

‘Drawing a line in the sand’: Physician diagnostic uncertainty in pediatric chronic pain

Alexandra Neville MSc¹, Melanie Noel PhD^{1,2}, Jacqueline Clinch MD³, Tamar Pincus PhD⁴, Abbie Jordan PhD⁵

¹Department of Psychology, University of Calgary and Alberta Children’s Hospital Research Institute, Calgary, AB, Canada

²Hotchkiss Brain Institute, Canada

³Bristol Royal Children’s Hospital, University of Bristol, Bristol, United Kingdom and Royal National Hospital for Rheumatic Diseases, Bath, United Kingdom

⁴Department of Psychology, Royal Holloway, University of London, United Kingdom

⁵Department of Psychology and Centre for Pain Research, University of Bath, United Kingdom

Corresponding Author:

Abbie Jordan, PhD

Department of Psychology, University of Bath, Bath, UK, BA2 7AY

Email: a.l.jordan@bath.ac.uk

Tel.: +44(0)-1225-386982

Category: Original Article

Funding Sources: This work was supported by charitable funding from the Royal United Hospitals Bath NHS Foundation Trust. Alexandra Neville is supported by a Canadian Institutes of Health Research Doctoral Award and an Alberta Innovates Graduate Studentship.

Conflicts of Interest: The authors have no conflicts of interest to declare.

Significance: Physicians experience diagnostic uncertainty in their role treating youth with chronic pain, which has potentially critical implications for youths’ pain experience and health outcomes.

Abstract

Background: Diagnostic uncertainty is the subjective perception of an inability to provide an accurate explanation of the patient's health problem or that a label is missing or incorrect. While recently explored in youth with chronic pain and families, this is the first study to investigate diagnostic uncertainty from the perspectives of physicians. **Methods:** Individual, semi-structured interviews were conducted with sixteen pediatricians who assess and/or treat youth who experience complex chronic pain. Interviews explored pediatricians' perceptions, beliefs, and confidence regarding the assessment and management of chronic pain in youth and how they manage uncertainty regarding the diagnosis. Interviews were analysed using inductive reflexive thematic analysis. **Results:** Analyses generated one prominent theme: 'drawing a line in the sand'. Within this theme, physicians discussed uncertainty as inherent to their role treating youth with chronic pain. The metaphor of 'drawing a line in the sand' was used to describe a process of identifying a point at which physicians no longer sought a new diagnosis for the child's pain or continued diagnostic investigations. This line was influenced by numerous factors, which are highlighted through four subthemes: physician training, experience, and mentorship; individual patient and family factors; perceived reassurance of diagnostic investigations; and the broader social context and implications. **Conclusions:** How physicians manage diagnostic uncertainty must be understood, as it is likely to critically impact how a diagnosis of chronic pain is communicated, the diagnostic investigations undertaken, the wait time to receiving a diagnosis, and ultimately youths' pain experiences.

Key Words: Chronic Pain, Diagnostic Uncertainty, Adolescents, Children, Physicians

Introduction

Many patients with chronic pain do not receive a diagnosis that precisely explains a pathological cause of their pain (Butow & Sharpe, 2013; Serbic & Pincus, 2013). Over 40% of adults with chronic pain experience diagnostic uncertainty (DU), the perception that a label or explanation for a patient's health problem is missing or inaccurate (Serbic & Pincus, 2013), which is associated with elevated levels of anxiety, depression, pain intensity (Serbic et al., 2016), and disability (Geisser & Roth, 1998). Individuals who perceive their pain as not appropriately labeled or legitimized, experience emotional distress and may continue to pursue medical investigations, which contribute to increasing economic burden (Newton et al., 2013).

DU has been identified as a critical area of research in pediatric chronic pain (Pincus et al., 2018). In a qualitative study, youth with chronic pain and their parents expressed needing further explanations for their pain beyond a diagnostic label, and an unrelenting search for an alternative diagnosis that many believed had been missed by clinicians (Neville et al., 2019). Notably, parents and youth perceived that clinicians appeared uncertain, which influenced their confidence in the clinician's understanding of their condition, the diagnosis, and the treatment plan. Thus, DU may have critical implications for pediatric chronic pain outcomes and treatment engagement, and is integrally tied to clinical encounters youth and parents experience.

Although clinicians regularly experience DU, defined as the subjective perception of an inability to provide an accurate explanation of the patient's health problem (Bhise et al., 2018), clinicians experience unique challenges caring for patients with chronic pain. Clinicians who manage adult chronic pain have reported feeling unsuccessful as physicians and in their ability to effectively manage their patients' chronic pain problem (Matthias et al., 2010). In the context of *pediatric* chronic pain, uncertainty has been studied among clinicians in terms of assessing pain

in children with profound cognitive impairment, with clinicians managing uncertainty through seeking mentorship and discussion with colleagues (Carter et al., 2016). The implications of uncertainty among clinicians are enormous at both individual (e.g., unnecessary and invasive medical investigations) and societal (e.g., increased healthcare costs and wait times) levels (Allison et al., 1998; Bhise et al., 2018; Kassirer, 1989; Schneider et al., 2010; Simpkin & Schwartzstein, 2016).

The experiences of DU for youth with chronic pain and their parents are likely linked to how clinicians themselves experience DU. Youth have been found to consult an average of 4 physicians prior to referral to a chronic pain program, with the majority of referrals made by pediatricians (Cucchiaro et al., 2017). Low agreement among pediatricians regarding primary cause and diagnostic approach to treating children with unexplained chronic pain suggests that pediatricians experience considerable uncertainty (Konijnenberg et al., 2004). Understanding DU from the perspectives of pediatricians is critical as they are often the first physicians youth consult when they develop a pain problem. The aim of the current study was to qualitatively explore pediatricians' own perceptions, beliefs, and confidence with regard to the assessment and management of chronic pain in youth.

Methods

Study Design

Adopting both a relativist epistemology and phenomenological approach, this study sought to gain an understanding of how pediatricians understand pediatric chronic pain, how they manage uncertainty regarding the diagnosis of chronic pain in the absence of underlying pathology, and how this diagnosis is delivered to and received by families. In brief, relativism refers to ideas which propose that “our experiences, moral judgments, claims to knowledge, and

so on can be understood only relative to something else such as particular languages and particular social and cultural practices”, and the lack of a single universal truth (Smith, 2008, p. 750), whilst phenomenology refers to a focus on the exploration of experience and meanings (Davidsen, 2013). This study adopted a qualitative semi-structured interview design to allow physicians the flexibility to describe their own personal perspectives and experiences (Mason, 2004). To ensure a rigorous approach to this qualitative analysis, the consolidated criteria for reporting qualitative research (COREQ) checklist were followed (Tong et al., 2007).

