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The Evolution of Pediatric Chronic Disease Treatment Decisions: A Qualitative, Longitudinal View of Parents' Decision-Making Process

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Abstract

Background—In the context of pediatric chronic conditions, patients and families are called upon repeatedly to make treatment decisions. However, little is known about how their decision making evolves over time.

Objective—To understand parents' process for treatment decision making in pediatric chronic conditions.

Methods—We conducted a qualitative, prospective longitudinal study using recorded clinic visits and individual interviews. After obtaining consent from healthcare providers, parents and patients, clinic visits where treatment decisions were expected to be discussed were video-recorded. Parents then participated in sequential telephone interviews about their decision-making experience. Data were coded by two people and analyzed using framework analysis with sequential, time-ordered matrices.

Results—21 families, including 29 parents, participated in video-recording and interviews. We found three dominant patterns of decision evolution. Each consisted of a series of decision events, including conversations, disease flares and researching of treatment options. Within all three patterns there were both constant and evolving elements of decision making, such as role perceptions and treatment expectations respectively. After parents made a treatment decision, they immediately turned to the next decision related to the chronic condition, creating an iterative cycle.

Conclusion—In this study, decision making was an iterative process occurring in three distinct patterns. Understanding these patterns and the varying elements of parents' decision processes is an essential step towards developing interventions that are appropriate to the setting and capitalize on the skills families may develop as they gain experience with a chronic condition. Future research should also consider the role of children and adolescents in this decision process.

INTRODUCTION

In living with chronic conditions, patients and families are called upon repeatedly to make treatment decisions.(1-5) In contrast, most medical decision making research focuses on decisions that are more discrete and time-bound, such as a screening test, surgical procedure or vaccination. This is particularly true for trials of decision aids(6) but also applies to many observational and qualitative studies. Such research develops a picture of decision making at a single point in time. It may serve as a building block for research about decisions in chronic conditions but does not adequately address the longitudinal nature of such decisions.

This longitudinal nature has led to decision making in chronic conditions being framed as a process rather than a discrete event.(1, 3, 7, 8) Both the ever-changing nature of chronic conditions and the evolving relationships between patients and healthcare providers likely contribute to the decision process. Most studies examining the decision process in chronic conditions have used retrospective methods.(1-3, 5, 9, 10) From such work we know that patients and families report having conversations with numerous individuals as part of the process. We also know that even after a decision is “made” individuals continue to think about the treatment options. A few studies have sought to examine the process longitudinally. One used a national dataset and found that parents of children with special health care needs report changes over time in the degree of shared decision making.(11) Others have conducted qualitative interviews at multiple time points and then analyzed the data as recurrent cross-sections.(12, 13) In other words, although the participants may be the same at each time point, there is little attempt to understand individual participants’ disease and decision trajectories.

In order to develop interventions designed to improve decision making for chronic conditions, a more nuanced understanding of the key decision events and processes is needed. To achieve this goal, we focused on a particular treatment decision. We sought to investigate prospectively families’ decisions about treatment with biologic therapies, specifically tumor necrosis factor- α inhibitors, for their children with inflammatory bowel disease (IBD) or juvenile idiopathic arthritis (JIA). These relatively common pediatric chronic conditions can lead to debilitating symptoms, need for surgery, and/or disability. Although biologics have been shown to be effective for each condition, there are significant risks and uncertainty associated with the treatment.(14-19) The limited long-term pediatric data and potential side-effects of biologics, such as immune suppression, risk for infections and possible increased risk of late-onset lymphoma, contribute to parents’ struggle with this decision.(2, 20) The complex trade-offs that characterize the decision about biologics make it a well-suited model for understanding the decision-making process in pediatric chronic conditions.

METHODS

Sample

Physicians and one nurse practitioner, hereafter referred to as providers, who treat patients with either JIA or IBD were recruited from the rheumatology and gastroenterology clinics of

a large academic children's hospital. All approached providers, except one, agreed to participate and written consent was obtained.

Eligible families were those who were English-speaking, their child had JIA or IBD, they had a clinic appointment scheduled with a consented provider and the provider anticipated discussing biologic treatment initiation at that visit. At the time of the clinic appointment, written consent and assent (for children age 8-17) was obtained from anyone who would be in the room during the visit. We recruited families until we reached informational saturation for each disease group,(21) defined, for this study, as the point at which two consecutive visits revealed no new approaches to discussing treatment decisions. Families who discussed biologic treatment initiation during the recorded visit (21 out of 30) then participated in sequential telephone interviews.

