

A specific literature comes from ‘health services research’ (HSR) perspectives and focuses on the desirability or effectiveness of patient involvement (e.g. Culyer, 2005; Facey & Hansen, 2011; Health Equality Europe, 2018), and specifically in guideline development. Patient involvement is regarded as beneficial, but the HSR evidence for more appropriate guideline outcomes is less positive. One of the stronger studies found no empirical evidence to support a beneficial effect (van de Bovencamp & Trapenburg, 2008). Patient perspectives may be seen as lacking objectivity and patients may have difficulties with technical language and scientific evidence (van Wersch & Eccles, 2001). However, Culyer (2005) points out in relation to NICE, that ‘client involvement in decision-making can work well, but demands commitment from the entire organization, specific managerial arrangements and funding. De Freitas (2017), again, states that lay citizens, alongside other stakeholders should be entitled to specific training in methods and processes.

## **Methodology**

The paper draws on a wide variety of documents in the public domain, and on the broad experience and specific fieldnotes made by author KB who leads the voluntary ‘*Lyme Research UK*’ organisation and who participated in some of the stakeholder engagement processes considered here, as both activist and critical observer. Author KB participated in workshops at both NICE and EPPI Centre (University College London), coordinated responses and feedback between several patient groups at different stages of the two processes, and compiled written notes during or reflecting on the processes, on which we draw as fieldnote sources. The key documents analysed are those published by NICE in the interest of transparency around their guideline development process, including responses from stakeholders to drafts of the ‘scope’ of its review and to the draft guideline itself and ‘evidence reviews’ informing them, and one linked document reporting on the ‘stakeholder experience’ of clinicians, researchers and patients, commissioned by the UK Department of Health (EPPI Centre, 2017). Further documents related to UK policy and practice, manuals on the NICE guideline process, website information – collected from both stakeholders and patient groups, statutory policy documents e.g. Public Health England, and its

Rare and Imported Pathogens Laboratory (RIPL). The small number of key documents analysed amounted to over 500 pages of text. We developed a timeline of events, in the NICE and Department of Health process in order to organise the analysis systematically. This included listing and categorising the registered stakeholders in the NICE process and examining their committee composition in order to interpret the interests, organisational positions and expertise represented. We also identified some stakeholders, that potentially could, but did not participate in the process.

We adopted a conventional approach to systematic content analysis of the documents. The documents related to each stage of the NICE and EPPI consultations were scrutinised systematically, extracting information on participant composition, topics of testing, and focusing on patient group interventions and NICE and EPPI responses. We chose the two most active patient stakeholder groups (Lyme Disease Action (LDA) and Lyme Disease UK (LDUK) to focus on primarily and noted information from other groups. We identified how various types of evidence had been employed by stakeholders in the consultation processes, and what the response – positive or negative – of NICE was. Two of the team scanned in full the main documents to identify a range of examples where patient groups had made critical or reflective comments.

We also note that our collaborative approach to research and authorship is an example of research-based activism, illustrating the alliance-building noted as a feature of certain patient organisation strategies (Rabeharisoa, 2003), and enacting a form of a 'joint epistemic and policy enterprise' (Rabeharisoa et al., 2014, p. 121). The collaborative team combines a concern with both diagnostic uncertainty as a personal and professional (medical sociology) issue and Lyme disease diagnosis and testing specifically as activist issues. Equally, while adhering to academic standards of data collection and analysis, we do so with a shared goal of contributing critically to policy and practice improvement.

## **Analysis**

In this section we present our analysis of the NICE guideline development process and the Department of Health review, focusing on the ways in which governance has been conceived and mobilised, and how

patient groups have been positioned and participated.

## **1. NICE guideline development**

### ***Participation in the NICE consultation***

NICE is the national health technology assessment agency of England and Wales. It employs powerful assessment methods of review of published studies, and elite sets of medical and other experts to guide NHS practice and adoption decisions about new drugs and devices. Its ‘technology appraisals’ are mandatory on the NHS, and its guidelines, although advisory, have levels of acknowledged authority that are difficult for practitioners to ignore. The NICE process on Lyme disease has resulted in publication of a full clinical guideline, a ‘Clinical Knowledge Summary’ (CKS) (NICE, 2015, updated 2018), and various other documents.

