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Illness Uncertainty in Parents of Children with Juvenile Idiopathic Arthritis

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Objective. To gain a better understanding of uncertainty regarding the illness experienced by parents of children with juvenile idiopathic arthritis (JIA).

Methods. Parents/guardians of a child or young person (aged less than 18 years) diagnosed JIA were recruited in the United Kingdom via the National Rheumatoid Arthritis Society JIA group. Semistructured telephone interviews were conducted with the parents.

Results. Twenty parents took part, including 19 mothers and one father. Their children with JIA were mostly female ($n = 15$; 75%) with polyarticular arthritis ($n = 12$; 60%), averaged 8 years of age, and had been diagnosed for a mean of 3.7 (SD 2.3) years. Parents expressed uncertainty in the following five key domains: diagnosis, cause, symptoms, and prognosis; medical management; impact; parenting uncertainty; and awareness of JIA. All participants expressed uncertainty in at least four of the five domains. Although parents' uncertainty in the early stages of the disease related to lack of information and understanding of JIA, much uncertainty could not be resolved by receipt of information. These included concerns about their child's future and a lack of support with managing the emotional aspects of living with JIA.

Conclusion. We found that parents' experiences of uncertainty went beyond dealing with the purely medical aspects of JIA. Provision of information about JIA, although essential, is not sufficient to help parents manage the considerable uncertainty they experience about many aspects of their child's JIA. Identifying ways to incorporate support for coping with uncertainty into routine care will be an important way of supporting parents to care for their child with JIA.

INTRODUCTION

Juvenile idiopathic arthritis (JIA) is the most common rheumatological condition in childhood, with a global prevalence of 32.6/100 000 (1). JIA comprises a group of heterogeneous diseases of unknown cause ("idiopathic") that begin before 16 years of age and cause inflammation of one or more joints, lasting at least 6 weeks (2). The current International League of Associations for Rheumatology classification of JIA specifies seven subtypes (systemic, oligoarticular, rheumatoid factor–negative polyarticular, rheumatoid factor–positive polyarticular, psoriatic; enthesitis-related, and undifferentiated), which are based on the number of affected joints and extra-articular involvement in the first 6 months of the disease (2). The robustness of this classification system has, however, been criticized (3,4), and its revision is underway (5).

There is currently no cure for JIA, and, without effective treatment, it can cause considerable pain, fatigue, disability, and impaired quality of life (6,7). Several pharmacological treatments are available that help to control the disease, including intraarticular glucocorticoids and disease-modifying antirheumatic drugs such as methotrexate and biological therapies, which have considerably improved JIA prognosis in recent decades. However, the medications do not work for everyone, and less than half of those with JIA achieve drug-free remission after 10 years (8). Often, children receive serial medications before finding one that works for them, during which time they can incur irreversible damage while experiencing debilitating medication side effects (9,10), pain, and psychosocial consequences of living with uncontrolled arthritis (11).

Living with JIA, in which the disease course and treatment effects are unpredictable, inevitably involves the experience

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SIGNIFICANCE & INNOVATIONS

- Although uncertainty around JIA prognosis is inevitable, this study found that uncertainty permeates many other aspects of parenting a child or young person with JIA. This included uncertainty about medical management, what impact JIA will have on the child and family, aspects of parenting a child with JIA, and a low level of awareness of JIA in the population.
- Our findings broaden conceptualizations of uncertainty in relation to childhood chronic disease. Prior research on parental illness uncertainty has mainly focused on the domains we termed “diagnosis, cause, symptoms, and prognosis” and “medical management.” Other important domains identified in the current research concerned impact, parenting uncertainty, and awareness of JIA.
- Information alone is not sufficient to enable parents to manage uncertainty. Identifying ways to incorporate support for coping with uncertainty into routine care is required to support parents in caring for their child with JIA.

of uncertainty (12,13). For parents, being able to manage the stresses of their child’s illness and treatment is important in maintaining their child’s wellbeing (14); however, uncertainty may limit the ability to cope (15). Higher levels of illness-related uncertainty have been found to be related to poorer wellbeing in both patients and carers in a range of long-term conditions (16–21).

Two studies of uncertainty have been conducted with parents of children with juvenile rheumatic diseases (a majority of whom had JIA) and found that parent perceptions of illness uncertainty were related to parent and child distress (22,23). These studies reported illness uncertainty using a single dimension score, whereas research by Cleanthous et al in adult rheumatological disease (24) found that illness uncertainty is multidimensional, incorporating five domains, namely, uncertainty relating to symptoms and prognosis, medical management, self-management, impact, and social functioning. Health professionals can play an important role in helping parents to manage uncertainty, but, to do so, they require an understanding of parents’ experiences. This study aimed to gain an in-depth understanding of the uncertainty experienced by parents of children and young persons with JIA.