Participants

Sixteen pediatricians who assess and/or treat youth who experience complex chronic pain were recruited through established networks, contacts, and healthcare organizations in the United Kingdom (UK). Sociodemographic information of participants is presented in Table 1. Pediatricians were eligible to participate in this study if they treated children and adolescents who experience chronic pain in a UK based healthcare setting. Participants were excluded from the study if they had never treated a child or adolescent with pain that had lasted for three months or longer (i.e., chronic pain) and/or were unable to speak and understand English. Eligibility for this study did not require physicians to have any specific training in pain medicine. Pediatricians, rather than pediatric pain specialists, were purposively sampled as they are the most likely physicians to encounter youth early in their pain problem and prior to their enrollment in a tertiary level pediatric chronic pain program.

As research transparency is perceived as an indicator of quality (Spencer et al., 2003), a focus was placed on being thorough and transparent in our appraisal and reporting of the sample size and research procedure. In keeping with this methodological approach, no attempt was made to achieve saturation (O'Reilly & Parker, 2012). Braun and Clarke contend that there are an

endless number of possible interpretations of a data set, suggesting the impossibility of generating all possible interpretations (Braun & Clarke, 2019b). Rather, specific characteristics of this study, including the study aims, the quality and richness of the data, and the researchers' skills in qualitative methods, are recommended in determining sample size and served as a guide in the current study (Vasileiou et al., 2018). We deliberately recruited a small sample of pediatricians who had experience treating youth with chronic pain in the UK. This is fitting with the theoretically flexible approach of inductive reflexive thematic analysis (Braun & Clarke, 2006). Given the depth of the analysis and richness of the data collected by co-author A.J., who is an expert in the field and an experienced and skilled qualitative researcher, the current sample size met the aims of the study and was methodologically appropriate for the analytical approach (Braun & Clarke, 2013).

Procedure

All study procedures were approved by the Psychology Research Ethics Committee at the University of Bath (ref 17-212). Invitations to take part in the study were circulated through professional paediatric networks. Snowballing was also used through encouraging interested participants to send emails to colleagues. All recruitment notices contained the contact information of co-author A.J., who screened all interested participants for eligibility. Study procedures were verbally reviewed with the physician prior to participation and verbal informed consent was obtained. Interested participants were then emailed a consent form via Qualtrics online survey software (Qualtrics, 2019). Following enrollment and consent to participate in the study, participants completed a demographic questionnaire online via Qualtrics. Semi-structured interviews were conducted over the phone by co-author A.J., who did not know any of the participants personally. Interview duration ranged between 18-45 minutes (mean duration = 29

minutes). Interviews were audio-recorded and transcribed verbatim. To ensure the anonymity of participants and related individuals, all identifying information was removed from the transcripts and participant numbers were used. Participants received a £20 Amazon voucher to thank them for their time.

Measures

Demographic Characteristics. Pediatricians reported on their gender, years of training, job title, medical specialty, and the clinical setting in which they worked at the time of participating in the study.

Interview Schedule

The interview schedule (Table 2) focused on exploring physician beliefs and confidence surrounding the diagnoses that they provide to youth with chronic pain and their families. In particular, physicians were asked to discuss how the diagnosis is offered, their confidence in the accuracy of the diagnosis, how they manage uncertainty regarding the diagnosis in the absence of underlying disease/pathology, and how they perceive that the diagnosis is received and understood by the child and their parent(s). The interview schedule was semi-structured and comprised open-ended questions and prompts in order to allow the interviewer to follow-up and expand on topics of interest as they occurred during the interviews (Mason, 2004).

Data Analysis

Data analysis was completed using QSR International's NVivo 12 qualitative data analysis software, a computer-assisted qualitative data analysis package (NVivo qualitative data analysis software, 2018). Data were analyzed using inductive reflexive thematic analysis, in accordance with Braun and Clarke's (Braun & Clarke, 2006, 2019a) six-phase thematic analysis guide. This is a theoretically flexible method of qualitative data analysis that is not limited to a

single epistemological view (Nowell et al., 2017) and allows for a detailed and complex account of qualitative data (Braun & Clarke, 2006). Inductive thematic analysis is a “bottom-up” approach aiming to identify patterns of meaning from data (Braun & Clarke, 2006, 2019a). In the reflexive thematic analysis approach, themes are conceptualized as meaning-based patterns (Braun & Clarke, 2006, 2019a). Regarding specific steps taken, following transcription of the data and verification of the transcriptions, A.N. and A.J. read and re-read the transcripts in order to familiarize themselves with the data. Through an iterative process, the data were thoroughly and comprehensively coded by A.N. and A.J. Codes were then collated to identify potential themes. In accordance with Braun & Clarke (Braun & Clarke, 2006), analysis extended beyond description and the data were interpreted for meaning. To address issues around quality of qualitative research, themes and interpretations were iteratively reviewed and refined through discussions with all co-authors, ensuring credibility and trustworthiness in the data and analytic interpretations (Elliott et al., 1999; Morrow, 2005). Definitions of each theme were generated, paying close attention to the specifics of each theme, the relation of each theme with others, as well as the overall story of the data (Braun & Clarke, 2006). Lastly, data were analysed within each theme. Refinement of themes led to the generation of one robust theme within the data with four subsequent sub-themes. Example quotations of this theme and corresponding sub-themes were selected from a variety of participants for the final written report.

Results

A single, complex, prominent theme was generated from the thematic analysis, which was labeled ‘drawing a line in the sand’. Within this single theme, four subthemes were generated including: physician training, experience, and mentorship; individual patient and family factors;

perceived reassurance of diagnostic investigations; and the broader social context and implications. Below, this theme and subthemes are presented along with supporting quotations from a range of participants. Identifying information has been removed from quotations and physician numbers have been used to protect the confidentiality of participants.

