

Appraisals of Pain in Pediatric Inflammatory Bowel Disease: A Qualitative Study With Youth, Parents, and Providers

Irina Gorbounova, MD,^{*}  Tanera R. van Diggelen, BS,[†] Katherine Slack, MEd,[‡] ,
Lexa K. Murphy, PhD,[§] and Tonya M. Palermo, PhD[¶]

^{*}Department of Pediatric Gastroenterology, Hasbro Children's Hospital, Brown University, Providence, Rhode Island, USA

[†]PGSP-Stanford PsyD Consortium, Palo Alto, California, USA

[‡]WSU's Elson S. Floyd College of Medicine, Spokane, Washington, USA

[§]Department of Psychology, Seattle University, Seattle, Washington, USA

[¶]Center for Child Health, Behavior and Development, Seattle Children's Research Institute, Department of Anesthesiology & Pain Medicine, University of Washington, Seattle, Washington, USA

Address correspondence to: Irina Gorbounova, MD, 593 Eddy Street, LL Providence, RI 02903, USA (igorbounova@lifespan.org).

Background: Pain is a predominant symptom of inflammatory bowel disease (IBD), and is influenced by cognitive, emotional, and behavioral factors. The cognitive-affective model of symptom appraisal (CAMSA) has been used to understand how youth view symptoms in chronic conditions. We sought to (1) determine how youth with IBD and their parents appraise pain, and how their perspectives fit within CAMSA, and (2) explore health care providers' understanding and communication about pain.

Methods: Participants included 19 youth ages 10–17 years with chronic IBD pain and their parents, and 5 IBD providers from a gastroenterology clinic. Separate semi-structured qualitative interviews with youth, parents, and providers were conducted. Interview prompts were adapted from CAMSA, previous studies of pediatric pain and symptom monitoring, and a qualitative study in adults with IBD pain. Interviews were analyzed according to principles of reflexive thematic analysis.

Results: Three key components of CAMSA (IBD Threat, Fear/Worry, and Biased Attending) were identified in youth and parent dyads. Some youth showed Biased Attending, including difficulty disengaging, while other youth simply monitored pain. The overarching theme for provider interviews was Gastroenterologists view pain as a secondary (rather than primary) treatment issue.

Conclusions: CAMSA is potentially applicable to pain appraisal in youth with IBD and their parents. When health care providers communicate about pain, they should consider how symptom uncertainty may be influenced by threat, fear/worry, and biased attending. Further studies are needed to develop and test psychosocial interventions to reduce fear and threat of pain in youth with IBD in partnership with families and providers.

Lay Summary

This qualitative study investigated monitoring and interpretation of pain among youth with inflammatory bowel disease (IBD), parents, and providers. Parents and youth demonstrated fear and worry with pain attributable to IBD, highlighting pain appraisal as an opportunity to mediate IBD-related pain.

Key Words: IBD, pain, qualitative, psychosocial

Introduction

Inflammatory bowel disease (IBD) is a chronic immune-mediated condition characterized by an unpredictable relapsing and remitting course and significant disease burden.¹ Chronic abdominal pain is a predominant symptom, with over half of youth reporting abdominal pain and associated disability during clinical remission.^{2,3} Abdominal pain severity and pain catastrophizing have been found to negatively affect quality of life and functional disability in children and youth with IBD.^{3,4} The etiology of abdominal pain in IBD is poorly understood, but likely involves the interplay of peripheral and central nervous system sensitization and biopsychosocial factors^{5,6} such as mood disorders,^{7,8} female sex,⁹ parental and family functioning,¹⁰ and coping style.¹¹ Adults with IBD report uncertainty in interpreting and managing painful

sensations, leading to anxiety and frustration¹²; however, little is known about how youth with IBD interpret and manage pain. This is important, as youth's pain experience is affected by individual factors, parent-child dyadic factors (such as parent protective responses,⁸ parent pain catastrophizing¹³) and family factors.^{14,15}

Psychosocial factors have emerged as key drivers of pain and associated disability, providing opportunity for intervention in IBD-related chronic pain management by targeting depression, coping techniques, and parental responses to pain.^{5,6,16} A recent systematic review revealed promising evidence for psychological treatments such as stress management interventions and Cognitive Behavioral Therapy for pain management in adults with IBD.¹⁷ Acceptance and Commitment Therapy for adults with IBD has been shown

Received for publication: August 15, 2022. Editorial Decision: October 31, 2022

© The Author(s) 2022. Published by Oxford University Press on behalf of Crohn's & Colitis Foundation.

This is an Open Access article distributed under the terms of the Creative Commons Attribution-Non-Commercial License (<https://creativecommons.org/licenses/by-nc/4.0/>), which permits non-commercial re-use, distribution, and reproduction in any medium, provided the original work is properly cited. For commercial re-use, please contact journals.permissions@oup.com

to reduce stress and improve functioning^{18,19}; and mindfulness intervention has been shown to improve quality of life and psychological distress.^{20,21} There is emerging research on cognitive-behavioral therapy approaches in pediatric IBD, which has been shown to improve child functioning and quality of life,^{22,23} but a limited evidence base to date. Psychological therapies aim to alter the individual's perception of pain, to increase adaptive pain-coping skills, and to increase confidence in pain management. To optimize psychosocial pain management intervention approaches in youth with IBD, further understanding of child and parent experiences with fear and threat of pain may be useful, as has been shown in other pediatric chronic pain conditions.²⁴⁻²⁷

Heathcote and Eccleston's cognitive-affective model of symptom appraisal (CAMSA) describes symptom appraisal in cancer survivorship,²⁸ and can be applied to other painful conditions. There are multiple parallels between cancer and IBD, including a potentially relapsing and remitting course,¹ fear of disease recurrence,¹² heightened risk for chronic pain during remission,^{2,3} and monitoring for pain as a potential symptom of recurrence.²⁹ This model identifies factors that influence how patients monitor and interpret painful symptoms, as well as consequences of interpreting pain as threatening (eg, Fear/Worry, Biased Attending that amplifies pain symptoms). See CAMSA adapted for IBD in Figure 1. In this model, IBD Threat, Fear/Worry, and Biased Attending are key components of the model in monitoring for and interpreting pain. The definitions of key components, modified from Heathcote and Eccleston's model of cancer survivorship,²⁸ is as follows: IBD Threat "*perceiving symptoms and sensations as a threat of IBD,*" Fear/Worry "*symptom uncertainty and anxiety,*" and Biased Attending "*hypervigilance, monitoring, difficulty disengaging, selective attention and interpreting ambiguous information as threatening.*" This model highlights that interpretation of pain as threatening (IBD Threat) can result in anxiety (Fear/Worry) and hypervigilance to pain (Biased Attending), contributing to a cycle of enhanced pain and worry.