PATIENTS AND METHODS

Participants. Participants were recruited from the United Kingdom-based National Rheumatoid Arthritis Society JIA group (JIA-at-NRAS). Inclusion criteria were 1) parent/guardian of a child or young person (aged less than 18 years) who has a diagnosis of JIA and 2) the ability to speak English. Interviews were conducted with one parent/guardian per child. Participants were recruited using convenience sampling, and we sought to recruit both mothers and fathers.

Procedure. Ethical approval was granted by the City, University of London School of Health Sciences Research Ethics Committee (reference number Staff/16-17/08).

The study was advertised via an email to members of the JIA-at-NRAS group, and information about the study was featured in their newsletter, website, and Twitter. The information contained a link to the participant information sheet and consent form and the contact details of the research team. For parents of a child aged 7 years or older, an information sheet for children and young persons was also provided via the link, and parents were asked to discuss the study with their child. Parents interested in taking part contacted the research team to arrange a convenient time for a telephone interview. As the interviews were conducted over the telephone, the consent procedure was audiorecorded.

Data collection and analysis. Semistructured telephone interviews were conducted by a postdoctoral research assistant experienced in qualitative methods (CP), using an interview topic guide informed by other research on illness uncertainty (24, 25) (Appendix A), which explored several areas of potential uncertainty concerning their child’s illness, including symptoms, prognosis, treatment, and impact. The interviews were recorded and transcribed verbatim. The transcripts were imported into NVivo, version 11, to facilitate analysis. Transcripts were analyzed using thematic analysis, which is a method for identifying, analyzing, and reporting patterns within data (26), and quotations that expressed aspects of uncertainty were coded. CP conducted preliminary coding of the interview data and developed an initial coding structure, which categorized codes into domains and subdomains. A second author (KM) read all transcripts and reviewed the initial coding structure along with the quotes coded within each domain/subdomain. Thereafter, CP and KM discussed and revised the coding structure in an iterative process until consensus on the final structure and the list of uncertainty domains/subdomains was reached. During this iterative process, the developing coding structure was cross-referenced with the uncertainty framework developed by Cleanthous et al [24], and, where themes in this framework reflected the findings of the current study, these categories were applied to the data.

RESULTS

Twenty parents participated between November 2016 and January 2017; sample characteristics are shown in Table 1. Almost all were mothers (95%), and a majority were of White British ethnicity (60%). Most (80%) were married/living with a partner, and 65% had attended higher education. Just over half (55%) were in paid employment. The characteristics of the participants’ children with JIA are shown in Table 2. Most were female (15; 75%), and 16 (80%) were less than 13 years old. The interview duration averaged 42 minutes, ranging from 17 to 80 minutes.

Table 1. Sample characteristics (parents)

Characteristics	Results
Sex, n (%)	
Female	19 (95)
Male	1 (5)
Age, mean (SD)	40.95 (5.7)
Ethnicity, n (%)	
White British	12 (60)
White European	3 (15)
Mixed (White and Black)	1 (5)
Mixed (White and Asian)	1 (5)
Mixed (White and Maori)	1 (5)
Mixed (not specified)	1 (5)
Asian (Chinese)	1 (5)
Marital status, n (%)	
Married/living with partner	16 (80)
Divorced	3 (15)
Single	1 (5)
Employment status, n (%)	
Full-time	4 (20)
Part-time	7 (35)
Full-time homemaker/carer	8 (40)
Student	1 (5)
Education (highest qualification), n (%)	
A levels*/vocational level 3 or equivalent	5 (25)
Undergraduate diploma/level 5 or equivalent	1 (5)
Higher education	13 (65)
Other qualifications, level unknown	1 (5)

* A levels are national exams taken at an approximate age of 18 years

Uncertainty domains. Five domains and 19 subdomains of uncertainty were identified. All participants expressed uncertainty in at least four of the five domains. Table 3 details the full list of uncertainty domains and subdomains, the number of participants who reported expressions of uncertainty within each of the subdomains, and illustrative quotes for each subdomain. These five domains captured the parents' feelings of uncertainty around JIA, its treatment, and impact as well as uncertainty around their role as a parent in managing their child's JIA and the social context of understanding of JIA. Each of the domains are described below.

Table 2. Child characteristics

Characteristics	Results
Sex, n (%)	
Female	15 (75)
Male	5 (25)
Age, yr, mean (SD)	7.95 (3.9)
JIA subtype, n (%)	
Polyarthritis	12 (60)
Oligoarthritis	4 (20)
Extended oligoarthritis	3 (15)
Systematic Onset	1 (5)
Years since diagnosis mean (SD)	3.7 (2.3)
Current medication, n (%)	
DMARDs	5 (25)
Biologics	2 (10)
DMARD and biologic treatment	8 (40)
Steroids	1 (5)
None	4 (20)

DMARD, disease-modifying antirheumatic drug; JIA, juvenile idiopathic arthritis.