NICE’s technology assessments and guideline work begin with defining the ‘scope’ of the task. This includes parameters such as the disease definition, the affected population, risk factors, symptomatology, the current standard of care, and the type of evidence to be sourced and assessed. The scope is especially important because it defines the ‘rules of engagement’ which may be the subject of basic disagreement amongst some stakeholders, especially in the case of contested medical conditions, where diagnosis and testing are especially crucial (Jutel & Nettleton, 2011). Overall leadership of the NICE process was vested in a 15-member committee consisting of two general practitioners, four ‘lay’ members, medical specialists including pharmacy, general physician, microbiology, paediatric/neurology, rheumatology, infectious disease, chaired by a Professor of Paediatric Immunology & Infectious Diseases. Of the ‘lay members’, these included a bioethicist, the long-time chair of Lyme Disease Action (one of the largest patient groups, organising clinical/scientific conferences amongst other activities) who had both a scientific/health information background, a ‘retired marketer’ suffering from Lyme disease, and the CEO of Caudwell LymeCo.

In the scoping work, the majority of stakeholders registered for the NICE consultation were NHS and medical-related. These included commissioning/service delivery bodies and NHS management groups, and professional medical associations. Of 135 registered stakeholders, 30 percent were professional groups, including two representing alternative or complementary medicine. The second largest group was NHS service organisations including health and social care trusts and hospitals (21 percent). Fifteen patient related organisations were registered. There were also four government bodies and six guidelines development related bodies or networks, research centres such as the Health Protection Research Unit in Emerging and Zoonotic Infections, and the MRC Centre for Neuromuscular Diseases. One was a deer management group (deer may carry infected ticks).

The NICE guideline development process for Lyme disease can be divided into two main stages, preceded by a workshop for stakeholders. This workshop, in February 2016 in London, was held for stakeholders to discuss the format and content of the initial 'scoping' document, understand the NICE development process, and get initial feedback from participants. It took the form of deliberative exchange between various representatives and NICE staff. Information about the initiative was passed around via social media and hence some relatively small patient groups became involved. The workshop attendees comprised representatives of four 'Lyme groups' (Lyme Disease UK, Caudwell LymeCo, Lyme Disease Action, Lyme Research UK (present author KB)) plus one related charity (Facial Palsy UK). Some attendees came from professional colleges such as the Royal College of General Practitioners, Royal College of Paediatrics and Child Health, Association of British Urologists, the British Society for Antimicrobial Chemotherapy, and the British Infection Association. This might be seen as a small number considering the broad range of professional groups with potential concern in Lyme disease. There was a noticeable absence of groups directly connected with testing issues such as microbiology and pathology. There was one company, Oxford Immunotec (supplier of the 'stage one' ELISA test kit), representatives from Public health England, the Department of Health, and NHS Highland.

The workshop was steered by NICE staff. The complex discussion covered many of issues that are longstanding themes amongst patient organisations. Concerns raised included that existing testing regimes should cover a broader range of strains and subspecies of the *Borrelia* bacteria, other currently non-endorsed tests should be included in the scope of NICE's review, and limitations of tests for special groups such as immunocompromised patients. A frequently cited concern was an alleged overreliance on testing in clinical diagnosis. Notably, one group proposed an additional question for the scope, which was eventually accepted: 'What is the best management strategy for patients with non-classic symptoms with positive/negative tests?' (fieldnotes author KB; NICE, 2016a)

### ***The NICE 'scoping' consultation***

Following this, consultation on the draft text of the scoping document was undertaken by online correspondence, the results of which were published in a 'Stakeholder comments table' (NICE, 2016b).. The most responses to this draft 'scope' came from patient organisations. Surprisingly, only one NHS organisation responded, NHS Highland, notably a very rural area of Scotland where tick-bites are a significant concern. The responses (approximately 167 separate statements from 13 different organisations (6 of which responded 'no comment'), varied from the apparently non-engaged (e.g. British Association of Dermatologists had no comments) to the very lengthy and critical (e.g. patient organisation VIRAS – Vector-borne Infection Research Analysis Strategy). Some 'single' comments were wide-ranging, citing academic articles and ranging over several pages.