Determining whether CAMSA is applicable to youth with IBD can further our understanding of cognitive-affective factors affecting the pain experience, and thus allow for future refinement of more targeted psychosocial interventions for chronic pain in this patient population. Thus, the aims are: (1) to determine how youth and their parents monitor for, interpret, and manage pain, and how their perspectives fit with key components of CAMSA (IBD Threat, Fear/Worry, and Biased Attending), and (2) to explore health care providers' understanding of and communication about pain with patients.

Materials and Methods

Participants and Procedure

Youth and parent participants were recruited from a gastroenterology clinic in a children's hospital in the Pacific Northwest. A purposive sampling technique with maximum variation was applied to recruit patients across different ages, sexes, and sociodemographic backgrounds (race/ethnicity). Inclusion criteria were: (1) age 10–17 years, (2) diagnosed with IBD, and (3) reports chronic pain (ie, weekly pain that has persisted for over 3 months). Exclusion criteria were: (1) significant medical comorbidity (eg, cancer), (2) significant developmental or intellectual disability, and (3) does not speak English. Potential participants were identified through medical chart review and approached via initial study letter, followed by a phone call from study staff.

Fifty-eight potential participants and their families were approached; 23 were unable to be reached, 8 were not eligible (6 no longer had pain, 1 did not speak English, 1 had a significant medical comorbidity), and 7 declined due to lack of time/interest. Twenty patients and their parents met criteria and were enrolled; one later was excluded due to a change in diagnosis (no longer had an IBD diagnosis). IBD providers (physicians, nurse practitioners) were recruited in-person and via email; 5 were approached and all 5 participated. All participants were compensated for their time with gift cards.

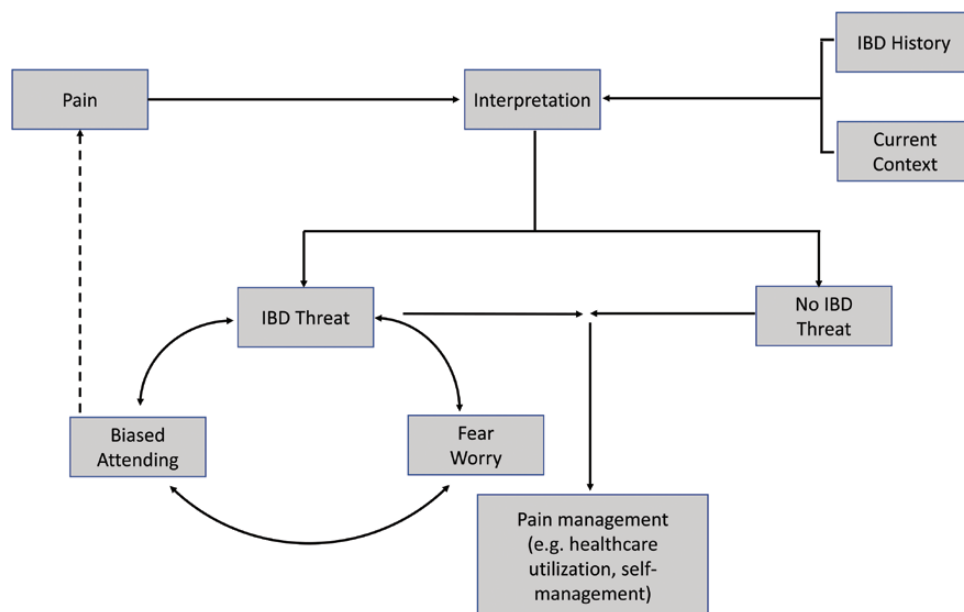


Figure 1. Cognitive-affective model of symptom appraisal,²⁸ adapted for pediatric IBD. Abbreviation: IBD, inflammatory bowel disease.

Participants were interviewed by postdoctoral pain researcher L.K.M. Interviews were conducted either in person or over the phone. The study was approved by the Seattle Children's Institutional Review Board.

Measures

Sociodemographics

Parents reported on their own sex as well as their child's age, race/ethnicity, and sex.

IBD disease characteristics

Parents reported on their child's IBD characteristics including diagnosis types (Crohn's disease, ulcerative colitis, indeterminate colitis), age at diagnosis, and current medications.

Abdominal pain severity

Youth and parents each completed the Abdominal Pain Index (API³⁰). The revised scoring method was used,³⁰ which yields a mean ranging from 0 to 4 based on ratings of pain frequency, duration, and intensity, with higher scores indicating greater abdominal pain severity.

Qualitative interview

All participants were interviewed privately over the phone. Interview prompts were adapted from the CAMSA, previous studies of pediatric pain and symptom monitoring, and a previous qualitative study in adults with IBD and pain.^{12,28,31,32} The youth and parent interview topics included (1) youth's experience with abdominal pain in the context of IBD, (2) youth's/parent's communication with providers about pain and pain management, (3) youth's/parent's experience with monitoring for and interpreting abdominal pain, and (4) youth's pain management (strategies and needs). The provider interview topics included (1) provider's opinion of the typical patient experience with pain in the context of IBD, (2) provider communication about pain and pain management with families of youth with IBD, (3) provider advice and experience with families monitoring for and interpreting abdominal pain in the context of IBD, (4) provider recommendations for pain management, and (5) provider experience with youth with IBD with abdominal pain in remission. The interview question prompts for youth, parents, and providers are available for review in [Supplementary Data Content](#).