Diagnosis, cause, symptoms, and prognosis. All participants expressed some uncertainty regarding diagnosis, cause, symptoms, and prognosis. The sense of uncertainty experienced by parents of a child with JIA began with the diagnosis. Six parents expressed uncertainty over whether their child had been given the correct diagnosis. In particular, this uncertainty stemmed from doubts around the definitions of the subtypes of JIA and how the disease was diagnosed. This uncertainty over the diagnosis was also evident when some children had their diagnosis changed or faced the possibility of a changed diagnosis.

Although the aetiology of JIA is unknown, in trying to make sense of the diagnosis, many parents contemplated factors that may have caused the disease. Nine parents expressed uncertainty over the cause of JIA and how their child developed the disease. Within this, three parents expressed some feeling of uncertainty over the parent's level of responsibility in causing their child's JIA and initially having had feelings of blame that they may have done something to provoke the onset of the disease.

In the face of these feelings of uncertainty, parents would at times attempt to attribute the cause of JIA to something, such as a virus. The fact there appeared "no rhyme or reason" for JIA starting meant many parents had difficulty accepting the diagnosis. One way the parents managed this uncertainty over the cause and diagnosis was to, as one parent described, remain in a state of "denial" over whether their child actually had JIA and view the onset of the disease as a "one off."

Uncertainty around symptom interpretation was a strong theme throughout the interviews and was expressed by 19 participants. This uncertainty was exacerbated when the child was too young to be able to articulate how they were feeling. In addition, parents described the difficulty in distinguishing their child's health status and symptoms from normal child behaviour, thereby making it difficult to judge whether their child was well or ill. When the child had a comorbid condition, it could increase uncertainty when interpreting symptoms as JIA related. Before a diagnosis, many of the parents described being faced with seemingly unexplainable symptoms and would attribute symptoms to JIA retrospectively.

Uncertainty over the prognosis of JIA was a strong theme in all of the interviews. This could be linked to the uncertainty over the diagnosis, which some considered made it difficult to predict the progression of the child's health. Parents described how they found it hard to deal with not knowing what was going to happen in the future. As one parent described it, the experience of having a child with JIA was "a very unpredictable journey," in which the child could be fine one day and unwell the next. Parents described the cyclical nature of JIA and how this influenced their feelings of uncertainty. They tended to oscillate between feelings of hope at the chance of recovery, which was experienced during periods of remission, and then uncertainty as to whether the JIA might ever go away if the child experienced a flare. Parents used words such as "cyclical," "cycle," "roller-coaster," and "fluctuate" to capture the sense of uncertainty

Table 3. Uncertainty domains and subdomains

Domain	Subdomain	Explanation	N	Illustrative Quotes
Diagnosis, cause, symptoms, and prognosis	Diagnosis	Unsure whether diagnosis is correct	20 6	"She's got Oligo JIA but yesterday, [at clinic] (...), they actually changed it to Poly. I think the whole uncertainty thing comes in. Every time we go somewhere it changes slightly."
	Cause	Uncertainty around the cause of JIA	9	"So I was blaming myself for a long time for her illness. That [there] was something I could have possibly done better or differently or not eaten."
	Symptom Interpretation	Interpreting physical sensations, distinguishing symptoms from normal child development and other conditions, interpreting symptoms prior to diagnosis	19	"In retrospect she had a little episode with a joint in her foot (...) a few months before with her foot where it had swollen up but we thought she'd injured herself and didn't do anything about that really, just waited for it to go away." "Is he having tantrums because something's hurting him and he can't communicate it? Or is he having tantrums because he's a toddler and that's what they do? Or because he's got a cold and it's nothing to do with arthritis?"
	Interpretation of health status	Judging how well child is	12	"Sometimes she'll be reluctant to tell us, I'll ask are you sore? Have you got a sore or something? No. I'm all good. (...) So I say, you'll tell daddy if you're sore eh? No. And so it can be a little bit difficult to say is something going on or not."
	Prognosis	Unsure of future progression of the disease	20	"And I'm scared will it extend further, is it somewhere else that we don't know about (...) I'm scared of what the future brings, (...) will he ever outgrow it, is it going to spread, (...)"
Medical management	Treatment	Uncertainty over treatment regime: necessity, changes to medication, side effects	20 20	"I don't know how long this one is going to work for and indeed when this stops working I don't know if there are any other options. I don't know what the next stage would be." "If he's been in remission for at least a year they will start talking about coming off and when you come off you're back in the wilderness and then you don't know if it's going to come back."
	Trust in doctor	Medical professional knowledge and ability to help with condition. Doctors' uncertainty around initial diagnosis and prognosis	13	"I have to trust in the consultant who's looking after him and the nurse practitioner because they know what they're doing and I don't and I have to put all my trust in them, is how I see it because I'm not an expert and they are and I don't particularly like that I have to say because you don't always know or feel, what I'm saying is you don't always know if some things are done because it's cost effective rather than the best thing." "But yeah at the same time I've come to realise all these years that even the consultants they don't really know. (...) they give you something and some kids start on something and then three months down the line they realise it doesn't work and then they have to start something else."
	Uncertainty in health services and support	Gaps in support, unreliable services, uncertainty around navigating different services	11	"The part that was also overwhelming is to understand how the NHS is working and the different parts of it." "The emotional side of it and coping with injections and needle phobias and how you manage being different and all that kind of stuff, I don't think there's nearly enough done about that."
	Transitioning to adult care	Uncertainty around child transitioning to adult care	2	"The other concern as well is of course that she will need to be, have some sort of transitional treatment by, there is a transition consultant who deals with adolescents. So that would be good to, we don't know how to get into that (...)"
Impact			16	
	Impact on child	Impact on child's future including emotional wellbeing, social functioning, and uncertainty around impact of JIA on child's personality and character.	15	"I wonder how she will accept the disease as she comes to know more about it and what her reaction to the disease will be. (...) she's grown up with it, it's all she's known, but as she becomes older I just wonder if she's bitter or resentful." "Yeah, I worry that he won't be able to make friends in the same way and he'll be ostracised because of it because he's a boy and boys do sport."
	Forward planning	Ability to plan for short- and long-term events	6	"We've got two weeks holiday booked (...) in the summer and we cannot plan, there's nothing you can plan, (...) we don't know whether we'll have to find local hospitals or there'll be medication down there and there's, it just does affect everything."
	Impact on family members	Impact on family as a whole and other children	9	"But balancing the needs of a child with what is I guess a long-term unpredictable condition with your other child or children and managing that... It is [settled] now and that's, but it still has a strain on us as a family I think."