Drawing a line in the sand

The experience of uncertainty and how to manage it were salient features of physician accounts, with participants describing this as inherent to their role of treating youth with chronic pain. A key aspect of managing DU for physicians related to their perceived need to be able to identify a point at which they no longer sought a new diagnosis for the child's pain. In particular, the identification of a defining point at which the decision is made to cease all diagnostic testing as part of this wider process was described as being akin to "drawing a line in the sand". This suggests a clear limit beyond which investigations would proceed any further. However, in reality, determining where and when this line should be drawn was far more complex than the metaphor implies. Despite the acknowledgment of needing to draw a line, the decision to do so was complicated by numerous factors including aspects of the individual case, as well as physicians' *own* need for reassurance. Further, even when this line in the sand had been drawn, it was not as definitive as one might first imagine. This is illustrated by Physician 4 below who explained that even when this line is drawn, some physicians may continue to follow the patient 'just in case' an underlying 'cause' had indeed been missed, even when they perceived no sinister cause for the child's pain.

And so, what I will do is set the line in the sand and then lead forward, make sure the diagnosis is there, and then keep seeing the child just in case I've missed something, and also to manage their pain. (Physician 4, woman, 17 years qualified)

Physician training, experience, and mentorship

Physicians' ability to sit with uncertainty and cease diagnostic testing was influenced by a variety of factors, such as the specific nature of their medical training. Interestingly, physicians described how their work treating youth with chronic pain was the antithesis of the focus of their clinical training. Consequently, physicians discussed how their medical training had focused on finding a diagnosis and "fixing" a problem, which conflicted with their current role of managing children's chronic pain where a clear underlying 'cause' and subsequent 'cure' was often lacking. In particular, physicians reported difficulties with adjusting to the requirements of chronic pain treatment, which were perceived to be based on principles that were in opposition with their lengthy clinical diagnostic training.

I think because it's not what we're trained to do, is it, as clinicians we are trained to find a problem, sort it out, give the medication or whatever it is, the treatment, and everybody's happy, everybody moves on, problem solved and that's it. But with chronic pain ... you can't do that, and so it's a completely different scenario, a different situation, and you just have to accept that sometimes there is no cause and there is no answer in terms of why, and whatever the situation is that you're faced with, that's the situation that you've got to deal with and try to improve. (Physician 1, woman, 28 years qualified)

Due to the perceived incongruence between managing DU in pediatric chronic pain and previous medical training, physicians described needing to adopt a new understanding of their role and identity as a physician. As part of this reconfiguration, they described how they needed to reformulate their own definition of success from that of symptom reduction to encouraging adaptive functioning in their patients. To achieve this, physicians described re-examining their entire aim and philosophy of care, how they approach this with families, and their role in supporting patients. These processes resulted in physicians actively redefining success as no longer being contingent on symptom management.

Well, for me it's not about getting them [patients] better, it's about allowing them to thrive as a person despite them having their symptoms. So, I'm not expecting to get anyone better, but I am expecting them to go through school, get their qualifications, do what they want to do in

life. But I don't tend to rely on symptoms to know that I've done a good job. (Physician 2, woman, 25 years qualified)

The 'line in the sand' was influenced by a multitude of factors, with one including the extent of physician experience associated with assessing and managing children with idiopathic chronic pain. Experience occurred 'on the job' and learning typically involved interacting with other more experienced colleagues in the form of regular meetings and case conferences. Consequently, with increasing experience, physicians noticed themselves becoming more confident in treating youth with chronic pain and in turn, in reducing the number of diagnostic tests that they ordered. Whilst such results are inherently positive, this developmental trajectory in physicians' learning and comfort with risk means that there are clear timepoints during which many families are likely exposed to repeated, and possibly unnecessary, diagnostic testing. Moreover, Physician 16's use of the word 'brave' below implies that this learning process is endured in the face of physician fears and conflicts with their previous training that underscored the importance of caution and certainty in the diagnostic process. This process of learning may also be linked to physicians' earlier conceptualizations of chronic pain as a symptom without an underlying cause rather than the disease in and of itself.

My colleagues have been doing it a little bit longer, but we've got braver over time. So, I was already moderately brave about the headache, I don't do a lot of scans compared to other colleagues who don't do headache much. But we've noticed with this more difficult group of patients actually, we're also getting braver and doing fewer tests as time goes by, because I think you get a reflective memory for how to manage it. (Physician 16, woman, 26 years qualified)

Physicians described that without an adequate mentorship and support system, such as is characteristic of a multidisciplinary team, they often felt isolated. Lack of experience and colleague support led some physicians to manage DU through continuing to see families past

their ‘the line in the sand’. Some physicians recognized the potential devastating impact this could have on youth and their families.

I think if you're not in a multidisciplinary team with these cases, it's very isolating... I think your gut feeling is to keep bringing them [youth with chronic pain] back, because you can't solve their problem but you don't know what else to do. And what I'm trying to make my colleagues realise is that perhaps just keeping on seeing them is possibly one of the worst things you can do. But I can totally see where they're coming from, it's exactly what I would have done a few years ago before we started working in this way.” (Physician 14, woman, 24 years qualified)

At times, managing uncertainty took an emotional toll on physicians. Strategies to manage uncertainty included sharing feelings of uncertainty with fellow physicians, although this process did not fully alleviate the uncertainty they experienced. Rather, it was described as a means to reduce the discomfort associated with experiencing uncertainty and to increase physicians’ sense of comfort with managing a particular level of risk. Many physicians sought mentorship from more senior clinicians, who were more experienced both in terms of employment duration as a clinician and in treating patients with chronic pain specifically, as a way to ease uncertainty. Particularly for less experienced physicians, support from colleagues appeared to serve as an unofficial training experience and reassurance that they were not missing a more sinister cause for the pain.

Yeah, there's been weekends where I've been really thinking, oh my goodness, have I missed something, what if it's this, what if it's that, I've never come across this before... So how do I cope with that? I basically seek mentorship from a senior... So, I would go down and I would sit down and say, look, I'm really worried about this patient, I'm pretty sure that is chronic pain or medically unexplained, but I'm just worried I could be missing something, and go through the story. And on the whole, she's always been really reassuring and will say this definitely fits this picture, I've seen patients exactly like this. (Physician 10, woman, 16 years qualified)

Individual patient and family factors

Explanations of physicians’ willingness or ability to sit with uncertainty were complex and multi-factorial. Physician 3, below, describes how patient differences and individual

thresholds were some reasons offered by physicians with regard to explaining their ability to sit within a particular level of uncertainty. For example, individual thresholds for severity of symptoms influenced whether a physician might seek out another colleague to share their uncertainty.