As noted above, and for reasons of space, we have made a selection of stakeholders' comments and NICE responses to illustrate the range of organisations, comments, types of knowledge and evidence being utilised, and NICE responses, to assess the characteristics of stakeholders' participation in the guideline development process.

Many different issues in the draft scoping document were raised around testing by patient groups, including themes such as those raised during the workshop. Further specific points included appropriate

timing for testing and methods of interpreting results for different clinical scenarios, manufacturers' instructions for testing, and other related tests (e.g. thyroid). Turning to specific examples, a particularly controversial issue is whether person-to-person contagion, for example via blood transfusion, congenital or sexual transmission is possible, given the medical view that Lyme disease is 'tick-borne'. This was raised by several patient groups (LDA, Caudwell LymeCo, LDUK and LRUK). They also wished that other non-tick vectors and symptomatology should be included in the scope. For example, patient group Caudwell LymeCo commented: 'Remove "Transmission of the disease between people" from the section "Areas that will not be covered"'. NICE responded, in this case positively (all NICE's responses are preceded by 'Thank you for your comment'):

Thank you for your comment. We have discussed your comment in detail and reviewed the decision to exclude other ways of transmission. Person-to-person transmission is now included as a key question in the scope. (NICE, 2016b, p. 35)

It is notable that NHS Highland made a number of comments reflecting an overall position very similar to some of the key patient organisations, including requesting that a greater range of types of tests should be evaluated - knowing that these may 'lack standardisation, sensitivity and specificity' (NICE, 2016b, p. 113). NICE responded that this would be considered by the Guideline Committee in defining the evidence to be reviewed.

There were also a number of comments from patient groups about recognising potential Lyme disease cases, for example in multiple or non-classic presentations of rashes. One patient group was concerned that circular EM rashes were not always typical (Lyme Research UK, 2016, p. 84). NICE replied:

We have changed the wording in the scope to read: "...in *some* people this is followed by....." to reflect the uncertainty about the true proportion of people (NICE, 2016b, p. 18; our emphasis).

Likewise, some patient organisations asserted that certain subgroups of patients should receive specific consideration for diagnostic testing. LDA stated: 'We feel that individual consideration should be given to immunocompromised people and pregnant women in whom diagnosis may be more difficult...' (NICE, 2016b, p28), evoking the following tentatively positive response:

The guideline committee will review the evidence about diagnostic test accuracy... in pregnant women and immunocompromised people. It is anticipated that these populations will form sub-groups in each of our evidence reviews (NICE, 2016b, pp. 171-2).

The question of persistent symptoms is one of the most controversial. Lyme Disease UK commented:

When there is currently no test available to distinguish past infection from ongoing infection or new infection, the evidence and tests that the term “relapse” is based on, should be reviewed... anecdotal evidence exists ...that patients who still have a positive test following “standard” treatment ... are told it is likely to be a false positive (NICE, 2016b, p. 76).

NICE responded: ‘Testing will be addressed by an evidence review ...The review question and protocol will be developed by the guideline committee’ (NICE, 2016b, p. 76). It is notable that in the workshop and scoping phase overall, NICE collected information on 34 test kits in its review. However, most non-standard non-mandated tests were later discounted on grounds of weaknesses in evidence, and the full list of tests scrutinised was not placed in the public domain.

Thus, in the patient organisations’ comments and NICE’s responses we can see a variety of types of evidence being put forward including both scientific and ‘anecdotal’ experiential evidence. Our examples show that NICE has responded with several adjustments to the ‘scope’ of the review, and at this stage ‘parked’ the question of the range of possible tests to the authority to be provided by its ‘evidence review’.

### ***The NICE draft guideline consultation***

Prestigious, elite, national-level organisations orchestrate and carry out the guidelines process. The second stage comprised the drafting and discussion of the guideline text itself and associated evidence reviews. Several groups work together; a committee that makes the recommendations; staff who undertake quality checking, based at NICE; an evidence review team; the developer, and a technical team that supports the committee. The protocol is periodically updated (NICE, 2014).