(Continued)

Table 3. (Cont'd)

Domain	Subdomain	Explanation	N	Illustrative Quotes
Parenting uncertainty	Coping	Ability to cope physically and emotionally with demands, and feelings of powerlessness	20 12	"I'm assuming I'll find a way to cope. I try not to think about not coping because then I might not cope." "You love your daughter, very caring, very close bond and then when you see her in pain you feel powerless about that. So powerlessness I think is one of the things that I struggle with. You think actually, I can, you can do things to make her more comfortable and you still feel this level of powerlessness."
	Managing child's JIA	Uncertainty around how best to manage child's JIA including symptom management, managing medication, child's behaviour and emotional wellbeing, managing JIA into the future including child's medication self-management	12	"You don't want to be overprotective, but you want to make sure you care for her, given her JIA, and at the same time you're parenting and trying to set boundaries. And, so definitely I think overall your parenting is influenced." "On the one hand I don't want to allow her to be rude and uncooperative but on the other hand I don't want to discipline her for those things if it's a manifestation of her trying to express how she's feeling if she's feeling sick or she's feeling tired or in pain."
	Role requirements	Unsure of role as parent and carer and role expectations	4	"I think my role as a mum has changed into more of a medical role sometimes. I don't want her to resent me as she gets older for being the one that's always holding her down for things. That's my main worry with my role with her."
	Making decisions about child's care	Uncertainty when making decisions about child's care	13	"Trying to make the best decisions I think on her behalf I think it would be easier if you're making the decision for yourself. It's just the responsibility of doing the right thing and safeguarding her emotional wellbeing as well as her physical wellbeing."
Awareness of JIA	Social awareness	Understanding among family, friends, and others of the condition and support received, invisibility of condition	19 18	"A lot of people still don't understand it, because we still get the 'What, children get arthritis? It cannot be, are you sure? It's an old people thing.' "School again, it's a bit like talking to your family, it's quickly brushed under the carpet, they don't really want to know, but they don't understand."
	Gaps in information	Areas parents feel are missing from JIA information, lack of awareness prior to diagnosis	11	"I had to find out about disability benefits and fight very hard for it and all the other things that I'm entitled to. You don't get given that information you have to find it for yourself really." "Perhaps there have been times when I would have liked more information and I didn't accept it. But I see, I get now why there isn't more out there, because it just, they can't tell you facts and certainties because there aren't many anyway."
	Confidence in information about JIA	How confident parents feel about the information they receive	4	"There's also some misinformation and there's also a lot of American information, which doesn't necessarily apply to the treatments that we have over here. So, I think you can go a bit wrong with some of the Googling and I think I did at the beginning, trying desperately to understand it all."