I'm very comfortable as a general pediatrician to sit with uncertainty. For me it's just about your threshold and the severity of the symptom. I suppose if the symptoms are within a realm that I'm comfortable with, then I'm happy to sit with uncertainty, if the symptoms are clearly worse then I'm going to struggle, because it's all about how severe the problem is, I think. I mean, I'm happy to sit with uncertainty, but I have my limit. (Physician 3, man, 14 years qualified)

Memory and experience were important for both physicians and patients, influencing individuals' perceptions of where the 'line in the sand' could be drawn in terms of no longer seeking a medical explanation for the child's pain. Physicians recognized that youth and parents were influenced by their own previous medical experiences, including interactions with clinicians along that journey. The physicians' sense of patient and family beliefs, 'buy-in', and past medical experiences (e.g., a family member diagnosed with a brain tumor, experiencing a misdiagnosis) led them to continue investigations *beyond* the point of actually needing further diagnostic information. Thus, the ability and willingness of physicians to 'draw a line in the sand' was also influenced at a more individual level, with characteristics and beliefs of each youth and their family influencing physicians' decisions about when to halt ordering diagnostic tests to identify a 'cause' for the pain.

And, unfortunately, the way the family are and their belief in you and how the consultation has gone, does affect probably what investigations you order. So, if they're dead set on the fact that this is definitely cancer, that might push you towards doing more investigation, for example. (Physician 8, woman, 15 years qualified)

Physicians also recognized that the previous experiences that patients had before arriving to their clinic influenced both the patients' alignment with the physician as well as their

understanding and acceptance of the diagnosis. However, physicians observed that this alliance was hampered for other patients and families in situations where many clinicians had been seen and multiple explanations of pain had been previously provided. Indeed, this was seen as resulting in mistrust in the medical system. In such situations, this mistrust due to past experience invariably affected ‘buy in’ and alignment with physicians. Misalignment of agendas, such as when families continue to believe that an underlying cause for the pain has yet to be found, was seen as a major barrier to moving forward with treatment. When a discrepancy in views occurs, how this is negotiated may have implications on patient acceptance of a diagnosis, treatment, and general trust in the medical system.

And I think it depends what their experience has been before, because I think if you get someone quite early on in the process then you are more likely to get a family that are going to be amenable to your way of thinking about it and buy into that explanation of chronic pain and not carry on thinking that they need investigations into that. But if you get somebody that's had this problem for quite a long time and they've had lots of different opinions from different doctors, and that's really difficult and then they lose trust and you've completely lost them then. (Physician 7, woman, 17 years qualified)

Additionally, this once considered definitive ‘line in the sand’ shifted according to physician perceptions of the severity of fears and anxieties held by individual families and the importance that the physician ascribed to managing these fears and anxieties.

But for some families they need a little bit more than that, and so they've got specific, focused concern and so in fact the risk-benefit is doing investigation to allay their anxiety around that particular thing as a line in the sand. (Physician 4, woman, 17 years qualified)

Perceived reassurance of diagnostic investigations

Physicians described, at times, adopting a cost-benefit analytic approach, such that they occasionally used diagnostic tests to curtail further, more invasive procedures. In an endeavor to prevent potential serious and unnecessary damage to a child, such as undergoing surgery,

physicians reported ordering further diagnostic testing for the sole means of assuaging family or patient worries of a sinister cause of the pain. Yet, ordering further investigations was reported to be contradictory to physicians' professional judgment and held the explicit expectation that nothing would be found. Indeed, this induced considerable discomfort in physicians. However, when benefits were perceived to be possible, such as reassuring the family and preventing invasive procedures, discomfort with acting against clinical judgment was perceived to be the more comfortable option and could be described as the lesser of two evils.

And then occasionally you meet people that are a little bit more entrenched in their views. So sometimes I have to in those situations done extra scans and things like that, like MRIs, just to say, look, there really is nothing, you can see there, we're not going to go and open you up to look at your tummy, you know this has to stop here sort of thing. (Physician 7, woman, 17 years qualified)

Physicians perceived families to be reassured by diagnostic testing and the return of test results that indicate the absence of pathology. As expressed by Physician 15 below, 'normal' or negative test results were believed by some physicians to reduce anxiety in children and families with regard to reinforcing the idea that the pain was not caused by something more serious.

It can actually be reassuring to have a normal test of some sort because it takes that worry [that something more sinister can explain the pain] away. It doesn't necessarily always cure the pain but it takes that worry away. (Physician 15, man, 24 years qualified)

In direct opposition to the perception of the reassuring and necessary nature of diagnostic investigations for families, a small number of physicians recognized the potentially harmful nature of unnecessary diagnostic tests for families in terms of exacerbating existing levels of patient anxiety and uncertainty about the cause of the pain.

I am aware that ordering investigations is not always reassuring for the patients, in fact it can make them feel that actually you do think there's something wrong, and then there's more anxiety leading up to the tests. And then when you've got the result it's sort of are you sure, you obviously thought this was really bad because you had to order this. On the other hand, you have to sort of do it on a patient-by-patient basis. (Physician 8, woman, 15 years qualified)

Furthermore, some physicians strongly reported that such tests were a necessary component to achieving family ‘buy-in’ to a chronic pain diagnosis. Until all possibilities had been ruled out, family beliefs about other diagnoses would remain a barrier to patient-physician communication and hamper any progress in terms of chronic pain treatment. This suggests that extraneous testing was seen by some as a necessary step in the direction towards initiating chronic pain treatment, providing a stark contrast with other physicians who worked hard to avoid such testing in their practice. This might also reflect differences in physician perceived self-efficacy in their ability to generate patient buy-in through effective communication.

My personal view is that there's no point in trying to counsel them in the management of chronic pain until you've ruled everything else out, because they're just hanging on that diagnosis and won't really listen to you. (Physician 2, woman, 25 years qualified)

The broader social context and implications

The decision of whether to conduct further investigations for potential ‘causes’ for the pain involved balancing a number of complex factors such as the ‘need’ for investigation and/or reassurance, the invasiveness of a test, and the broader societal implications, such as costs to the medical system. Physicians described following a stepped care approach according to the nature and invasiveness of a test. At times, physicians compromised with families in terms of deciding where to draw their ‘line in the sand’ with regard to deciding to stop ordering investigative tests. This was often presented as a compromise between the physicians’ desires to stop testing, costs to the family and healthcare system, and individual desires of the child and their family. Often ordering medical tests were used by physicians as a tool to align with families, even when they believed that these tests were medically unnecessary.