The Lyme guideline development process was undertaken by the National Guideline Centre, commissioned by NICE but based in the Royal College of Physicians. It is said (on its own website) to be one of the largest guidelines development centres in the world. Registered stakeholders were encouraged to respond to the draft document within a short, 6-week period. All the comments from participants were considered by the committee, supported by the technical technology assessment staff, and published online (NICE, 2018). 635 responses came from six patient organisations (one non-UK) and 17 other organisations. Thirteen organisations provided substantive responses, including one private medical provider, a US-based professional association, the deer management group and NHS Highland. The remainder were medical professional groups. It is notable again that various professional organisations/societies in microbiology and infectious diseases did not take part. The number of active stakeholders can thus be regarded as relatively small.

How 'evidence' is construed is key to the NICE process. One of the key reports was 'Evidence reviews diagnostic tests', presented in a 300-page document (NICE, 2017a). Most of the report consists of tables summarising published clinical studies, critically appraised and reviewed, showing reasons for including or excluding them as evidence acceptable to NICE. About 120 clinical studies of testing were included. In a section of this review entitled 'Why the committee made the recommendations', it is stated:

There is uncertainty over which test or combination of tests are most helpful.... The committee agreed that initial testing with a combination IgM and IgG ELISA [antibody tests] for Lyme disease should be offered because the evidence generally showed better accuracy... for combined tests (NICE, 2017a, p. 187).

Although this reads like a new finding ('the committee agreed'), it in essence re-confirms the pre-existing standard of care. As an example of officially endorsed technical discourse, we can see that the authoritative presence of 'the committee' is dominant and the use of the passive voice – 'there is uncertainty' – asserts the scientific basis of this authority. The Centre also tried to include health-economic studies but could not include any studies of sufficient quality. They nevertheless conducted an independent cost modelling of the two tests involved in the standard 'two-tier' testing regime,

concluding that ‘2-tier testing is very likely to be at least cost neutral compared to initial testing only...’ (NICE, 2017a, p. 184). This conclusion, based on what can be called a quantification of uncertainty, thus provided stronger support than previously for the use of the follow-up second-stage test. NICE also supported future research on the ‘seroprevalence of antibodies to tick-borne infections in the UK and to clarify further the most effective tests at different stages’ (NICE, 2018; Recommendations). This policy position may be construed as offering hope to patients (of future recognition of a chronic form of the disease), a conspicuous feature of contemporary biopolitics (Novas, 2006).

Patient groups made substantial and voluminous comments in the process, mobilising not only experiential knowledge but also scientific and survey-based insights. Their contributions ranged from questioning the foundation of the standard testing regime, through absence of manufacturers’ data, to the validation status of non-UK laboratories. *Fight Lyme Now*, for example, describing themselves in their response as ‘a science-based lobby group’, make a lengthy submission that questioned the basis on which the standard diagnostic tests were developed:

...one has to question the rationale of establishing the cut-off (or the point at which the assay is considered negative), using samples from individuals with very well characterised Lyme disease and applying the this (*sic*) test to other groups of individuals (NICE, 2016c, p. 9).

NICE responded, supporting the desire to ensure ‘tests used in UK are appropriate for the UK population’, and they ‘emphasised’ this in their research recommendations. Validation of tests was a key point of contention, and *Lyme Disease UK* sought more detailed information on this:

It is critical that patients are given advice on how to assess whether a test is validated rather than simply given a blanket statement about all non-NHS labs (NICE, 2016c, p. 145).

NICE replied, on a concessionary note:

It is not the intention to suggest all private and foreign laboratories are using tests that have not been validated (*ibid*, p. 145).

False negative tests are also contentious. *Lyme Disease Action* commented:

There is no test of disease activity... This is why it is unsafe to restrict...the guideline to seropositive patients only. There is an extensive literature on seronegative Lyme disease in humans, confirmed by culture or PCR [polymerase chain reaction] even after antibiotic treatment (NICE, 2016c, p. 49).

NICE's response pointed out, however, that in this case their evidence review and the associated recommendations were not restricted to seropositive cases (NICE, 2016, p. 49).