Families were compensated \$30 for video-recording of the clinic visit and \$10 per telephone interview. Providers received no compensation. Cincinnati Children's Hospital Medical Center's Institutional Review Board approved this study.

Data Collection

Recorded Visits—As fully described elsewhere,(22) we collected clinic observation data via video-recording with audio back-up. A camera was set-up in the exam room and recording continued until the provider left the room. Following the visit, the audio was professionally transcribed and non-verbal aspects of the video added to the transcripts by study staff. These non-verbal aspects included visible emotions (such as tears), individuals entering and leaving the room, and other behaviors (such as texting).

Follow-up Interviews—We arranged follow-up interviews with any parent who was at the recorded visit where starting treatment with biologics was discussed. Adolescent patients (those over age 11) were also interviewed; however this analysis focuses solely on parents. Interviews were conducted by a trained interviewer approximately 2 weeks after the recorded clinic visit, with a second interview 4 weeks later. For families (n=3) who had not made a treatment decision at the time of the second interview, we conducted a third interview 4 weeks later (10 weeks after the recorded clinic visit). Interviews were semi-structured using an interview guide (see Appendix) developed based on parents' descriptions of decision-making processes from our retrospective studies of decision making about biologics.(2, 20, 23) The interview guide included questions about the recorded visit, decisions being made, prior decision-making conversations, and the decision process, including timing, influencing factors and participating individuals. Interviews were professionally transcribed and the transcription verified by a study staff member.(24)

Data Coding and Analysis

Our analysis used a framework approach(25) combined with sequential time-ordered matrices.(26) Following the established steps of framework analysis, we familiarized ourselves with the data and then developed a thematic framework (see table 1) based on the data, as well as prior research related to decision making about biologics treatment,(2, 20,

27) and decision making in other pediatric chronic conditions.(1, 3, 28) The framework was modified slightly based on themes that emerged in analyzing the clinic recordings.

We used this framework to index and chart the data in sequential matrices. In the first set of matrices (figure 1), each family was charted in a matrix with framework elements along the y-axis and time points (clinic visit and each interview) along the x-axis. Different fonts were used to distinguish individual family members. Once a family's matrix was complete, the data were re-coded in a second matrix (figure 2) focused on the longitudinal aspects of the decision-making process. Again framework elements were placed on the y-axis but in this matrix the x-axis consisted of family units, with retention of different fonts for each family member. The codes in this matrix focused on how aspects of the decision-making process did or did not change over time. In addition to noting ideas that were present at all time points in matrix 1, but may have changed, we also took note of codes that were present at only some time points.(29) The data in this second matrix were then analyzed with a focus on variations between and within families.

In addition to this framework approach, we created a decision timeline for each family that indicated the order of events related to decision making and the time over which they occurred. These timelines were then compared between families.

Two individuals, with prior qualitative research experience and training, coded all data with disagreements resolved through discussion.(30, 31) Due to the need for sequential matrices, data were coded by hand rather than by using data software tools. The primary analysis of the recorded clinical encounter is reported elsewhere.(22)

RESULTS

Overview

In total 21 families, including 29 parents, and 10 providers participated. (Table 2) All patients were white and non-Hispanic, reflecting the general make-up of the clinics from which families were recruited. Results are organized to highlight the evolving nature of treatment decision making, including patterns of decision evolution and descriptions of the ways in which decision-making elements evolve or remain constant over time. Illustrative quotations are presented in table 3.

Decision Evolution

For all participating families, the treatment decision evolved over time, though the time over which it evolved varied from weeks to years, according to participants' descriptions of their experience. During this time numerous decision-making events occurred. Such events included conversations with healthcare providers and other family members; researching treatment options; experiencing disease flares and related medical testing; and seeking insurance approval. The specific decision-making events varied between families, especially with regard to the order of events.

The type of event that occurred most often was conversation about the treatment options. Participants' first awareness of biologics as a treatment option commonly occurred around

the time of diagnosis, during a conversation with members of their child's healthcare team or from parents' own research. However, many parents recalled having thought, at that early time point, that such treatment was not for their child or might be an option "down the line" rather than now. For other families, early conversations mentioned biologics in passing, as part of a larger conversation about treatment options. Most parents reported that, prior to the visit recorded for this study, at least one conversation about biologics as a treatment option had occurred with a healthcare provider.