I think tummy aches, again might do a blood test and a urine test, and in some cases might go as far as an ultrasound scan, on the basis that it's not invasive, non-painful, relatively cheap

thing to do and often is one that you can easily draw a line under with the families. I try really hard to avoid more expensive, more invasive imaging if I possibly can... I guess in my mind I'm balancing up not wanting to propagate the medical belief, but also only considering something ... because I think sometimes we do order tests even though we know that they're going to be normal, so you are using the test as a way of drawing a line in what you're going to do... (Physician 14, woman, 24 years qualified)

Interestingly, some physicians perceived that unnecessary medical investigations could be financially beneficial for the health care system if they prevented families from continuing to present for further more costly investigations.

You could argue, yeah, don't do an MRI, that costs the health service, but if it stops that person keep presenting and is also a saving as well, if it stops loads of medicines being prescribed, then there's a saving. So, I suppose it's like balancing that up in your mind. (Physician 7, woman, 17 years qualified)

Some physicians felt reassured by the number of clinicians that patients had previously seen and the amount of investigations completed, prior to the youth being in their care. Previous consultations with colleagues served to dilute this sense of individual responsibility with regard to managing a young person with idiopathic chronic pain. Physician 10 below acknowledges the wider social context in which uncertainty is experienced and managed.

...because they've seen so many other people before me, generally rheumatologists in [city] or neurosurgeons and had every scan under the sun, I feel really reassured by that and it's quite clear and barn door [obvious] to me, I think, that there doesn't need to be any worry about missing something and it's chronic pain. (Physician 10, woman, 16 years qualified)

Physicians' experiences and beliefs about uncertainty and how this might affect the patient-physician relationship influenced the extent to which physicians shared their uncertainty with families. For some, sharing uncertainty with families was considered to be risky and to undermine their competence as physicians with families. Others felt confident sharing their uncertainty with families, claiming that sharing reduced the power differential between physician, patient, and family. For these physicians, uniting with the family in the face of

uncertainty was perceived to demonstrate humanity and increase the therapeutic alliance between the physician and family.

I think people often worry when they're new to this [managing children with unexplained pain] and with the new consultants, that if you demonstrate uncertainty that you undermine a family's confidence in you. But I don't think that's the case, I think that what happens is with uncertainty I think they respond to you as a human being and as a professional and they respect the honesty, and they don't feel patronised, I think, by that, I think they feel more like it's a team effort. (Physician 11, woman, 19 years qualified)

Discussion

This study explored physicians' experience of DU regarding the assessment and management of chronic pain in youth. Through the theme 'drawing a line in the sand', physicians described how they manage uncertainty in their role, and the complex factors involved in their decisions of whether to continue or cease diagnostic testing. Even after a 'line in the sand' had been drawn, this point after which no further investigations would be sought was likely to shift depending on multiple factors, which were highlighted through four subthemes: physician training, experience, and mentorship; individual patient and family factors; perceived reassurance of diagnostic investigations; and the broader social context and implications.

Degree of experience and stage of training were key dimensions of physicians' experience of uncertainty. Many physicians reported receiving no formal training in chronic pain and described all of their learning as occurring 'on the job'. This highlights important gaps in pain education, which has been found to be inadequate both in the UK and internationally (Bhatia et al., 2008; Briggs et al., 2015; Hurley-Wallace et al., 2019; Shipton et al., 2018). Physicians discussed incongruence between their medical training, which focused on diagnosing and "fixing" a problem, and their current role in managing chronic pain in youth, which required supporting adaptive functioning despite pain. A lack of training was evident through physicians'

descriptions of having to redefine their aim of care and turn to more experienced colleagues to learn about managing chronic pain. However, more experienced physicians may not necessarily be more knowledgeable regarding chronic pain and may have received training at a time when pain was even less understood. If physicians were not part of a multidisciplinary team, which could provide this type of mentorship, they described feeling isolated in managing DU and also continuing to see families past the ‘the line in the sand’. A lack of training in pediatric chronic pain likely drives physicians’ perceptions of its etiology. Physicians expressed not subscribing to the notion and conceptualization of chronic pain as being the disease in and of itself. The communication of this belief to families may contribute to uncertainty among youth and their parents (Neville et al., 2019).

While some physicians perceived sharing their uncertainty as being beneficial, research shows that this belief is not shared by families. Indeed, perceptions of clinician uncertainty can fuel DU in youth and parents, and can lead to mistrust in clinicians and the medical system (Neville et al., 2019). Physician intolerance of uncertainty has been found to be positively associated with diagnostic action (Schneider et al., 2010). In the current study, physician experience was noted to influence their actions; physicians described becoming “braver” over time and ordering fewer tests. This parallels findings in other areas of medicine. In a recent vignette study, more experienced emergency department physicians reported less risk averse decision making (ordering fewer tests, offering fewer treatments, and making fewer referrals) (Lawton et al., 2019). Unfortunately, the consequence of this ‘on the job’ learning and gradual development of tolerance of uncertainty, is that youth are repeatedly exposed to potentially unnecessary and harmful diagnostic testing (e.g. MRI, ultrasound, x-ray, endoscopy). A retrospective study of medical records of youth seen in a chronic pain clinic found that 15% of

youth underwent surgery due to their pain problem prior to referral to the pain clinic, which resulted in no change to their pain (Cucchiaro et al., 2017).