*Lyme Disease UK (LDUK)* wanted evidence from manufacturers of tests to be included: 'Why is the view of test manufacturers ignored?' NICE responded that they were not aware of relevant evidence provided by manufacturers, the only study being 'a series of case studies published in 1989' (NICE, 2016:76), showing the methodological restrictions imposed by the NICE review protocols. *LDUK* also made a broad-ranging critique of the draft guideline, pointing to experiential knowledge as well as 'lay epidemiology' ('symptoms which we see as common'):

We have a daily overview of the intense suffering as well as the pervasive lack of knowledge about Lyme disease amongst GPs and NHS specialists. Our members found much of the guideline misleading and ambiguous. Doctors...are likely to come away with the message that Lyme disease is rare, easy to treat and that it cannot persist beyond two short courses of antibiotics... (NICE, 2016c, pp. 82-3)

NICE responded robustly to this strong critical statement, again pointing to 'research recommendations' as an avenue of hope for future possible evidence-based improvements:

The committee were aware of the limitations of the evidence base and have drafted research recommendations... confident that this guideline provides a useful tool for healthcare professionals (NICE, 2016c, p. 82-3).

Again, as with the scope consultation process, we see a range of adaptations accepted by NICE, robust defence of the standard NHS testing position, appeals to the NICE-defined evidence base, the strategic

offer of hope through research, and a range of different types of knowledge claims deployed by patient groups.

### ***The 'information needs of patients'***

A further strand of NICE's/NGC's evidence review was focused on the 'information needs' of patients (NICE, 2017b). We can understand from this framing, that patients here are configured as passive actors properly requiring education and information as healthcare recipients, rather than as active 'biocitizens' (Jorgensen, 2015).

Interestingly, this evidence review included only research with 'qualitative study designs' (NICE, 2017b, p. 6). However, only two such studies were found relevant after screening some 16,000 articles. Of the two, although the review's 'evidence tables' do not make it clear, one was conducted in the USA (in year 2000) and the other in Canada (2015), so the direct relevance for UK patients and healthcare system can be questioned. Nevertheless, it is of interest to summarise the main conclusions that the National Guidelines Centre drew, since this is important to patients' representation as stakeholders:

diagnostic tests correctly identify most people with Lyme disease but can produce false-negative results.... People with Lyme disease may feel anxious that they will not be helped or about the possibility of receiving alternative diagnoses. It was therefore decided that people with Lyme disease should be informed about how the tests work, factors that may reduce their accuracy and the importance of using validated tests. People with Lyme disease should also be reassured that they would continue to be ...reviewed (NICE, 2017b:14).

Although the reassurance of clinical review is promised here, guidance may remain unclear about how uncertainties about test accuracy could be shared with patients. It is also notable that the literature search was conducted only in medically-oriented databases, potentially marginalising the experiences of Lyme disease sufferers as well as sympathetic researchers such as medical sociologists and anthropologists. In spite of these apparent paternalist framings, however, our observation was that

patients saw information needs as important, while wanting information to reflect their specific concerns.

Overall, we see in the NICE draft guideline process a similar mixture of patient organisation interventions and NICE responses. Although the patient organisation contributions differ, we see a combination of forms of knowledge and 'experience' being drawn upon. Likewise, NICE responded with some adjustments, for example to allow stronger expression of scientific uncertainties in the 'evidence' and advice on clinical sharing of information with patients, but actually reinforcing fundamental, established positions on the standard testing regimes.

## **2. The 'stakeholder experience' review on diagnostics and testing**

The Department of Health commissioned a set of 'reviews' on Lyme disease at the same time as the NICE process, one of which focused on 'stakeholder experiences' of testing and diagnosis (EPPI Centre, 2017), stakeholders being defined as clinicians, 'researchers', and patients. The aim was 'to help interpret the findings of the reviews undertaken by NICE... to aid the use of these tests in practice' (EPPI Centre, 2017, p. 38). The review addressed 'patients', clinicians' and researchers' perspectives and experiences of diagnosis of Lyme disease' and a need for guidance about how to 'implement findings about the accuracy of different diagnostic approaches' (*ibid*, p. 5). A consultation process comprised two stages. The primary focus was on literature review using 'systematic review' methods, similar to those of NICE, with the difference that qualitative social science and related studies were to be included. Patient groups, which had to have a 'national focus', were then invited to provide feedback. Face-to-face consultations with eight UK-based patient advocacy groups were undertaken 'to understand how our draft emerging findings from international studies of patient views and clinician experiences resonated with UK experiences.' (*ibid*, p. 11). The findings were presented as 'bullet points via an online survey and stakeholder groups were invited to comment.' (*ibid*, p. 11).