In reviewing the timeline for each participant's decision-making process, we found that these conversations and the other decision events consistently occurred as part of a process that followed one of three patterns. The first is a slow, steady progression in which families are aware of the choices and, over time, progress to a decision about biologics. The second pattern is more step-wise than the first. In this, the family members perceived the decision about biologics as the next step in a series of treatment decisions. These families articulated having understood, from an early point in the patient's disease course, that there were established, ordered treatment steps. The final pattern is one of abrupt decision making. Here an acute event, such as a disease flare, leads to a rapid decision regarding biologics. In all 3 patterns, after a treatment decision is made, family members nearly immediately began considering the next decision, such as when another medication could be discontinued.

Constant Elements of Decision Making

As decisions evolved, individuals' roles and perceptions of others' roles in decision making remained constant. Most participants described the same people as participating in decision-making, in the same manner, in each sequential interview. For example one mother described the physician's role as providing information, hers as weighing that information. Another mother (#1) said that she and her family "were at a point that [they were] just going to do whatever [the doctor] thought was in the best interest of my child." Additionally, when more than one parent participated in the study, each member of the pair gave a similar description of the decision participants' roles. Although most individuals' role perceptions were constant, two families did have changes in the perception of who made the final decision. In one the mother initially stated that she made the final decision but in later interviews stated that the doctor made the decision. In the other, in the first interview the mother described making the decision together with the doctor, but in a later interview stated that she made the decision alone.

Although specifics varied both within and between families, another constant was parents' descriptions of a prominent role for internal processing throughout the decision-making process. For example, many parents mentioned thinking about the options. Others talked about "deciding what's best" or "coming around to a decision."

Evolving Elements of Decision Making

Other aspects of the decision process, such as expectations regarding treatment with biologics, changed over time. Commonly, when participants had chosen to start biologics, their expectations initially focused on the anticipated improvement in symptoms. However,

once symptoms improved, participants expressed new expectations, often related to limiting side-effects or decreasing other medications.

For most participants the decision about biologics was characterized by a combination of worry and hope that fluctuated over the course of the process. Parents expressed that sometimes they felt deep worry about side effects and other times felt more hopeful that the new treatment would be effective. For some parents, after making the treatment decision, they experienced a sense of relief, especially when their child's symptoms improved and the child had not yet experienced any side effects. Others, even if their child's symptoms improved, seemed to experience little relief and began to worry whether they made the right decision, when side-effects would appear, or how long the treatment would continue to work. Within families, the emotional changes over time tended to be similar. For example, if one parent continued to worry after the decision was made, the other parent often did as well. In a few cases, one parent would refer to the others' emotions rather than describing their own, saying something like, "my wife was concerned." This cycle of worry, decision making and relief, often followed by worrying about the next decision overlaid the general patterns of decision evolution.

Elements of Decision Making with Both Constant and Evolving Components

Many aspects of decision making had both constant and evolving components. During decision making, the options considered by most participants did not change, but the framing of the options changed for some families. For example, one family initially discussed the options as being between two classes of medications but in a follow-up interview described the options as a choice between on-going symptoms or using biologics.

The concrete factors that parents mentioned as influencing their decision-making process changed in emphasis but not content over time. In other words, some factors that were initially minor influences became more influential over time and vice versa. These shifts in emphasis often appeared situational rather than temporal. Such factors included, but were not limited to, data on long-term risks and benefits, disease severity, experiences with other treatments and the patients' other diagnoses. For example, as one patient's symptoms worsened over time, her mother discussed less about side-effects and more about the patient's disease severity as influencing the decision. When more than one parent participated in interviews, they typically mentioned the same influences but may have emphasized them differently.

Similarly, we found that participants' treatment concerns did not change over time, but for many the emphasis placed on such concerns did change. For example, once families made a decision to start treatment with a biologic, many placed less emphasis on previously expressed concerns about treatment. One father (#19), whose child started biologics between the first and second interview, stated in the first interview "...the side effects...Even when I'm at work, I'm thinking about the side effects." Four weeks later he said, "the only concern is I want to know why it isn't working." In this instance the lack of effect from treatment had apparently led to his concern about side effects having less salience. In contrast, in families where the decision was not to start treatment with biologics, the participants continued to emphasize their treatment concerns. In only one family did concerns notably

differ between family members, with the mother concerned about side effects and the father concerned about the treatment's impact on the patient's social life. Nevertheless, this family, who chose not to start treatment, followed the overall pattern of continuing to emphasize their treatment concerns.