The ‘line in the sand’ was also influenced by physicians’ perceptions of individual parent and youth characteristics, including severity of symptoms. Perceptions of parent and youth DU influenced physicians’ decision making. The nature of idiopathic chronic pain in terms of its limited improvement of symptom severity, lack of underlying pathology, and use of non-pharmacological treatments (Fisher et al., 2018; Friedrichsdorf et al., 2016; Lynch-Jordan et al., 2014), make certainty difficult to attain. Many physicians believed that diagnostic tests would serve to reassure and assuage family anxiety that a sinister cause of the pain had been missed, even if the pediatrician expected tests would not indicate pathology. Physicians described taking a stepped care approach to investigations, according to the nature, invasiveness of a test, potential for harm, and need to align with the family. Nevertheless, unnecessary testing increases healthcare expenditures and carries risks of false positives and iatrogenic injury (Simpkin & Schwartzstein, 2016). Further, continuous negative test results and prolonged time with a lack of definitive answers may lead to increased psychological issues and distress for youth with chronic pain and their families, and exacerbate their uncertainty and fears that something is still being missed (Neville et al., 2019). Thus, where many pediatricians reported ordering further tests to alleviate uncertainty around a diagnosis of idiopathic chronic pain, this may have the unintended opposite effect. Understanding the impact of unnecessary diagnostic testing is also important given that patients’ wellbeing declines while waiting for chronic pain treatment (Lynch et al., 2008; Palermo et al., 2019). Only a minority of physicians acknowledged that unnecessary diagnostic investigations may cause psychological harm, pointing to the need for improved pain education for pediatricians. Physicians acknowledged the impact that their patients’ past

experiences with the medical system (e.g., misdiagnosis) had on their ability to ‘buy in’ to a chronic pain diagnosis. The number and nature of the explanations for pain that patients and families were previously provided were seen to have a negative influence on their current diagnostic orientation. This is in line with previous literature showing DU is fueled by time spent waiting for a diagnosis, previous interactions with clinicians, and past experiences when a diagnosis had been missed (Neville et al., 2019).

In terms of further diagnostic testing, the lengths that physicians were willing to go to reflected their perceptions of a critical need to align with families. Misalignment in agendas between the physician and family was seen as a crucial barrier in treatment. Physicians' beliefs in, and reliance on, the ‘reassurance’ of tests may reflect a lack of self-efficacy and training in the communication and delivery of a diagnosis to patients, particularly of idiopathic chronic pain. How physicians explain chronic pain (e.g., pain neuroscience education), and tailor this explanation to the developmental stage of the child, has been identified as a critical area of future research (Robins et al., 2016) and may have an important impact on alignment of physician and patient agendas. The oral testimonies of patients with chronic pain revealed that a breakdown in communication between patients and clinicians led youth to perceive that clinicians did not attend to their lived experiences, which resulted in them doubting that their clinicians were able to help (Natkiewicz, 2008).

The current study should be viewed in light of limitations. Results are based on interviews with pediatricians in the UK and may differ geographically. While not expressed in the current interviews, in certain contexts, the ‘line in the sand’ described in these interviews may also be influenced by physician fears of litigation and socio-cultural and political factors. This should be explored in future research. Due to a recruitment process wherein pediatricians

self-selected in to the study, the current sample likely represents pediatricians who have a particular interest in chronic pain and findings may thus even underrepresent the uncertainty physicians experience in assessing and managing pediatric idiopathic chronic pain. We are unable to report on additional context regarding the healthcare settings (e.g., typical pattern of care, how long physicians typically follow patients) of participants. Nevertheless, these physicians reported little to no formal training in pediatric chronic pain and are the most likely clinicians to encounter youth in the early stages of their pain problem (Cucchiaro et al., 2017). This is important, as these interactions leave a lasting impact on families and set the stage for DU at the point of entry into a chronic pain program (Neville et al., 2019). Future research is needed to understand the experience of diagnostic uncertainty among pediatric pain specialists and the potential impact of pain education on DU among physicians and other healthcare professionals. Notably, the current findings emphasize a need for improved pain education for pediatricians, access to multidisciplinary pain teams and formalised mentorship schemes.

The experience of DU is inherent to the role of pediatric chronic pain provider and invariably influences how DU is experienced by youth and their parents (Neville et al., 2019). Given the widespread problem and economic toll of pediatric chronic pain (Groenewald et al., 2014) and potential consequences of delayed treatment (Lynch et al., 2008), understanding the factors involved in DU for youth with chronic pain, their parents, and clinicians is crucial. These physician accounts showed that, in the face of DU, physicians order more tests to reassure *themselves*, in addition to youth and their families. In this way, the management of DU may lead to potentially unnecessary and harmful diagnostic investigations in youth and longer wait times for a diagnosis, which can have devastating consequences for youths' health, trust in the medical system, and pain outcomes (Lynch et al., 2008; Neville et al., 2019). Thus, understanding DU

among clinicians has critical implications for harm reduction. Recent development of a new classification system for chronic pain as part of the Classification of Diseases (ICD-11) offers opportunity for consistency in language and internationally recognized labels for children's pain. Future research is needed to examine optimal ways in which physicians can deliver a diagnosis and communicate with youth and their parents about chronic pain. This messaging is likely influenced by physicians' own uncertainty and invariably influences DU of patients and families.

Author Contributions: All authors discussed the results and commented on the manuscript.

Alexandra Neville: analysis, writing – original draft, and writing – review and editing

Melanie Noel: analysis, writing – review and editing

Jacqueline Clinch: writing – review and editing

Tamar Pincus: writing – review and editing

Abbie Jordan: conceptualization, methodology, data collection, analysis, writing – review and editing