Data Analysis

Qualitative interviews were transcribed verbatim and analyzed with the qualitative analysis software NVivo³³ according to principles of reflexive thematic analysis developed by Braun and Clarke.³⁴ This analysis method emphasized flexibility in generating and interpreting patterns within data and is conducted in 6 stages: familiarization, coding, generating initial themes, reviewing themes, defining/naming themes, and writing. Qualitative interviews were coded using principles of deductive and inductive thematic analysis. Components of Heathcote and Eccleston's CAMSA were used to deductively guide coding, while inductive coding methods were used to enrich and modify the model.^{28,34} Because CAMSA is based on patient experiences, the parent and youth interviews were analyzed with a deductive approach. Three predetermined themes of CAMSA were "IBD Threat," "Fear/Worry," and

"Biased Attending." Supporting quotes for each theme were identified in parent and youth interview transcripts. Provider interviews were analyzed with an inductive approach to allow new themes to be identified and explore how provider communication about pain potentially contributes to parental and youth perspectives.

In the familiarization phase, 2 primary coders (T.R.V. and K.S.) reviewed the collected parent and youth interview transcripts. During the coding phase, T.R.V. and K.S. again reviewed all transcripts independently and began to identify potential codes, which were reviewed by I.G. and T.M.P. to ensure reliability. Codes were then reviewed iteratively for generalizability. Following the coding phase, T.R.V., K.S., I.G., and T.M.P. met to review codes and discuss initial themes with a deductive approach of the cognitive-affective model. The 3 themes were "IBD Threat," "Fear/Worry" and "Biased Attending." The definition of the themes were taken from Heathcote and Eccleston's CAMSA.²⁸ All authors then reviewed and agreed upon codes that were the best fit for each theme.

Provider interview transcripts were reviewed by 2 primary coders (I.G. and T.R.V.). During the coding phase, I.G. and T.R.V. again reviewed all transcripts independently and began to identify potential codes, which were reviewed by T.M.P. to ensure reliability. Following the coding phase, I.G., T.R.V., and T.M.P. met to review codes and discuss initial themes with an inductive approach. All authors then discussed final definitions and themes prior to writing the results.

Quality of coding was addressed through the use of Braun and Clarke's³⁴ 15-point Checklist of Criteria for Good Thematic Analysis and Yardley's^{35,36} Quality Principles. This includes transcription, coding processes, sensitivity to context, sensitivity to participants' perspectives by allowing participants to "tell the story" of their IBD, commitment and rigor, and transparency and coherence. Regarding professional expertise of the coding team, the primary coders consisted of 2 post-baccalaureate research coordinators (T.R.V. and K.S.) with experience in pediatric research and qualitative analysis, as well as a physician-researcher and fellow in pediatric gastroenterology (I.G.), and a faculty researcher (T.M.P.) with extensive experience conducting observational and treatment research in pediatric pain.

Ethical Considerations

The study was approved by Seattle Children's Institutional Review Board. All participants provided informed consent (parents) and assent (children) at the time of enrollment.

Results

Descriptives

See [Table 1](#) for descriptive information about the sample. Average pain severity was similar across parent-report and adolescent self-report ($M = 1.86$ on the API), indicating mild-moderate severity.

Qualitative Analysis—Youth and Parents

The adapted CAMSA for pediatric IBD shown in [Figure 1](#) was used to generate 3 key themes (IBD Threat, Fear/Worry, and Biased Attending). These themes emerged in youth and parent interviews. Some youth showed Biased Attending, with

Table 1. Participant characteristics (N = 19 adolescents and parents, N = 5 providers).

	M (SD) or N (%)	Range
Patient and parent sociodemographics		
Adolescent age (years)	14.21 (2.13)	12–17
Adolescent sex	10 (53%) male; 9 (47%) female	—
Parent sex	1 (5%) male; 18 (95%) female	—
Adolescent race/ethnicity	14 (74%) White 2 (11%) Asian 1 (5%) Black 2 (11%) multiple races 2 (11%) Hispanic/Latinx	—
Provider characteristics	4 (80%) physicians 1 (20%) nurse practitioner	—
Adolescent disease factors		
IBD type	13 (69%) Crohn's disease 5 (26%) ulcerative colitis 1 (5%) ulcerative colitis	—
Age at IBD diagnosis	10.05 (3.55)	3–17
Medication	2 (11%) antispasmodic 8 (42%) biologic 1 (5%) methotrexate 1 (5%) JAK inhibitor 1 (5%) anti-nausea medication 3 (16%) adjuvant (essential oils, tums, supplements)	—
Adolescent pain	Abdominal pain severity—self-report ^a 1.86 (0.89) Abdominal pain severity—parent report ^a 1.86 (0.98)	0.85–3.60 0.00–3.40

Abbreviations: IBD, inflammatory bowel disease; M, mean.

^aAbdominal Pain Index, possible range 0–4.

difficulty disengaging, while other youth simply monitored for pain (Table 2).

Theme 1: IBD Threat

IBD Threat was defined as “*perceiving symptoms and sensations as a threat of IBD.*” As a child with IBD experiences pain, uncertainty arises, and pain demands interpretation of whether it is a sign of IBD Threat or another process. Both parents and youth expressed concerns of IBD Threat (Table 2).

When a child experiences pain, parents described using pain characteristics (intensity and duration) to decide if their child's pain was an IBD Threat—one parent said, “*It's usually both linked with degree and duration.*” Youth described using the pain location, progression, and quality to determine if pain was an IBD Threat or a separate process. Youth used words such as “*sharp,*” “*punchy,*” and “*stabby*” to signify IBD Threat and “*achy*” or “*less intense*” when it was not

an IBD Threat. Youth were concerned if their pain worsened over time, or if the pain was more “*localized*” and “*lower down.*”

Theme 2: Fear/Worry

Fear/Worry was defined as “*symptom uncertainty and anxiety.*” Once pain is identified as an IBD Threat, it can lead to Fear/Worry that in turn cause Biased Attending to pain, and maintenance of Fear/Worry. This process thereby amplifies the pain experience. Fear/Worry was identified in youth and parents (Table 2).