JIA, juvenile idiopathic arthritis.

over the prognosis, which included much anticipation and waiting, either for remission or another flare. A sense of hope allowed the parents to try to remain positive despite the uncertainty, for instance the hope that their child would recover or that soon a cure would be discovered. However, the fear that their child would enter adulthood and continue to struggle with symptoms of the disease was a real concern and a source of uncertainty for many of the parents.

Medical management. The role of medical management was a key theme in the parents' interviews, in particular uncertainty around treatment for JIA, which was expressed by all 20 participants. Two parents expressed uncertainty around the necessity of the treatment, for example, if their child was on the most

appropriate treatment or whether medication was still necessary once symptom-free. Twelve parents expressed uncertainty and worry about whether their child's current medication would continue to work and what would happen if it stopped working. Although hoping that their child could eventually come off medication, three parents also expressed the fear about the medication being stopped, with the possibility that the disease could flare but the medication not work when restarted.

Sixteen parents expressed concern about the side effects of medication. These included the immediate impact of the treatment on their child's immune system and potential longer-term impact, such as effects on fertility. Further, parents had difficulty in identifying the impact of the treatment on their child's normal development. There was also uncertainty around distinguishing

the side effects of the treatment from the symptoms of the disease itself.

Parents mostly felt well supported by their medical teams, and although they described wanting a clearer picture from medical professionals around the prognosis, they did acknowledge that this is not possible. Ten of the parents described uncertainty among doctors before receiving a diagnosis, where there was often a long process before symptoms were identified as JIA. This was, for some of the parents, a frustrating time of feeling dismissed or “fobbed off” by health care professionals.

Almost all parents reported receiving very good care from their clinical teams, and they were an important source of support when a parent felt uncertain about an aspect of their child's illness. However, several had faced uncertainty when trying to navigate health services early on in their child's illness. Gaps in care included lack of availability of support for dealing with the emotional or psychological impact on the child and family as a whole, which was mentioned by two parents, and another referred to an inability to raise all her concerns with the rheumatologist in front of her child. In parents whose children were in their teens, this uncertainty around the support from hospitals and medical professionals also extended to uncertainty about how and when their child would transition to adult care.

Impact. Besides uncertainty relating to the general prognosis, reported above, 16 participants expressed uncertainty and worry regarding the impact JIA would have on their child. This included their child's future emotional wellbeing and social functioning and how their child might cope without them as they grow older. Parents had concerns about whether their child would be able to engage with school activities, for example, sports classes. They also highlighted uncertainty about the emotional impact of JIA, including the experience of having medical treatment from a young age. The uncertainty of the emotional impact was also emphasized if the child was unable to describe their emotions to their parent. Parents had concerns about how their child will feel as they get older and begin to understand the disease. As many of the children were diagnosed at a young age, parents mentioned uncertainty around the impact of JIA on their child's personality and character. Some parents contemplated the extent to which JIA may have exacerbated mood swings or toddler “tantrums” or affected psychological wellbeing.

Forward planning was also difficult because of the unpredictable nature of JIA. Parents described how JIA could affect the ability to plan for short- and long-term events. They also spoke of the impact that having a child with JIA had on the family as a whole and concerns about the impact on other siblings.

Parenting uncertainty. All participants expressed some uncertainty around their role as a parent and carer for their child and their ability to cope and manage their child's JIA. Some

described difficulty in distinguishing their roles as parent and carer and feeling unsure whether they were doing a “good job.”

Twelve parents described their worries in physically and emotionally coping with being a parent of a child with JIA. They described coping on a day-to-day basis rather than thinking too far into the future. Indeed, some parents mentioned they tried not to think about future problems in order for them to cope with the present. This uncertainty about the ability to cope also caused feelings of powerlessness. Parents felt that JIA was “calling the shots” and struggled with seeing their child in pain and being unable to remedy it. Specifically, the parents were uncertain about how to manage different aspects of their child's illness, including how to manage pain and treatment as well as their child's emotional wellbeing. Many of the parents expressed a sense of doing the best they can but feeling unsure whether they were doing enough.

Another dimension the parents highlighted was uncertainty about how to manage their child's behaviour, for example, setting the right boundaries while taking their child's illness into consideration. Parents also felt uncertain about their role in managing their child's illness as they entered adolescence and adulthood. There was some concern about what would happen when the child would come to manage their treatment themselves, as most parents still had control over their child's treatment regime.

Thirteen parents described uncertainty when making decisions about their child's care, particularly when making decisions on medication and treatment, which could relate to commencing, changing, or coming off medication. For parents, the responsibility of their child's care could feel great. They described a process of continually weighing up the options before making a decision. However, a number of parents did note that though they struggled initially with making decisions, their confidence in their own judgement had grown over time.