Reporting on researchers' views, the authors found they believed that laboratory tests are limited, precise and timely diagnosis is important, symptoms make diagnosis a challenge and there are gaps in

the evidence base around diagnosis. Face-to-face consultations found that several of the issues raised by patient advocacy groups corresponded with these researchers' views. Several of the convened groups discussed the limitations of existing laboratory tests and clinicians' alleged overreliance on these. In two consultation events, they report, the limitations of testing for antibodies as opposed to the *Borrelia* organism itself was raised, as was variability in interpretation of test results. Further concerns included the timeliness of testing and diagnosis, barriers to conducting useful research, and a perceived need for rigorous evaluations of alternative, non-standard NHS tests. Like the researchers-patients' convergences, clinicians are also argued to have similar views.

The report also presented 'Findings from studies of patient views and experiences'. Nine studies, all using qualitative research methods, were included. Given that these studies were themselves conducted by researchers interacting directly with patients, we can understand them as in some way representing patients' 'voices' and experiences. Thus, patients can be seen as participants, even if configured as passive rather than as organised participating groups. In fact, the authors suggest that the evidence of patients' views gained was biased:

All of the studies focused on patients who had experienced persistent symptoms... The experiences of those who were diagnosed and treated promptly and who experienced no ongoing problems are notably absent (*ibid*, p. 22-23).

This latter comment may be seen as an attempt to balance the 'evidence' as defined by the review methodology. The results were exposed to 'Feedback from face to face consultations and online survey', concluding that the overall findings were 'felt to resemble their own experiences' (*ibid*, p. 28). The EPPI team summarised:

The UK evidence base on patient experiences of Lyme disease appears to be in the early stages... Several online surveys of UK patient experience were identified...and more were provided by several public advocacy groups (Caudwell Lyme Disease 2016, Newton and et al. 2017, Tick Talk 2016, VIRAS 2014, 2015). However, we were unable to include most of them...because of limitations in reporting methods of sampling, data collection and analysis... (*ibid*, p. 39).

One of the patient surveys cited in the report was that undertaken by Lyme Research UK (Lyme Research UK, undated). Arguably, the authors' filtering approach to critical appraisal of the studies limits what might be called this *secondary participation* of patient organisations in the policy process. Most of the patient experience studies reviewed were non-UK, but the feedback suggested that the UK experiences were very similar. Thus, we can understand one outcome as a strengthening of the credibility of the patient voice as a stakeholder. In fact, this review process was well received by some patient groups, *Fight Lyme Now* saying that it was 'groundbreaking' in taking into account patients' concerns, involving them directly, and noting that the review authors believed that 'Consulting with UK patient advocacy groups helped to mitigate some of these weaknesses (i.e. of international evidence review) as it enabled us to assess the validity of our findings and their relevance to the UK context' (FLN, 2018). Conversely, others believed that the methodology was restrictive, being based on published evidence that was limited, for example on clinicians' knowledge of Lyme disease science, and using feedback methods such as online survey with restricted space for responses, rather than face-to-face discussion (Lyme Research UK/author KB reflective fieldnote, October 2017).

In summary, we have considered patients' participation in this review in two ways, first as direct participants in the organisational processes of interactions through various media, and second as actors-at-a-distance represented in the studies selected and reviewed. This second form of participation has antecedents in STS study of bias in populations constructed for medical research, although focused on racial and gender representation (Epstein, 2007). The convening of face-to-face group discussions and design of explicit feedback processes shows a notable degree of opening up of the policy development process to the 'stakeholders', including activist patient groups that valued this degree of participation, while remaining critical.

## **Discussion**

Healthcare governance is enacted by many powerful actors and institutions, which claim and mobilise a variety of specialist knowledge and expertise. Diagnosis and testing for Lyme disease is a controversial subject where official policy and its evidence base are strongly contested. It is clear that some of the

patient groups bring detailed technical knowledge and evidentiary resources to bear, alongside experiential knowledge. Issues of testing are fundamental to claims to defining the disease and the legitimacy of patients' experiences. The spate of recent policy developments represents a step-change in actors' involvement and processes of healthcare governance around this medical condition. We have seen this in the adoption of the language of 'stakeholders' and 'engagement' and the evidence of the engagement processes that we have analysed.