References

- Allison, J. J., Kiefe, C. I., Cook, F., Gerrity, M. S., Orav, J., & Centor, R. (1998). The association of physician attitudes about uncertainty and risk taking resource use in a medicare HMO. *Medical Decision Making, 18*(3), 320-329.
- Bhatia, A., Brennan, L., Abrahams, M., & Gilder, F. (2008). Chronic pain in children in the UK: a survey of pain clinicians and general practitioners. *Pediatric Anesthesia, 18*(10), 957-966. doi:10.1111/j.1460-9592.2008.02710.x
- Bhise, V., Rajan, S. S., Sittig, D. F., Morgan, R. O., Chaudhary, P., & Singh, H. (2018). Defining and measuring diagnostic uncertainty in medicine: A systematic review. *Journal of General Internal Medicine, 33*(1), 103-115. doi:10.1007/s11606-017-4164-1
- Braun, V., & Clarke, V. (2013). *Successful Qualitative Research: A Practical Guide for Beginners*. Los Angeles, CA: Sage
- Braun, V., & Clarke, V. (2006). Using thematic analysis in psychology. *Qualitative Research in Psychology, 3*(2), 77-101.
- Braun, V., & Clarke, V. (2019a). Reflecting on reflexive thematic analysis. *Qualitative Research in Sport, Exercise and Health, 11*(4), 589-597. doi:10.1080/2159676X.2019.1628806
- Braun, V., & Clarke, V. (2019b). To saturate or not to saturate? Questioning data saturation as a useful concept for thematic analysis and sample-size rationales. *Qualitative Research in Sport, Exercise and Health*. doi:10.1080/2159676X.2019.1704846
- Briggs, E. V., Battelli, D., Gordon, D., Kopf, A., Ribeiro, S., Puig, M. M., & Kress, H. G. (2015). Current pain education within undergraduate medical studies across Europe: Advancing the Provision of Pain Education and Learning (APPEAL) study. *BMJ Open, 5*(8), e006984. doi:10.1136/bmjopen-2014-006984
- Butow, P., & Sharpe, L. (2013). The impact of communication on adherence in pain management. *Pain, 154 Suppl 1*, S101-107. doi:10.1016/j.pain.2013.07.048
- Carter, B., Simons, J., Bray, L., & Arnott, J. (2016). Navigating uncertainty: Health professionals' knowledge, skill, and confidence in assessing and managing pain in children with profound cognitive impairment. *Pain Research and Management, 2016*, 8617182. doi:10.1155/2016/8617182
- Cucchiario, G., Schwartz, J., Hutchason, A., & Ornelas, B. (2017). Chronic pain in children: A look at the referral process to a pediatric pain clinic. *International Journal of Pediatrics, 2017*, 8769402. doi:10.1155/2017/8769402
- Davidsen, A. S. (2013). Phenomenological approaches in psychology and health sciences. *Qualitative Research in Psychology, 10*(3), 318-339. doi:10.1080/14780887.2011.608466
- Elliott, R., Fischer, C. T., & Rennie, D. L. (1999). Evolving guidelines for publication of qualitative research studies in psychology and related fields. *British Journal of Clinical Psychology, 38*, 215-229.
- Fisher, E., Law, E., Dudeney, J., Palermo, T. M., Stewart, G., & Eccleston, C. (2018). Psychological therapies for the management of chronic and recurrent pain in children and adolescents. *Cochrane Database Systematic Reviews, 9*, CD003968. doi:10.1002/14651858.CD003968.pub5
- Friedrichsdorf, S. J., Giordano, J., Desai Dakoji, K., Warmuth, A., Daughtry, C., & Schulz, C. A. (2016). Chronic pain in children and adolescents: Diagnosis and treatment of primary pain disorders in head, abdomen, muscles and joints. *Children (Basel), 3*(4). doi:10.3390/children3040042

- Geisser, M. E., & Roth, R. S. (1998). Knowledge of and agreement with chronic pain diagnosis: Relation to affective distress, pain beliefs and coping, pain intensity, and disability. *Journal of Occupational Rehabilitation, 8*(1), 73-88. doi: 10.1023/A:1023060616201
- Groenewald, C. B., Essner, B. S., Wright, D., Fesinmeyer, M. D., & Palermo, T. M. (2014). The economic costs of chronic pain among a cohort of treatment-seeking adolescents in the United States. *The Journal of Pain, 15*(9), 925-933. doi:10.1016/j.jpain.2014.06.002
- Hurley-Wallace, A., Wood, C., Franck, L. S., Howard, R. F., & Liossi, C. (2019). Paediatric pain education for health care professionals. *Pain Reports, 4*(1), e701. doi:10.1097/PR9.0000000000000701
- Kassirer, J. P. (1989). Our stubborn quest for diagnostic certainty. A cause of excessive testing. *The New England Journal of Medicine, 320*(22), 1489-1491. doi:10.1056/NEJM198906013202211
- Konijnenberg, A. Y., De Graeff-Meeder, E. R., Kimpen, J. L., van der Hoeven, J., Buitelaar, J. K., Uiterwaal, C. S., & Pain of Unknown Origin in Children Study Group. (2004). Children with unexplained chronic pain: Do pediatricians agree regarding the diagnostic approach and presumed primary cause? *Pediatrics, 114*(5), 1220-1226. doi: 10.1542/peds.2004-0355
- Lawton, R., Robinson, O., Harrison, R., Mason, S., Conner, M., & Wilson, B. (2019). Are more experienced clinicians better able to tolerate uncertainty and manage risks? A vignette study of doctors in three NHS emergency departments in England. *BMJ Quality & Safety, 28*(5), 382-388. doi:10.1136/bmjqs-2018-008390
- Lynch, M. E., Campbell, F., Clark, A. J., Dunbar, M. J., Goldstein, D., Peng, P., Stinson, J., & Tupper, H. (2008). A systematic review of the effect of waiting for treatment for chronic pain. *Pain, 136*(1-2), 97-116. doi:10.1016/j.pain.2007.06.018
- Lynch-Jordan, A. M., Sil, S., Peugh, J., Cunningham, N., Kashikar-Zuck, S., & Goldschneider, K. R. (2014). Differential changes in functional disability and pain intensity over the course of psychological treatment for children with chronic pain. *Pain, 155*(10), 1955-1961. doi:10.1016/j.pain.2014.06.008
- Mason, J. (2004). Semi structured interview. In M. Lewis-Beck, Bryman, A, Liao, TF. (Ed.), *Encyclopedia of Social Science Research Methods*. Thousand Oaks, CA: SAGE.
- Matthias, M. S., Parpart, A. L., Nyland, K. A., Huffman, M. A., Stubbs, D. L., Sargent, C., & Bair, M. J. (2010). The patient-provider relationship in chronic pain care: providers' perspectives. *Pain Medicine, 11*(11), 1688-1697. doi:10.1111/j.1526-4637.2010.00980.x
- Morrow, S. L. (2005). Quality and trustworthiness in qualitative research in counseling psychology. *Journal of Counseling Psychology, 52*(2), 250-260. doi:10.1037/0022-0167.52.2.250
- Neville, A., Jordan, A., Beveridge, J. K., Pincus, T., & Noel, M. (2019). Diagnostic uncertainty in youth with chronic pain and their parents. *The Journal of Pain*. doi:10.1016/j.jpain.2019.03.004
- Newton, B. J., Southall, J. L., Raphael, J. H., Ashford, R. L., & LeMarchand, K. (2013). A narrative review of the impact of disbelief in chronic pain. *Pain Management Nursing, 14*(3), 161-171. doi:10.1016/j.pmn.2010.09.001
- Nowell, L., Norris, J., White, D., & Moules, N. (2017). Thematic analysis: Striving to meet the trustworthiness criteria. *International Journal of Qualitative Methods, 16*, 1-13.
- Nutkiewicz, M. (2008). Diagnosis versus dialogue: oral testimony and the study of pediatric pain. *Oral History Review, 35*(1), 11-21. doi:10.1093/ohr/ohm002