Awareness of JIA. The uncertainty the parents experienced was set within a broader context of a lack of understanding and awareness about JIA, which was mentioned by 19 participants. Seven remarked that they had little awareness and knowledge of JIA before their child was diagnosed. They also described a lack of understanding among friends and family and felt people did not appreciate the realities of living with JIA. This was made more difficult by the invisibility of the condition, with parents describing JIA as a “hidden disability.” The parents found that other people did not acknowledge the seriousness of the disease if they saw the child looking well. Four parents mentioned problems with accessing support from their child's school, where they faced a lack of understanding of JIA. Related to this, 17 of the parents mentioned the use of peer support, mostly via an online social media group, that provided mutual understanding and feelings of reassurance as well as practical help and information.

Parents noted some gaps in knowledge; however, they acknowledged that these were due to a general uncertainty around aspects of JIA, such as prognosis, rather than being unable to access available information. All except one participant felt they had received sufficient disease information; this came from a range of sources, including the hospital team, relevant charities, the internet, and other parents. More disease information was not necessarily viewed as helpful to the parents, as it was felt that such information did not reduce feelings of uncertainty. Three parents actually felt they had received too much information and felt overwhelmed by it. Specific areas on which parents wanted more information and support were on managing the day-to-day problems and the emotional side of the disease for parents and children ($n = 5$) and information on accessing disability benefits ($n = 2$).

DISCUSSION

To date, there has been little research on the experiences of parents of children with JIA. Although uncertainty around JIA prognosis is inevitable, this study found that uncertainty permeates many other aspects of parenting a child or young person with JIA. We identified the following five domains of uncertainty: diagnosis, cause, symptoms, and prognosis; medical management; impact; parenting uncertainty; and awareness of JIA.

Other studies of uncertainty in parents of children with rheumatic diseases (22,23) have used the Parental Perceptions of Uncertainty Scale (PPUS) (27), which covers some, but not all, of the domains of uncertainty identified in the current study. The PPUS focuses mainly on uncertainty in the domains that we termed "diagnosis, cause, symptoms, and prognosis" and 'medical management' (although it does not include questions on uncertainty around making decisions relating to their child's care, which was expressed by participants in the current study). We found that parents' experiences of uncertainty went beyond dealing with the purely medical aspects of JIA, which has implications for how parents are able to support their children in managing their JIA. Corbin and Strauss (28) proposed that living with a chronic disease involves the following three key tasks: medical management (eg, taking medication and attending hospital appointments), adapting life roles such as school and relationships to the demands of the condition, and managing the emotional consequences of having a chronic disease. Parents' reports of uncertainty spanned these three tasks, highlighting a need for support that goes beyond the provision of information about the disease and its treatment.

Some of the uncertainty experienced by parents may be helped by a broader awareness of JIA in the population. A lack of awareness, both among the participants themselves and among nonspecialist health professionals, was a source of uncertainty during the early stage of the disease before diagnosis. Feelings of confusion, anxiety, and frustration with the health care system during this period have also been reported elsewhere (13). A

recent United Kingdom study reported that the median time from symptom onset to first pediatric rheumatology appointment was 23.6 weeks (interquartile range 12.3-50.4)(29) and did not vary significantly across the 10-year study period. The authors referred to several steps that have been taken in recent years to try to raise awareness of JIA in all doctors, including training for medical students and education for primary-care physicians and pediatricians, but they recognized a continued need for efforts to assist health professionals in the early recognition of JIA. They also recommend targeted education for schools, nurseries, and health visitors. A lack of awareness among the general population continued to be an issue for parents after diagnosis because teachers, family, and friends did not always appreciate the difficulties of living with JIA. Recent campaigns such as the World Young Rheumatic Diseases Day (30) may help to raise awareness of JIA.

The study findings highlight the important role that health professionals played in helping parents to manage illness uncertainty. Once a diagnosis was received, parents sought out and received information about JIA from their clinical teams. They also reported seeking information and support from clinicians as new issues arose. Almost all parents reported having very supportive clinical teams and having received sufficient information about JIA and medication.

Although some aspects of parent uncertainty can be alleviated by attending to unmet information needs, uncertainty management theory states that the relationship between information and feelings of uncertainty is complex (31), and, although information can help to reduce uncertainty, it may also maintain or even increase uncertainty by introducing new concerns or questions about the illness (32). The uncertainty of JIA is an ongoing challenge for parents throughout their child's illness, and new issues arise as their child develops, many of which cannot be mitigated by the provision of information. For example, some parents expressed uncertainty and worry about the extent of the emotional impact that JIA had or could have on their child and highlighted a lack of support for their child in dealing with the emotional side of living with JIA. This issue has also been identified by young people with rheumatic conditions who felt that psychosocial research should be given greater priority (33).