When youth experienced pain, youth and their parents worried that something was seriously wrong. One youth said, “*...I think for them [parents] not knowing what's causing it [pain] is kind of, um—I'm trying to find a good word. It, it worries them, I guess.*” Another parent echoed, “*... it continued, and of course, he [child] was worried. And then we were worried, ... and I was feeling extremely upset and scared, and I felt like my child was going to die.*”

Youth talked about IBD Threat giving rise to anxiety about their daily functioning, such as being able to perform adequately at school, work, or sports (Table 2). Meanwhile, parents talked about their worries about their child's disease progression and worries about future management of IBD when their children are away from home (Table 2).

Theme 3: Biased Attending

Biased Attending was defined as “*hypervigilance, monitoring, difficulty disengaging, selective attention and interpreting ambiguous information as threatening.*”

We found that parents exhibited Biased Attending, while youth showed only partial evidence for Biased Attending (Table 2). Some youth demonstrated Biased Attending, while others expressed monitoring for and paying attention to pain with no difficulty disengaging or hypervigilance.

Parents explained that they try to determine whether their child is in pain by asking them direct questions and by looking at “*facial expressions*” or their “*behavior.*” Other parents paid attention to whether their child was more quiet than usual, and the way their child “*walks*” and “*talks.*”

Youth demonstrated partial evidence for Biased Attending. Some youth expressed hypervigilance and difficulty disengaging—one of the youth said “*And I'm always on the lookout in case it comes up, what's going to be my plan of action,*” while another youth confided “*... and it's hurting, it's very distracting...So unless I find something that I'm very focused into, it's kind of always on my mind.*” Meanwhile, other youth reported monitoring for pain to determine whether there was a “*pattern,*” rather than perseverating about the pain experience, “*So we usually look out for it, and we do try to see if there's any pattern.*” Some youth expressed that they learned to live with pain, and although they monitor their pain, they do not get consumed by it, “*Um, I guess I do monitor it [pain]. At this point, though, I've kind of gotten used to it. So it's kind of hard to tell whether or not I'm having it unless I like focus on it.*” One youth expressed his insight, “*But sometimes if I think too much about it [pain], it like makes it happen.*”

Qualitative Analysis—Providers

The second aim was to explore health care providers' understanding of and communication about pain with youth

Table 2. Parent and youth themes and example quotes based on 3 key aspects (IBD Threat, Fear/Worry, and Biased Attending) of the cognitive-affective model of symptom appraisal (CAMSA).

Theme	Parent quote	Youth quote
IBD Threat	<p>“I think when he says he’s [youth’s] in more pain, he’s definitely probably flared.”</p> <p>“I would say maybe five is when we start to say, ‘Okay, obviously you’re really uncomfortable. We should probably send an email and leave a voicemail.’ And then if it’s climbing even higher than it’s more, ‘Okay. We need to call the on-call or we need to just go ahead—and—if it’s an 8, we’re just going to get in the car and go ahead and go to the emergency room.’”</p> <p>“It’s usually both linked with degree and duration... If the degree and the duration has consistently been extending over a period of days, then that’s our sign for, ‘We need to reach out.’”</p> <p>“...we just go into a whole lot of questionnaires and guesswork about what could have cause it. Why is that there? ‘I hope it’s not a flare-up.’”</p>	<p>“If the pain gets too bad to the point where I can’t get up or like I’m not making it to school regularly, then, uh then we’ll call them [providers] and then we’ll tell them about it.”</p> <p>“It depends on where the pain is located and how long it lasts, because most of my IBD pain, it’s more like low down, and it’s definitely more of a deep-rooted pain...”</p> <p>“Stress and food are usually, like an achy pain where flares are the sharpest pain.”</p> <p>“...the less intense stomach pain is hunger pain.... And pain it’s usually punchy feeling. And the medium level stabby and the high level punchy are Crohn’s.”</p>
Fear/Worry	<p>“Any time she [youth] comes to me, and she says she has bad pain, I worry. I worry about whether or not she’s starting to flare.”</p> <p>“Well, just, ‘Okay. What’s this mean? Is this going to last? You know, do I need to start packing a bag?’ Because I know that if we’re going to the ER, they’re going to admit us. You know, and it’s just I start kind of running through the checklist in my head of everything that needs to be taken care of if this is going to persist or get worse, or whatever it is I need to do. So there’s kind of a constant worry.”</p> <p>“Stress definitely aggravates his [youth’s] pain. Any kind of anxiety... he’s prone to getting stressed very quickly. And so that leads to tightening, I feel, of his stomach, and pain after that.”</p> <p>“[Youth] has a diagnosis of post-traumatic stress disorder, which is an anxiety disorder. And anxiety and stress, they’re like oxygen to fire with something like IBD.”</p> <p>“I worry about her a lot. And she’s getting ready to go to college this next fall, and it terrifies me that she’s going to be there, and I’m always there, and I’m not going to be there”</p> <p>“How much damage? Will you be able to recover? What is your life going to be like going forward with everything, college, and just normal social stuff and all that?”</p>	<p>“I do get worried when I’m in a lot of pain, because it just brings me back to when I was in the flare, and when I was undiagnosed, and then diagnosed and still flaring... if it lasts more than a day or two, then I’ll really start getting worried.”</p> <p>“... kind of panic a little bit in the sense that I’ll think about, ‘If I’m flaring, how is this going to affect if I can play a softball game, if I can work, if I can do all these other things that I can do when I’m in remission.’ So I definitely start to stress about how it’s going to change what I can do.”</p> <p>“I do get worried when I’m in a lot of pain, because it just brings me back to when I was in the flare.”</p> <p>“It can be scary. Um, when I get the pain—because I’m always so scared that I’ll start seeing blood again because I don’t, I don’t know if it’s normal and if my medication isn’t working anymore. And it’s just all kind of not, not clear”</p> <p>“I definitely have to calm myself down. I’m a person who, when I feel pain, I want to shelter that and guard it, make sure that nothing worse happens to it. And so sometimes I can get a little overwhelmed by smaller pain by thinking it’s bigger.”</p> <p>“... I would not be able to go to work and that really took a toll on like, my performance [laughter]. So I used to think about it [pain] like every single day.”</p> <p>“Because I know one of my big triggers is stress. And so when I get stressed out and anxious, I start getting really sick. And I will actually throw up and be in a lot of pain.”</p> <p>“I think that every time I get pain again. I’m so worried that my medication has stopped working and I, you know, it’s back.”</p>
Biased Attending	<p>“And I kind of look out for signs, any kind of discomfort on his face, and I ask him, ‘Is your tummy hurting?’ ... ‘Hey. How are you feeling? Are you uncomfortable?’”</p> <p>“I will say, ‘How’d you feel today?’ ... especially if I can tell. Like, if he’s super quiet, I’ll say, ‘How was your day?’ You know, ‘Did your stomach hurt?’ So, yeah, we do, I do often ask him about that.”</p> <p>“I don’t want it to be the only thing that we discuss... And I think immediately after he was diagnosed, and for several months, it kind of was. I mean, it kind of took over.”</p> <p>“It’s been such a rollercoaster ride... we really haven’t reached remission. So I guess I don’t feel like the guard’s been let down entirely yet. So we’re constantly in a state of watching for things.”</p> <p>“...but we also try to not make it [pain] what every day is about”</p>	<p>“Oh, it never leaves. It’s always in the background. It’s always there. It’s just a consistent part of my life now... And I’m always on the lookout in case it comes up, what’s going to be my plan of action.”</p> <p>“When I’m at school or something like that, and it’s hurting, it’s very distracting...So unless I find something that I’m very focused into, it’s kind of always on my mind.”</p> <p>“So we usually look out for it, and we do try to see if there’s any pattern”</p> <p>“Um, I guess I do monitor it [pain]. At this point, though, I’ve kind of gotten used to it. So it’s kind of hard to tell whether or not I’m having it unless I like focus on it”</p>