In the Lyme disease testing case, on the one hand, the presence and extent of contributions of many patient groups in the recent consultations is symbolically significant in itself, but the evidence of the extent of influence is mixed. As we have seen, the technology assessment/review techniques employed by the NICE/NGC and EPPI staff do not pay attention to 'local' factors that might be specific to UK experience of Lyme disease, relying on patient group contributions to contextualise the analysis.

Recalling the spectrum of forms of engagement suggested by de Freitas (2017), the Lyme disease groups and individuals clearly lie toward the 'one-off' end of the spectrum of participatory 'spaces', although the processes have been lengthy and the *amount* of participation in number of contributions is high. The engagement processes observed are clearly based around consultation models orchestrated by the healthcare state. The crux of this is the extreme technical evidence review procedures of the 'systematic review' methods associated with health technology assessment and NICE. The sheer extent of this work lends authority and credibility to the actual guidelines and recommendations eventually produced.

Nevertheless, the presence and activity of patient groups in these processes has been substantial, even though there remains a significant level of dissatisfaction with the process and contents, especially of the eventual NICE guideline.

What further contributions to policy development can be envisaged? Patient group involvement, whether grassroots activist or co-opted or elicited, as in the cases examined here, can have a role in highlighting emerging knowledge in public health and other credentialed forums, including forums such as an academic journal. NICE concluded that the extent to which different tests can be compared is

slight, so further research into tests deemed of higher likely validity could be undertaken. Furthermore, some recent scientific innovations appear promising (e.g. Mavin et al., 2014), and we can note for example development of legitimate scientific evidence about the conversion of microbial infections into chronic conditions, including Lyme disease (O'Connor et al., 2006). Non-mainstream science such as conversion to chronic disease was not considered within the evidence review processes. The variable definition of this type of 'non-evidence' has been described as the procedural crux on which evidence-related guideline production depends (Knaapen, 2012), and patient groups indeed found the definition of relevant evidence to be too narrow, disallowing their own multidimensional experience and forms of knowledge (cf. Rabeharisoa et al., 2014). The NICE review can be regarded as enacting a quantification of large areas of uncertainty, and sidelining of certain types of research as non-evidence, which many patient groups regard as very important.

Lyme disease patient groups' reactions to the NICE guideline varied somewhat between feeling a 'definite achievement through to considerable disappointment' (Lyme Research UK author KB notes), especially in the limitations of the testing and diagnostic processes. Groups believed that participation in the reviews enabled them to orientate their own thinking and increase their own knowledge, so orchestrated consultation has had a mobilising effect. However, it was felt the NICE timelines were extremely tight especially to review and link evidence and experience to very long, detailed evidence reviews (Author KB reflective note), echoing the challenges revealed by studies of guideline participation cited in our Introduction.

From patient groups' perspectives, it was felt by some that patient experience was not properly incorporated in policy formation. NICE sets great store by its patient and patient group engagement procedures (e.g. Findacure.org.uk, 2016) but in our case study a strong form of interactional deliberation was seen primarily only in the initial NICE workshop, rather than the ensuing consultations. The role of 'lay members' (of the NICE committee) and the status of patient groups' consultation comments was prominent. However, for reasons of committee confidentiality, NICE prevented its lay members from deliberating during the process with 'external' patient groups. And while the lived

experience of four individuals with the disease was valued, given the number of medical experts on this key committee, an even broader representation of patients could be considered. Thus, there are reasons to suggest that the NICE and other guideline processes could be improved, including the procedures by which their stakeholder inclusiveness and deliberation is designed and implemented at different stages of a guideline development process.

## Conclusion

Patient groups have made a significant amount of contribution in the governance in Lyme disease testing. The step-change in degree of participation of patient groups is notable but it has not fundamentally altered the selectively evidence-based position of the public health authorities. Aside from the substantive content of the new guidelines, patient groups' participation can be seen to reinforce their legitimacy as both rightful stakeholders in guideline and policy development *and* as patients legitimately in need of both recognition and care. A consequence of the participation process may, paradoxically, be to bolster the credibility of the existing guidance in the eyes of health authorities and practitioners on the one hand, and on the other, to enhance the political identity of the patient organisations taking part - as significant acknowledged actors in high-level policy processes.

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