Parents also experienced uncertainty in their role as carer, which has been identified by other carers of those with both adult (25) and childhood illnesses (34,35). This highlights the importance of supporting parents in managing their uncertainty both for their own wellbeing and for the wellbeing of their child. Young people with rheumatic conditions have also recognised the effect that their illness has on their parents and the need for research into how their families could be better supported (33). Many parents turned to an existing online parent group, which they found helpful, but more routinely available support from health professionals for psychological issues requires greater consideration.

Although health professionals cannot eliminate all uncertainty about JIA, they can help parents to manage it. For example,

the development of decision aids (36) may help parents manage the uncertainty experienced around treatment changes. There is also evidence that illness uncertainty is modifiable; research has found that parents who use more positive coping styles experience less uncertainty (21), suggesting that interventions to teach coping strategies may help to alleviate some of the stress of illness uncertainty. Fedele et al (37) developed an intervention to help mothers of children newly diagnosed with cancer to cope with the uncertainty of their child's illness and treatment. The intervention, which included sessions on communication skills, methods of coping, problem-solving strategies, and how to use social support effectively (38), was found to have a significant benefit on child adjustment (37). The intervention of Hoff et al (39) for parents of children newly diagnosed with type 1 diabetes included techniques to manage uncertainty, such as information, role clarification, and training in problem-solving and communication skills. Although the level of uncertainty did not change, parental distress reduced over time in the intervention group but not in the control group, suggesting that parents became better at coping with the uncertainty. Maternal ratings of their child's behaviour also improved in the intervention group. Identifying ways to incorporate support for coping with uncertainty into routine care would be an important way of supporting parents to care for their child with JIA.

This study had a number of limitations. Only one father was recruited to the study; the absence of fathers in research about children's illness is widely recognized (40). We recruited by advertising for volunteers through an arthritis charity, but it may be that a more targeted approach is required to ensure that fathers are recruited. Ours was a highly educated sample who may be better equipped than many to navigate health and other services relating to chronic illness. For example, although only two parents spoke about uncertainty relating to claiming disability benefits, others may require more support with this issue. A majority of parents in the current study had children with JIA who were less than 12 years old. Fedele et al (23) found that the association between parental uncertainty and parental distress was greater in parents of older children and that parental uncertainty was related to increased child depressive symptoms in older, but not younger, children. It may be that other issues around uncertainty would have been identified if we had recruited more parents of older children. As this is a qualitative study with 20 parents, we were not able to examine the strength of the relationship between uncertainty and outcomes such as psychological wellbeing. To do so will require the development of a measure that captures the breadth of parental uncertainty identified in the current study.

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All authors were involved in drafting the article or revising it critically for important intellectual content, and all authors approved the final version to be published. Dr. Mulligan had full access to all of the data in the study and takes responsibility for the integrity of the data and the accuracy of the data analysis.

Study conception and design. Mulligan, Newman.

Acquisition of data. Pearce.

Analysis and interpretation of data. Pearce, Mulligan.

REFERENCES

1. Thierry S, Fautrel B, Lemelle I, Guillemin F. Prevalence and incidence of juvenile idiopathic arthritis: a systematic review. *Joint Bone Spine* 2014;81:112–7.
2. Petty RE, Southwood TR, Manners P, Baum J, Glass DN, Goldenberg J, et al. International League of Associations for Rheumatology classification of juvenile idiopathic arthritis: second revision, Edmonton, 2001. *J Rheumatol* 2004;31:390–2.
3. Petty RE, Laxer RM, Wedderburn LR. Juvenile idiopathic arthritis. In: Petty RE, Lindsley RM, Wedderburn LR, editors. *Textbook of pediatric rheumatology*. Amsterdam: Elsevier; 2016. p. 188–204.e6.
4. Martini A. It is time to rethink juvenile idiopathic arthritis classification and nomenclature. *Ann Rheum Dis* 2012;71:1437–9.
5. Martini A, Ravelli A, Avcin T, Beresford M, Burgos-Vargas R, Cuttica R, et al. Toward new classification criteria for juvenile idiopathic arthritis: first steps, Pediatric Rheumatology International Trials Organization international consensus. *J Rheumatol* 2019;46:190–7.
6. Selvaag AM, Aulie HA, Lilleby V, Flato B. Disease progression into adulthood and predictors of long-term active disease in juvenile idiopathic arthritis. *Ann Rheum Dis* 2016;75:190–5.
7. Nugent J, Ruperto N, Grainger J, Machado C, Sawhney S, Baildam E, et al. The British version of the Childhood Health Assessment Questionnaire (CHAQ) and the Child Health Questionnaire (CHQ). *Clin Exp Rheumatol* 2001;19 Suppl 23:S163–7.
8. Shoop-Worrall SJ, Kearsley-Fleet L, Thomson W, Verstappen SM, Hyrich KL. How common is remission in juvenile idiopathic arthritis: a systematic review. *Semin Arthritis Rheum* 2017;47:331–7.
9. Mulligan K, Wedderburn LR, Newman S. The experience of taking methotrexate for juvenile idiopathic arthritis: results of a cross-sectional survey with children and young people. *Pediatr Rheumatol Online J* 2015;13:58.
10. Mulligan K, Kassoumeri L, Etheridge A, Moncrieffe H, Wedderburn LR, Newman S. Mothers' reports of the difficulties that their children experience in taking methotrexate for juvenile idiopathic arthritis and how these impact on quality of life. *Pediatr Rheumatol Online J* 2013;11:23.
11. Fair DC, et al. Depression And Anxiety In Patients With Juvenile Idiopathic Arthritis: Current Insights And Impact On Quality Of Life, A Systematic Review. *Open Access Rheumatol*, 2019. 11: p. 237–252.
12. Mishel MH. Uncertainty in illness. *Image J Nurs Sch* 1988;20:225–32.
13. Gomez-Ramirez O, Gibbon M, Berard R, Jurencak R, Green J, Tucker L, et al. A recurring rollercoaster ride: a qualitative study of the emotional experiences of parents of children with juvenile idiopathic arthritis. *Pediatr Rheumatol Online J* 2016;14:13.
14. Cousino MK, Hazen RA. Parenting stress among caregivers of children with chronic illness: a systematic review. *J Pediatr Psychol* 2013;38:809–28.
15. Johnson LM, Zautra AJ, Davis MC. The role of illness uncertainty on coping with fibromyalgia symptoms. *Health Psychol* 2006;25:696–703.