Three key components of CAMSA were identified in youth and parent dyads. Youth showed partial evidence for Biased Attending. Abbreviation: IBD, inflammatory bowel disease.

Table 3. Health care provider subthemes and example quotes.

Subtheme	Provider quote
<p><i>Providers assume pain indicates inflammation and will resolve with treatment</i></p> <p>Providers evaluate for inflammation by performing diagnostic studies before attributing pain to non-IBD causes</p>	<p>“So as long as the inflammation’s active, I will default most, if not all pain, to it until I get the inflammation under control”</p> <p>“So if they’re [youth] having pain and there’s active inflammation, there’s no sense in focusing on, ‘Here’s what’s bothering them,’ at that point. You’ve got to focus on the inflammation first.”</p> <p>“And so if somebody’s got pain, umm you’ve got to evaluate, and you have to find objective evidence of disease activity that says, <i>aha</i>, this is truly pain, and that can be as simple as a blood test, as simple as a stool test, or you know again, a scope or endoscopy.”</p> <p>“I’m usually operating under the assumption that the pain will go away after the inflammation is better. So I will sort of responsively—if this appears to be a problem, then I will begin that conversation. And to be honest, the vast majority of my patients get better.”</p> <p>“Fortunately, once we see them in two to four weeks with treating the inflammation, the pain significantly subsides, and that’s the case for the overwhelming majority”</p> <p>“I would imagine that most individuals’ abdominal pain umm becomes significantly less, umm and it resolves <i>uhh</i> in the majority of patients that <i>uh</i> that I see, once we get them onto appropriate therapy.”</p>
<p><i>Providers are confident they can differentiate IBD and non-IBD pain</i></p> <p>Providers encourage patients to contact them about ongoing symptoms and pain, as they can help determine if pain is due to IBD</p>	<p>“I would say, ‘If you have symptoms and you can’t explain those symptoms by something you ate or something you were exposed to, then we need to hear from you, and we can certainly help you—we can certainly help you tease out the cause.’”</p> <p>“I don’t want somebody to over- or under-analyze their symptoms... That’s the role of the doctor to really be able to say, you know ‘You’re having these symptoms right now, but this is not Crohn’s disease. This is the fact that you drink too liters of soda pop,’ or, ‘You’re constipated.’”</p> <p>“Oftentimes, if that’s different than what they have been experiencing before, oftentimes, they will that will be something we want to know, and then we can assess whether there’s something new going on.”</p>
<p><i>Patients should monitor for a bundle of symptoms</i></p> <p>Providers do not explicitly ask patients to monitor for pain</p>	<p>“I don’t know if I over-emphasize monitoring for pain. I would say monitor for a bundle of these symptoms and then monitor for any symptom you cannot explain with common things that would cause pain for everyone”</p> <p>“Well, we don’t really talk too much about the abdominal pain objectively. We sort of talk about sort of the constellation of symptoms, so sort of diarrheal symptoms, the difficulty eating, weight loss, elevated inflammatory markers. Abdominal pain is part of that, but we usually don’t focus in on the abdominal pain portion of things”</p> <p>“I mean, I think I use it in the context of, ‘Okay, if you’re having increasing symptoms, and that’s one of the symptoms, then to call us.’ But I don’t have them like keep a pain diary.”</p> <p>“I -I don’t tell people to monitor for pain per se... if an individual has symptoms you know such as pain or something else, that you know they should... let us know to see if we need to do something about that”</p>

The overarching theme was “Gastroenterologists view pain as a secondary issue.” Abbreviation: IBD, inflammatory bowel disease.

with IBD and their parents. The overarching theme was, *Gastroenterologists view pain as a secondary treatment issue*. IBD providers expressed their concern about assessing for and treating inflammation when patients report pain. Three subthemes emerged that are described below (see [Table 3](#) for example quotes).