Disease specific differences

Our analysis was not designed to compare and contrast families of patients with IBD with those of patients with JIA. During the course of analysis, however, some differences were observed. The first difference related to the pattern of decision evolution, in that abrupt decision making associated with an acute event such as a disease flare, occurred more frequently in IBD than JIA. Also, compared to parents of patients with JIA, parents of patients with IBD more often reported having had multiple conversations with other individuals, such as friends or family members, and having done independent research about treatment options.

As to the process of decision making, nearly all parents of children with JIA reported making a decision either to start or not to start biologics during the recorded visit. In contrast, about half the parents of children with IBD indicated that the decision was made some time after the visit. Similarly, after the recorded visit and within the follow-up period of this study, few parents of children with JIA had additional conversations with a healthcare provider, but nearly half of parents of children with IBD had such conversations.

DISCUSSION

By examining a specific clinical decision, treatment with biologics, and using repeated interviews, we were able to trace families' decision making over time. As suggested by prior retrospective and cross-sectional studies,(1-3, 8) our prospective analysis demonstrated that decision making in chronic conditions is more of a process than an event. Other treatment decisions in pediatric chronic conditions, such as ADHD treatment or gastrostomy feeding, have also been seen to involve a long process of deliberation, although the details have not been well elucidated.(32, 33)

Most models of decision making depict a process with a clear beginning and end,(34-36) however our findings suggest that in many chronic conditions the beginning and end may be less clear, with each little decision leading to the next. By considering decision making in this way (no clear beginning or end), providers and families may be better prepared to discuss decisions as part of a longitudinal process and prepare for the next decision-making opportunity. In turn, this may alleviate some of the recurrent worry that we found families experienced shortly after making a decision.

Although we focused on a specific decision, the three decision-making trajectories we found (slow evolution, step-wise and abrupt decision making) likely occur in other medical decisions as well. For example, they are similar to help-seeking trajectories outlined by Leslie et al related to diagnosis and treatment of attention deficit disorder.(1) We believe our study is the first to map prospectively these trajectories and is an important step towards developing a holistic view of decision making in chronic conditions. Further research in the

area may help to determine whether such decision trajectories are traits associated with individuals and families, consequences of the natural course of the disease, or some combination of the two.

Cross-sectional studies have shown that numerous factors influence treatment decisions and that the factors considered by families may be different than those used by physicians.(2, 3, 20, 37) The finding that some aspects of decision making are constant and others change over time contributes new insight into the complexity of this process and highlights the dynamic nature of decision making in chronic conditions. Some of the changes could be related to changes in disease course, particularly given that Fraenkel et al found that in adult patients with arthritis their perceptions of the treatment options depended partially upon the impact the disease had on their lives.(37) Additionally, the numerous factors parents consider, as well as parents' own descriptions of decision making, suggest a very deliberative process of weighing pros and cons. This may contribute to the lengthy decision process we observed, given that using more explicit heuristics, such as fitting unfamiliar decisions into familiar patterns or using "rules of thumb", may lead to more efficient parental decision making.(38)

Like other studies in pediatrics,(28, 39-42) we found that the patients were perceived as having a relatively small role in decision making, although this analysis does not include interviews with patients themselves. These results are consistent with our prior study of adolescents' roles in decision making about biologics.(23) The participation of children and adolescents in decision making is an area deserving of further research and intervention, particularly as medical decision making is a crucial skill for adolescents to develop as they transition from the pediatric to adult setting.(43, 44)

Although our study was not structured to formally compare families of patients with IBD with those of patients with JIA, some differences emerged. These differences, namely the longer decision process in IBD which involved conversations with more individuals and the more common occurrence of an abrupt decision evolution, may be related to characteristics of the disease or to demographic differences, such as maternal education, between the groups. Our results are not intended to be generalizable beyond the conditions studied; however such disease variation may provide clues into how decision making occurs in other chronic disease treatment decisions. For example, knowing that reaching a treatment decision is taking a long time may portend a future, abrupt decision evolution, particularly in diseases such as IBD that can have relatively sudden, severe flares. Such insight may help in the development of decision support interventions that address variability in the decision process. Moreover, in both IBD and JIA, decision making was an iterative process, suggesting that this may be common in chronic disease decisions. For other chronic disease treatment decisions in which there exists a trade-off between treatment risks and benefits, the process is likely similar.