- NVivo qualitative data analysis software. (2018). Version 12. QSR International Pty Ltd.
- O'Reilly, M., & Parker, N. (2012). 'Unsatisfactory saturation': A critical exploration of the notion of saturated sample sizes in qualitative research. *Qualitative Research, 13*(2), 190-197.
- Palermo, T. M., Slack, M., Zhou, C., Aaron, R., Fisher, E., & Rodriguez, S. (2019). Waiting for a pediatric chronic pain clinic evaluation: A prospective study characterizing waiting times and symptom trajectories. *The Journal of Pain, 20*(3), 339-347. doi:10.1016/j.jpain.2018.09.009
- Pincus, T., Noel, M., Jordan, A., & Serbic, D. (2018). Perceived diagnostic uncertainty in pediatric chronic pain. *Pain*. doi: 10.1097/j.pain.0000000000001180
- Qualtrics. (2019). Qualtrics software. Provo, UT, USA.
- Robins, H., Perron, V., Heathcote, L. C., & Simons, L. E. (2016). Pain neuroscience education: State of the art and application in pediatrics. *Children (Basel), 3*(4). doi: 10.3390/children3040043
- Schneider, A., Lowe, B., Barie, S., Joos, S., Engeser, P., & Szecsenyi, J. (2010). How do primary care doctors deal with uncertainty in making diagnostic decisions? The development of the 'Dealing with Uncertainty Questionnaire' (DUQ). *Journal of Evaluation in Clinical Practice, 16*(3), 431-437. doi: 10.1111/j.1365-2753.2010.01464.x
- Serbic, D., & Pincus, T. (2013). Chasing the ghosts: the impact of diagnostic labelling on self-management and pain-related guilt in chronic low back pain patients. *Journal of Pain Management, 1*(6), 25-35.
- Serbic, D., Pincus, T., Fife-Schaw, C., & Dawson, H. (2016). Diagnostic uncertainty, guilt, mood, and disability in back pain. *Health Psychology, 35*(1), 50-59. doi: 10.1037/hea0000272
- Shipton, E. E., Bate, F., Garrick, R., Steketee, C., Shipton, E. A., & Visser, E. J. (2018). Systematic review of pain medicine content, teaching, and assessment in medical school curricula internationally. *Pain and Therapy, 7*(2), 139-161. doi: 10.1007/s40122-018-0103-z
- Simpkin, A. L., & Schwartzstein, R. M. (2016). Tolerating uncertainty - The next medical revolution? *The New England Journal of Medicine, 375*(18), 1713-1715. doi: 10.1056/NEJMp1606402
- Smith, J. K. (2008). Relativism. In L. M. Given (Ed.), *The SAGE Encyclopedia of Qualitative Research Methods* (Vol. 1). Thousand Oaks, CA: SAGE Publications, Inc.
- Spencer, L., Ritchie, J., Lewis, J., & Dillon, L. (2003). *Quality in qualitative evaluation: A framework for assessing research evidence*. National Centre for Social Research
- Tong, A., Sainsbury, P., & Craig, J. (2007). Consolidated criteria for reporting qualitative research (COREQ): A 32-item checklist for interviews and focus groups. *International Journal for Quality in Health Care, 19*(6), 349-357. doi:10.1093/intqhc/mzm042
- Vasileiou, K., Barnett, J., Thorpe, S., & Young, T. (2018). Characterising and justifying sample size sufficiency in interview-based studies: Systematic analysis of qualitative health research over a 15-year period. *BMC Medical Research Methodology, 18*(1), 148. doi:10.1186/s12874-018-0594-7

Table 1. Sociodemographic Characteristics of the Sample

Physician	Gender	Number of Years qualified as a physician	Clinical Setting
1	Woman	28	Rheumatology
2	Woman	25	Chronic pain
3	Man	14	General pediatrics
4	Woman	17	General pediatrics
5	Man	23	Epilepsy
6	Woman	17	General pediatrics
7	Woman	17	Diabetes and metabolic disorders
8	Woman	15	Rheumatology
9	Woman	24	General pediatrics, neonates
10	Woman	16	General pediatrics
11	Woman	19	General pediatrics
12	Woman	19	General pediatrics
13	Woman	15	Community
14	Woman	24	Pain
15	Man	24	General Pediatrics
16	Woman	26	General Pediatrics and pediatric Neurology

Table 2. Semi-Structured Interview Schedule

Question number	Question and prompts
1	<p>Can you tell us a bit about any training you have received about treating individuals with chronic pain?</p> <p>Prompts:</p> <ol style="list-style-type: none"> a. What about pediatric pain specifically?
2	<p>Can you please tell us about your experiences of treating children with chronic pain?</p> <p>Prompts:</p> <ol style="list-style-type: none"> a. What kinds of patients have you treated? b. Can you walk us through the process of seeing a child with chronic pain and providing a diagnosis? c. How have these patients and their families managed the diagnosis and treatment?
3	<p>Can you please tell us about what happens once a child with chronic pain arrives in your clinic at an initial assessment appointment?</p> <p>Prompts:</p> <ol style="list-style-type: none"> a. Can you tell us about any referrals that you might make at this point? b. Can you tell us about any tests that you might administer?
4	<p>How do you sit with uncertainty regarding diagnosing children with idiopathic chronic pain?</p> <p>Prompts:</p> <ol style="list-style-type: none"> a. What is it like to diagnose a child with chronic pain in the absence of a test result? b. How does this experience fit with your training and understanding of what it means to be a physician?
5	<p>How do you explain chronic pain to pediatric patients and their parents?</p> <p>Prompts:</p> <ol style="list-style-type: none"> a. Why do you choose to explain pain in that manner?
6	<p>What happens when you provide the diagnosis and it is not believed or accepted by children and/or parents?</p> <p>Prompts:</p> <ol style="list-style-type: none"> a. How do you negotiate the ‘disagreement’?
7	<p>Is there anything else that we have not talked about that you think is important for us to understand about clinician uncertainty in pediatric chronic pain?</p>