The first subtheme was *Providers assume pain indicates inflammation and will resolve with treatment*. Providers evaluate for inflammation by performing diagnostic studies before attributing pain to non-IBD causes. Providers report that pain is a sign of inflammation and will improve after attaining remission. Providers did not comment on discussing chronic pain as a primary diagnosis in our interviews.

The second subtheme was *Providers are confident that they can differentiate IBD and non-IBD pain*. Providers encourage patients to contact them about ongoing symptoms and pain, as they can help figure out if pain is due to IBD.

The third subtheme was *Patients should monitor for a bundle of symptoms*. Providers encourage their patients to watch out for several symptoms such as diarrhea, blood in the stool, and abdominal pain; yet they do not explicitly ask patients to actively monitor for pain.

Discussion

This study examined the experience of youth with IBD-related chronic pain through semi-structured qualitative interviews of youth, their parents, and health care providers. We found that many elements of Heathcote and Eccleston’s CAMSA model²⁸ were relevant in youth with IBD-related chronic pain. Three key components of the model (IBD Threat, Fear/Worry, and Biased Attending) were identified in our patient sample, similar to survivors of childhood cancer.^{28,37} Youth showed partial evidence for Biased Attending with some youth exhibiting hypervigilance and difficulty disengaging, while others simply monitor for pain without perseverating on the pain experience.

In interviews with health care providers, we found that providers viewed pain as a secondary treatment issue and primarily focused on inflammation when a patient is in pain, with the expectation that pain will improve once inflammation is under control. Provider communication that pain is a sign of inflammation could contribute to youth and parent’s Biased Attending to pain and reinforce the notion that pain is a possible IBD Threat, which would then increase Fear/Worry that pain is a sign of an IBD flare.

Prior qualitative studies have found that children with IBD struggle with the unpredictable nature of the disease,³⁸ and view pain's impact on emotional and physical functioning as a barrier to effective management of pain.¹² Our study adds to these findings as it shows that pain experienced by youth in the context of unpredictable disease may give rise to IBD Threat, Fear/Worry, and further Biased Attending to pain; and communication with families and providers has the potential to alter the experience of pain.

Youth with IBD and abdominal pain are at risk for depression,³⁹ experience a lower quality of life and increased disability independent of disease activity.^{2,40} There is growing evidence that parent- and family-level psychosocial factors are associated with pain and disability in children with IBD.¹⁶ Depressive symptoms in youth and their parents have been found to mediate the relationship between youth quality of life and IBD disease activity,^{10,15,41,42} and parental response toward pain in children with IBD has been shown to predict the intensity of pain.⁸ In our study, we found that parents likely play a key role in youth's experience with IBD pain. Parents described Fear/Worry and Biased Attending to pain, suggesting that there may be opportunities to intervene with parents around their cognitions and behaviors. Future studies may use validated measures of pain-specific cognitive-affective variables that are reflected in the CAMSA model, such as pain catastrophizing and fear of pain.

Prior studies have shown that youth with IBD express significant worry over their gastrointestinal symptoms, including abdominal pain.⁴³ Our study adds to these findings, as youth reported significant Fear/Worry when they experienced abdominal pain. Specifically, youth frequently worried about their ability to function during an IBD flare; pain-related fear is associated with increased disability in other chronic pain populations.⁴⁴

Providers play a pivotal role in the management of IBD inflammation and psychosocial comorbidities. Other studies have demonstrated that communication with providers partially mediates the relationship between gastrointestinal symptoms and gastrointestinal worry.⁴³ To our knowledge, our study is the first to examine provider understanding and communication about pain with youth with IBD and their parents. Provider communication about pain has the potential to indirectly affect youth's experience of pain. In our interviews, we found that providers did not comment on discussing chronic pain as a primary diagnosis, and expressed that pain likely indicates inflammation, and if present after medical workup, will resolve with treatment. This is despite data demonstrating that pain is very common for youth with IBD during clinical remission.^{2,3} This perspective might explain provider inclination to perform investigative studies to rule out inflammation, but may also set the expectation for the family that pain is a sign of inflammation. Our data suggest that the expectation that pain will resolve with the resolution of inflammation may impact the way that youth and families interpret and respond to pain symptoms in the context of IBD.

Our findings highlight the need for multifaceted intervention approaches for youth with IBD-related chronic pain that target symptom appraisals in parents, youth, and providers. Psychological therapies that include parents and adolescents with IBD are limited,⁴⁵ but have been shown to be beneficial in children with other chronic illnesses. Based on our

findings of Biased Attending to pain in parents of youth with IBD; parent cognitions, beliefs, and behaviors toward their child when in pain may be important targets for psychosocial interventions for IBD-related pain. For example, Levy et al found positive benefits for their brief social learning and cognitive-behavioral intervention targeting children with IBD and their parents on improving child functioning and quality of life.²³

Our study examined provider beliefs about pain in youth with IBD, but did not directly observe communication of the provider with the parent-child dyad. Future studies are needed to examine patient-provider communication about pain in the context of IBD to inform provider interventions. Interventions to increase gastroenterologist comfort in discussing chronic pain as a primary diagnosis in youth with IBD and providing patient and family education about pain may be relevant targets. Suggestions on how to approach pediatric chronic pain discussion and education as a health care provider was outlined in narrative review by Koechlin et al,⁴⁶ and could be applied to pediatric IBD. Studies in other pediatric chronic pain conditions reported that health care provider's plausible treatment narrative and positive patient-health care provider relationship can have a positive impact on the pain experience.⁴⁶ Future studies should examine whether early education of youth and parents on chronic pain decreases fear and worry and biased attending in youth with IBD-related chronic pain and their parents; and examine the impact of plausible treatment narrative on daily function and quality of life in youth with IBD-related chronic pain.