A key limitation of our study is that participants were asked to recall events and emotions that occurred between the recorded visit and the interviews. We aimed to minimize such recall bias by informing participants of the interview schedule so they would be aware of the planned, future discussions about decision making. We did not use phone diaries or other

structured recall techniques, however, some recall bias may still have been present. Additionally, it is possible that having a camera in the exam room during the recorded visit influenced the interactions among providers, parents and patients. However, others have shown only minimal effects from video recording clinic visits.(45) Additionally, this study was not structured to examine the influence the child's age may have on parent decision making.

By analyzing the trajectory of each individual's decision-making process we were able to detect subtle differences that may have been lost in a cross-sectional analysis. Moreover, the ability to compare responses between parents, over time, allowed us to develop a more complete view of how family decision making occurs. Using these methods, our prospective longitudinal study expanded what prior research had suggested,(1, 3, 7, 8) namely, that decision making in chronic conditions is a process, not an event. These findings lead us to envision a future where, at the time of their child's diagnosis, parents are invited to participate in workshops, on-line learning or other modalities that help them anticipate future decision trajectories and develop decision-making skills such as articulating treatment goals, asking questions and evaluating evidence. These skills would be capitalized upon later in the disease process by using decision-specific interventions, such as decision coaching programs or values clarification exercises, which are appropriate to the decision at hand and utilize skills families develop as they gain experience with the chronic condition. Future work should consider the roles of children and adolescents in this decision process and how such roles may be influenced by developmental trajectories.(46, 47) Additionally, research is needed to understand the generalizability of this decision-making process to other conditions and settings, such as adult patients.

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Appendix: Interview Guides

2-WEEK FOLLOW-UP INTERVIEW

Use questions as general guide to conversation. Questions need not be asked verbatim. Do not ask questions that participant already answered spontaneously.

Section A: Introduction

Thank for taking the time to talk with me today and for letting us record _____'s GI/Rheumatology visit on _____. Today I want to talk with you about what happened at that visit and since then. As we talk, if there are any questions you don't know the answer to or don't feel comfortable answering, feel free to tell me.

To begin with, I would like for us to talk about the actual doctor's appointment.

- Tell me about that appointment.

Prompts: What was discussed? Summarize what happened during the visit.

- What decisions, if any, were made during the appointment?
- What decisions were discussed but not made during the visit?

Section B: Non-biologics decisions

If a non-biologics related decision was made in clinic:

- Who participated in making the decision to _____? In what way did each person participate?
- What things did you consider in making this decision?
- What information did you want to help make this decision?
- Where did you find that information?

If a non-biologics related decision started in clinic:

- Who is participating in making the decision to _____? In what way is each person participating?
- What things are you considering in making this decision?
- What information is being used to make this decision?
- Where are you looking for this information?

Section C: Biologics decision

(If biologics decision not mentioned, prompt interviewee)

Have you and _____'s doctor discussed treatment with Humira/Remicade/Enbrel?

- Tell me about your discussion about (Humira/Remicade/Enbrel).
- Was this the first discussion about (Humira/Remicade/Enbrel) with the doctor? In general?
- Had you thought about/known about this before this appointment? Prompt: What were your thoughts leading into this appointment?
- What is the decision you are making/had to make? (i.e., what are you choosing between?)
- Where are you in this decision?

Prompts: Totally undecided, gathering information, leaning one way, decided etc.

- Tell me about what has happened since the appointment.

Prompts: Who have you spoken to? Where did you look for information? Did you and your child discuss this? [Follow up with, "Tell me more about that" to avoid specific questions]

[If Decision was Made]

- Who participated in making the decision? In what way did each person participate?
- What concerns about starting TNF α i treatment did you voice to [the provider]?
 - Have you shared your concerns with physician?
- What expectations about starting TNF α i treatment did you voice to [the provider]?
 - Have you shared your hopes with physician?
- What things did you consider in making this decision?
- What information did you get from provider/clinic regarding the risks/benefits of this treatment option?
 - What information was used to make this decision (see prompts below)?
- Is there any information that you wish you had but were unable to get before making this decision?
- Have you told your provider/clinic of the decision?
 - When did you tell your provider/clinic that the decision was made?
 - How did you tell your provider/ clinic that the decision was made?