The findings of our study should be interpreted in light of several limitations including the limited racial diversity in the sample, small sample size, lack of comparison with healthy controls or those with other chronic pain conditions, and the imbalance of mothers participating in the study. Thus, findings may not be representative of more diverse populations and may not reflect fathers' experiences with youth with IBD-related chronic pain.

Conclusion

This qualitative study was the first to examine the experience of and communication about pain in 3 stakeholder groups (youth with IBD, parents, and providers). The CAMSA model is potentially applicable to pain appraisal in youth with IBD and their parents. Gastroenterologists view pain as a secondary treatment issue, and their communication about pain may contribute to symptom uncertainty. Further studies are needed to develop and test multifaceted intervention approaches for youth with IBD-related chronic pain that target symptom appraisals in partnership with parents and providers.

Supplementary Data

Supplementary data is available at *Crohn's and Colitis* 360 online.

Funding

Irina Gorbounova was supported by NIH postdoctoral training award (5T32DK 007742-24) and Lexa K. Murphy

was supported by the Seattle Children's Hospital Hearst Fellowship grant.

Acknowledgments

We gratefully acknowledge the participation of the children, parents, and providers who participated in the study. We also thank Lauren Heathcote, PhD for her comments on our interview scripts.

Authors' Contributions

I.G. participated in qualitative analysis, drafted and revised the manuscript, and approved the final draft as submitted. T.R.D. and K.S. participated in qualitative analysis and revised the manuscript, and approved the final draft as submitted. L.K.M. conceptualized the study, drafted and revised the manuscript, and approved the final draft as submitted. T.M.P. supervised the study, participated in qualitative analysis, and revised the manuscript, and approved the final draft as submitted.

Conflicts of Interest

All of the authors are responsible for the drafting or revision of the manuscript and have approved the manuscript as submitted. All authors agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. Authors have no financial disclosures or conflicts of interest to report.

Data Availability

Transcribed interviews are available upon request.

References

- Rosen MJ, Dhawan A, Saeed SA. Inflammatory bowel disease in children and adolescents. *JAMA Pediatr.* 2015;169(11):1053–1060.
- Greenley RN, Kunz JH, Schurman JV, Swanson E. Abdominal pain and health related quality of life in pediatric inflammatory bowel disease. *J Pediatr Psychol.* 2013;38(1):63–71.
- Wojtowicz AA, Greenley RN, Gumidyal AP, Rosen A, Williams SE. Pain severity and pain catastrophizing predict functional disability in youth with inflammatory bowel disease. *J Crohns Colitis.* 2014;8(9):1118–1124.
- De Carlo C, Bramuzzo M, Canaletti C, et al. The role of distress and pain catastrophizing on the health-related quality of life of children with inflammatory bowel disease. *J Pediatr Gastroenterol Nutr.* 2019;69(4):e99–e104.
- Bielefeldt K, Davis B, Binion DG. Pain and inflammatory bowel disease. *Inflamm Bowel Dis.* 2009;15(5):778–788.
- Srinath AI, Walter C, Newara MC, Szigethy EM. Pain management in patients with inflammatory bowel disease: insights for the clinician. *Therap Adv Gastroenterol.* 2012;5(5):339–357.
- Srinath AI, Goyal A, Zimmerman LA, et al. Predictors of abdominal pain in depressed pediatric inflammatory bowel disease patients. *Inflamm Bowel Dis.* 2014;20(8):1329–1340.
- Murphy LK, Rights JD, Ricciuto A, Church PC, Ahola Kohut S. Biopsychosocial correlates of presence and intensity of pain in adolescents with inflammatory bowel disease. *Front Pediatr.* 2020;8:559.
- Murphy LK, Suskind DL, Qu P, et al. Abdominal pain after pediatric inflammatory bowel disease diagnosis: results from the ImproveCareNow network. *J Pediatr Gastroenterol Nutr.* 2020;71(6):749–754.
- Caes L, Chambers CT, Otley A, Stinson J. Pain and quality of life in youth with inflammatory bowel disease: the role of parent and youth perspectives on family functioning. *Pain Rep.* 2019;4(2):e715.
- van Tilburg MA, Claar RL, Romano JM, et al. Role of coping with symptoms in depression and disability: comparison between inflammatory bowel disease and abdominal pain. *J Pediatr Gastroenterol Nutr.* 2015;61(4):431–436.
- Sweeney L, Moss-Morris R, Czuber-Dochan W, Belotti L, Kabeli Z, Norton C. 'It's about willpower in the end. You've got to keep going': a qualitative study exploring the experience of pain in inflammatory bowel disease. *Br J Pain.* 2019;13(4):201–213.
- Langer SL, Romano JM, Mancl L, Levy RL. Parental catastrophizing partially mediates the association between parent-reported child pain behavior and parental protective responses. *Pain Res Treat.* 2014;2014:751097.
- Palermo TM, Chambers CT. Parent and family factors in pediatric chronic pain and disability: an integrative approach. *Pain.* 2005;119(1–3):1–4.
- Reed-Knight B, van Tilburg MAL, Levy RL, et al. Maladaptive coping and depressive symptoms partially explain the association between family stress and pain-related distress in youth with IBD. *J Pediatr Psychol.* 2018;43(1):94–103.
- Murphy LK, de la Vega R, Kohut SA, et al. Systematic review: psychosocial correlates of pain in pediatric inflammatory bowel disease. *Inflamm Bowel Dis.* 2021;27(5):697–710.
- Norton C, Czuber-Dochan W, Artom M, Sweeney L, Hart A. Systematic review: interventions for abdominal pain management in inflammatory bowel disease. *Aliment Pharmacol Ther.* 2017;46(2):115–125.
- Wynne B, McHugh L, Gao W, et al. Acceptance and commitment therapy reduces psychological stress in patients with inflammatory bowel diseases. *Gastroenterology.* 2019;156(4):935–945.e1.
- Lavelle J, Storan D, Eswara Murthy V, et al. Brief and telehealth acceptance and commitment therapy (ACT) interventions for stress in inflammatory bowel disease (IBD): a series of single case experimental design (SCED) studies. *J Clin Med.* 2022;11(10):2757. doi:10.3390/jcm11102757. PMID: 35628884.
- Neilson K, Ftanou M, Monshat K, et al. A controlled study of a group mindfulness intervention for individuals living with inflammatory bowel disease. *Inflamm Bowel Dis.* 2016;22(3):694–701.
- Schultz M, Atherton I, Watson A. Mindfulness-based cognitive therapy for inflammatory bowel disease patients: findings from an exploratory pilot randomised controlled trial. *Trials.* 2015;16:379–391.
- Stapersma L, van den Brink G, van der Ende J, et al. Psychological outcomes of a cognitive behavioral therapy for youth with inflammatory bowel disease: results of the HAPPY-IBD randomized controlled trial at 6- and 12-month follow-up. *J Clin Psychol Med Settings.* 2020;27(3):490–506.
- Levy RL, van Tilburg MA, Langer SL, et al. Effects of a cognitive behavioral therapy intervention trial to improve disease outcomes in children with inflammatory bowel disease. *Inflamm Bowel Dis.* 2016;22(9):2134–2148.
- Neville A, Griep Y, Palermo TM, et al. A "dyadic dance": pain catastrophizing moderates the daily relationships between parent mood and protective responses and child chronic pain. *Pain.* 2020;161(5):1072–1082.
- Dougherty BL, Zelikovsky N, Miller KS, Rodriguez D, Armstrong SL, Sherry DD. Longitudinal impact of parental catastrophizing on child functional disability in pediatric amplified pain. *J Pediatr Psychol.* 2021;46(4):474–484.
- Miller MM, Wuest D, Williams AE, Scott EL, Trost Z, Hirsh AT. Injustice perceptions about pain: parent-child discordance is associated with worse functional outcomes. *Pain.* 2018;159(6):1083–1089.
- Connelly M, Anthony KK, Sarniak R, Bromberg MH, Gil KM, Schanberg LE. Parent pain responses as predictors of daily activities and mood in children with juvenile idiopathic arthritis: the utility of electronic diaries. *J Pain Symptom Manage.* 2010;39(3):579–590.