[If No Decision Made Yet]

- Who is participating in making this decision? In what way is each person participating?
- What concerns about starting TNF α i treatment did you voice to [the provider]?
- What expectations about starting TNF α i treatment did you voice to [the provider]?
- What things are you considering in making this decision?
- What information did you get from provider/clinic regarding the risks/benefits of this treatment option?
 - What information are you using to help make this decision?
 - What information are you still looking for?
- What steps are you planning to take regarding this decision? Did you make plans with provider to discuss the decision?

6- AND 10-WEEK FOLLOW-UP INTERVIEW

Use questions as general guide to conversation. Questions need not be asked verbatim. Do not ask questions that participant already answered spontaneously.

Section A: Introduction

This interview is going to be very similar to our last one. As a refresher, if you don't know the answer or don't feel comfortable answering a question, feel free to tell me.

Once again, I would like for us to talk about your son's/daughter's clinic visit on [give date]
[6 weeks ago/10 weeks ago]

- What part of the visit is now the most memorable?
- What, if anything, related to the appointment have you continued to think about?
- How have your thoughts about that visit on _____ changed since our interview a month ago?
- What new decisions related to [patient's] JIA/IBD, if any, have been discussed since our last interview? Tell me about that discussion.

[ONLY IF A NEW DECISION HAS COME UP SINCE LAST INTERVIEW THAT IS A NONBIOLOGICS DECISION]

Section B: Non-biologics decisions

For a non-biologics decision made since last interview:

- Who participated in making the decision to _____? In what way did each person participate?
- What things did you consider in making this decision?
- What information did you want to help make this decision?
- Where did you find that information?

For a non-biologics decision started since last interview:

Who is participating in making the decision to _____? In what way is each person participating?

What factors are you considering in making this decision?

What information is being used to make this decision?

Where are you looking for this information?

Section C: Biologics decision

Now I'd like to revisit the decision you made/are making about [specific biologic from prior interview].

When you think about it now, what is the decision you are making/had to make? (i.e., what are you choosing between?)

Where are you in this decision?

Tell me about what has happened since the appointment. Since our last interview.

Prompts: Who have you spoken to? Where did you look for information? Did you and your child discuss this? [Follow up with, "Tell me more about that" to avoid specific questions]

[If Decision was Made]

- Who participated in making the decision? In what way did each person participate?

- What concerns about starting TNF α i treatment did you voice to [the provider]?
 - ☐ Have you shared your concerns with the physician?
- What do you hope to get from biologics treatment?
 - ☐ Have you shared your hopes with the physician?
- What things did you consider in making this decision?
- What information did you get from provider/clinic regarding the risks/benefits of this treatment option?
 - ☐ What information did you use to help make this decision?
 - ☐ What information are you still looking for?
- Is there any information that you wish you had but were unable to get before making this decision?
- Have you told your provider/clinic of the decision?
 - ☐ When did you tell your provider/clinic that the decision was made?
 - ☐ How did you tell your provider/ clinic that the decision was made?

[If No Decision Made Yet]

Who is participating in making this decision? In what way is each person participating?

What concerns about starting TNF α i treatment did you voice to [the provider]?

What do you hope to get from TNF α i treatment?

What factors are you considering in making this decision?

What information did you get from provider/clinic regarding the risks/benefits of this treatment option?

- ☐ What information are you using to help make this decision?
- ☐ What information are you still looking for?

What steps are you planning to take regarding this decision? Did you make plans with provider to discuss the decision?

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Themes	Index Visit	2 Week Interview	6 Week Interview
Treatment Options	6-MP vs methotrexate vs biologic	<u>Current med vs biologic</u> <i>biologic vs 2nd opinion</i>	<u>biologic vs current med vs do nothing</u> <i>Symptoms vs biologic</i>
Treatment Concerns	Infection; insurance Lymphoma risk Physician's potential bias	<u>Lymphoma, fungal infection</u> <i>Lymphoma, cost, what's the next step</i>	<u>Lymphoma, patient experiencing flu-like symptoms, and headaches</u> <i>Leukemia</i>
Decision Factors	Side effects Potential length of treatment Quality of life	Risks < benefits; <u>fewer pills, improved quality of life</u> <i>Trust physician; local experience with treatment</i>	<u>Patient's perspective</u> <i>Likelihood of improvement; local experience</i>
Decision Making Roles	Physician gave recommendation Parents listened Patient not in room	<u>physician gives recommendation;</u> <u>parents decide</u> <i>Parents decide and discuss with patient</i>	<u>physician answers questions; parents decided; patient asked questions</u> <i>MD gave rec; parents decided; told patient the decision</i>

Underline indicates mother; *italics* indicate father.

Figure 1.
Sample (partial) Family Matrix

Themes	Family 1	Family 2	Family 3
Treatment Options	<u>Added do nothing option</u> <i>Changed biologic alternative</i>	<u>No change</u> <i>Added third option of "something else"</i>	<u>Biologic alternative switched from current med to methotrexate</u> <i>From choosing between meds to choosing between symptoms vs no symptoms</i>
Treatment Concerns	<u>Potential risks to actual side effects</u> <i>Focus on cancer risk, no cost concerns at the end</i>	<u>Less specific concerns over time</u> <i>Persistent side effect concerns</i>	<u>Decreased focus on side effects; increased concern about IV</u> <i>From minimal concerns to no concerns</i>
Decision Factors	<u>From risk/benefit to focus on patient</u> <i>On-going focus on local experience; decreased focus on trust</i>	<u>Increase focus on patient experience with current treatment and lack of long term data</u> <i>Increased side effect concerns; increased "belief in current treatment"</i>	<u>Increased focus on treatment safety and efficacy</u> <i>Initial focus on physician advice; increased focus on quality of life</i>
Decision Making Roles	<u>Stable role for parents; increased role for patient</u> <i>Stable roles</i>	<u>Decreasing role for physician in decision</u> <i>Stable role for self and doctor; decreased role for wife</i>	<u>Stable roles for physician, parents and patient</u> <i>Stable roles</i>

Underline indicates mother; *italics* indicate father.

Figure 2.
Sample (partial) Longitudinal Analysis Matrix

Table 1**Major Coding Framework Themes and Definitions**

Treatment options

- The options participant considered relative to each treatment decision

Decision making events

- Active or passive events that participant believe relates to the treatment decision, such as medical testing or disease flares
- Actions the participant has taken or plans to take to facilitate decision making
- May proceed recorded clinic visit

Thought processes and emotions

- Comments that reflect what the participant has or is currently thinking or feeling regarding the decision making process

Treatment concerns

- Any concern or worry mentioned regarding treatment with biologics
- Must include some sense of emotion rather than just a listing of side effects

Treatment expectations

- Any hope or expectation mentioned regarding treatment with biologics
- Must include some sense of emotion not just a listing of data regarding treatment effectiveness

Decision factors

- Any event, emotion, concern or expectation that the participant indicates is influencing decision making about biologics

Decision making roles

- Description of what each participant did as part of the decision making process
-

Table 2**Participants**

Characteristic	IBD	JIA
Visit participants—providers		
Years in Subspecialty		
Median (Range)	9.5 (3-19)	16.2 (1-33)
Sex (n)		
Male	3	3
Female	1	3
Provider type (n)		
Physician	4	5
Nurse Practitioner	0	1
Ethnicity (n)		
Non-Hispanic	4	6
Race (n)		
White	3	5
Asian	1	1
Visit participants—family members		
Patient (n)	12	9
Median Age in Years (Range)	11.5 (7-16)	9 (2-18)
Mother (n)	12	9
Father (n)	2	6*
Maternal Education		
< College Graduate	2	5
College Graduate	6	1
Graduate or professional degree	4	3