28. Heathcote LC, Eccleston C. Pain and cancer survival: a cognitive-affective model of symptom appraisal and the uncertain threat of disease recurrence. *Pain*. 2017;158(7):1187–1191.
29. Szigethy E. Pain management in patients with inflammatory bowel disease. *Gastroenterol Hepatol (N Y)*. 2018;14(1):53–56.
30. Laird KT, Sherman AL, Smith CA, Walker LS. Validation of the abdominal pain index using a revised scoring method. *J Pediatr Psychol*. 2015;40(5):517–525.
31. Jordan AL, Eccleston C, Osborn M. Being a parent of the adolescent with complex chronic pain: an interpretative phenomenological analysis. *Eur J Pain*. 2007;11(1):49–56.
32. Stinson J, White M, Isaac L, et al. Understanding the information and service needs of young adults with chronic pain: perspectives of young adults and their providers. *Clin J Pain*. 2013;29(7):600–612.
33. QSR. *NVivo Qualitative Data Analysis Software for Windows, v. 10*. QSR International Pty Ltd; 2012.
34. Braun V, Clarke V. Using thematic analysis in psychology. *Qual Res Psychol*. 2006;3(2):77–101.
35. Yardley L. Dilemmas in qualitative health research. *Psychol Health*. 2000;15(2):215–228.
36. Yardley L. Demonstrating validity in qualitative psychology. In: Smith JA, eds. *Qualitative Psychology: A Practical Guide to Research Methods*. SAGE Ltd.; 2007:235–51.
37. Heathcote LC, Loecher N, Simon P, et al. Symptom appraisal in uncertainty: a theory-driven thematic analysis with survivors of childhood cancer. *Psychol Health*. 2021;36(10):1182–1199.
38. Easterlin MC, Berdahl CT, Rabizadeh S, et al. Child and family perspectives on adjustment to and coping with pediatric inflammatory bowel disease. *J Pediatr Gastroenterol Nutr*. 2020;71(1):e16–e27.
39. Zimmerman LA, Srinath AI, Goyal A, et al. The overlap of functional abdominal pain in pediatric Crohn's disease. *Inflamm Bowel Dis*. 2013;19(4):826–831.
40. Claar RL, van Tilburg MAL, Abdullah B, et al. Psychological distress and quality of life in pediatric Crohn disease: impact of pain and disease state. *J Pediatr Gastroenterol Nutr*. 2017;65(4):420–424.
41. Reed-Knight B, Lee JL, Greenley RN, Lewis JD, Blount RL. Disease activity does not explain it all: how internalizing symptoms and caregiver depressive symptoms relate to health-related quality of life among youth with inflammatory bowel disease. *Inflamm Bowel Dis*. 2016;22(4):963–967.
42. Bramuzzo M, De Carlo C, Arrigo S, et al. Parental psychological factors and quality of life of children with inflammatory bowel disease. *J Pediatr Gastroenterol Nutr*. 2020;70(2):211–217.
43. Varni JW, Shulman RJ, Self MM, et al. Patient health communication mediating effects between gastrointestinal symptoms and gastrointestinal worry in pediatric inflammatory bowel disease. *Inflamm Bowel Dis*. 2017;23(5):704–711.
44. Asmundson GJ, Noel M, Petter M, Parkerson HA. Pediatric fear-avoidance model of chronic pain: foundation, application and future directions. *Pain Res Manag*. 2012;17(6):397–405.
45. Eccleston C, Fisher E, Law E, et al. Psychological interventions for parents of children and adolescents with chronic illness. *Cochrane Database Syst Rev*. 9660;2015(4):CD009660.
46. Koechlin H, Locher C, Prchal A. Talking to children and families about chronic pain: the importance of pain education—an introduction for pediatricians and other health care providers. *Children (Basel)*. 2020;7(10):179. doi:10.3390/children7100179. PMID: 33053802.