* one father present at the visit declined to participate in interviews

Table 3

Illustrative Quotations

Themes and Subthemes	Quotations
Decision Evolution: <i>Decision Events</i>	
Research	<p>"I spend hours on forums, threads, on blogs, drug company websites..." (mother #12)</p> <p>"I search the internet once in a while..." (mother #4)</p>
Conversations	<p>"...I had actually discussed it with [2 doctors] and had asked the few questions I have because I had looked into it before." (mother #8)</p> <p>"Mostly me and [the patient] have talked about it..." (mother #21)</p> <p>"I spoke with [the nurse] and we had a 20-minute discussion on the phone." (mother #1)</p>
Medical events	<p>"They did a Doppler [of his swollen knee] in one of the visits..." (mother #19)</p> <p>"We take her off of those medications and then she gets sick again." (mother #21)</p> <p>"...then his pain came back between the two visits and when I called we just enacted the plan we already had in place." (mother #28)</p>
Constant Element: <i>Role Perceptions</i>	<p>"[the doctor] pretty much explained what the new medicine was and how it works and me and my wife pretty much decided that we wanted to stay with the methotrexate" (father #31; 2 week interview)</p> <p>"...He just kind of discussed it with us and then we were all pretty much like, we'll all just stay on the methotrexate." (father #31; 6 week interview)</p> <p>"[The doctor] offered the suggestion and then my husband and I talked it over and I think both of us were on board with trying something." (mother #19; 2 week interview)</p> <p>"Dr. L suggested it and explained it to us and we decided that was the next, natural step to take." (mother #19; 10 week interview)</p> <p>"Well, we listened to the doctor's advice...we talked it over and we just made the decision..." (father #19; 10 week interview)</p>
Constant Element: <i>Internal Processing</i>	<p>"We're trying to figure out what to do next." (mother #6; 2 week interview)</p> <p>"I still think about which one will be the best one to start." (mother #6; 10 week interview)</p> <p>"I guess we were just trying to decide with all of her other medical issues" (mother #25; 2 week interview)</p> <p>"We were trying to decide what would be the next course of treatment..." (mother #25; 6 week interview)</p> <p>"Basically I thought, 'well, are we at that point? Are we at the end of the rope?' I was just thinking about the [biologic therapy]." (mother #1; 2 week interview)</p> <p>"...now that she did start it...I'm thinking, 'Oh no, did we do the right thing?'" (mother #1; 6 week interview)</p>
Evolving Element: <i>Expectations</i>	<p>"Definitely, I just think letting him and seeing him feel better is my biggest goal. Trying to kick it in to remission where he's not walking stiff in the morning and things like that." (mother #28; 2 week interview)</p> <p>"Just, you know, minimal side effects since that seems to be what was bothering him the most with [prior treatment]." (mother #28; 6 week interview)</p>
Evolving Element: <i>Worry and Hope</i>	<p>"Can we get her off of her 15 pills and go down to the [biologic therapy] in one pill? You know, is this going to be the right treatment? Are we going to have to switch again? Just a lot of unknowns right now." (mother #9; 2 week interview)</p> <p>"I hope she stays well with less flare-ups." (mother #9; 6 week interview)</p>

Themes and Subthemes	Quotations
	interview)
	“We still both had some reservations about the medication particularly because of the severity of the side effects.” (mother #3; 2 week interview)
	“It seems like he’s feeling much better...I’m just glad at this point that we moved forward with that decision.” (mother #3; 6 week interview)
	“I, eventually, would like to see [my son] go into remission with is. That’s our hope.” (mother #21; 2 week interview)
	“You always worry about any medications that something shows up years later. ...you know, this has been taken off the market because of this, this and this.” (mother #21; 6 week interview)
Element with Constant and Evolving Components: <i>Option Framing</i>	<p>“So that was the biggest decision...whether to try a different treatment option or hold off on therapy or to continue with the therapy and try to get rid of side effects.” (mother #28; 2 week interview)</p> <p>“So one is the biological, then they also talked about leflunomide and then we talked about methotrexate.” (mother #28; 6 week interview)</p> <p>Interviewer: What were you choosing between?</p> <p>“You know, letting the methotrexate do a slow, I guess you could say slow healing process where when we add the [biologic therapy], it will be a faster heal.” (mother #23; 2 week interview)</p> <p>“Whether it was the right time to put her on an additional injection. If it would be worth her having the two shots a week instead of one shot.” (mother #23; 6 week interview)</p> <p>“Well, I know [the nurse practitioner] had gone over [injected biologic] and I know there’s [different biologic] and I know she talked about one, I think it was [third biologic] and I can’t remember there might have been a couple other ones.” (mother #26; 2 week interview)</p> <p>“[We were] choosing between her starting the [biologic] to make her feel better or her staying where she was without that...” (mother #26; 6 week interview)</p>