16. Eastwood JA, Doering L, Roper J, Hays RD. Uncertainty and health-related quality of life 1 year after coronary angiography. *Am J Crit Care* 2008;17:232–42; quiz 243.
17. McCormick KM, Naimark BJ, Tate RB. Uncertainty, symptom distress, anxiety, and functional status in patients awaiting coronary artery bypass surgery. *Heart Lung* 2006;35:34–45.
18. Van Pelt JC, Mullins LL, Carpentier MY, Wolfe-Christensen C. Brief report: illness uncertainty and dispositional self-focus in adolescents and young adults with childhood-onset asthma. *J Pediatr Psychol* 2006;31:840–5.
19. Maikranz JM, Steele RG, Dreyer ML, Stratman AC, Bovaird JA. The relationship of hope and illness-related uncertainty to emotional adjustment and adherence among pediatric renal and liver transplant recipients. *J Pediatr Psychol* 2007;32:571–81.
20. Waldron-Perrine B, Rapport LJ, Ryan KA, Harper KT. Predictors of life satisfaction among caregivers of individuals with multiple sclerosis. *Clin Neuropsychol* 2009;23:462–78.
21. Szulcowski L, Mullins LL, Bidwell SL, Eddington AR, Pai AL. Meta-analysis: caregiver and youth uncertainty in pediatric chronic illness. *J Pediatr Psychol* 2017;42:395–421.
22. Chaney JM, Gamwell KL, Baraldi AN, Ramsey RR, Cushing CC, Mullins AJ, et al. Parent perceptions of illness uncertainty and child depressive symptoms in juvenile rheumatic diseases: examining caregiver demand and parent distress as mediators. *J Pediatr Psychol* 2016;41:941–51.
23. Fedele DA, Ramsey RR, Ryan JL, Bonner MS, Mullins LL, Jarvis JN, et al. The association of illness uncertainty to parent and youth adjustment in juvenile rheumatic diseases: effect of youth age. *J Dev Behav Pediatr* 2011;32:361–7.
24. Cleanthous S, et al. What constitutes uncertainty in systemic lupus erythematosus and rheumatoid arthritis? [Reports primary research]. *Psychol Health* 2013;28(2):171–88.
25. Hurt CS, Cleanthous S, Newman SP. Further explorations of illness uncertainty: carers' experiences of Parkinson's disease. *Psychol Health* 2017;32:549–66.
26. Braun V, Clarke V. Using thematic analysis in psychology. *Qualitative Research in Psychology* 2006;3:24.
27. Mishel MH. Parents' perception of uncertainty concerning their hospitalized child. *Nurs Res* 1983;32:324–30.
28. Corbin JM, Strauss A. *Unending work and care: managing chronic illness at home*. San Francisco: Jossey-Bass; 1988.
29. McErlane F, Foster HE, Carrasco R, Baidam EM, Chieng SE, Davidson JE, et al. Trends in paediatric rheumatology referral times and disease activity indices over a ten-year period among children and young people with juvenile idiopathic arthritis: results from the childhood arthritis prospective study. *Rheumatology* 2016;55:1225–34.
30. WORD day. About WORD day. URL: <https://wordday.org/about-word-day/>.
31. Hogan TP, Brashers DE. The theory of communication and uncertainty management: implications from the wider realm of information behavior. In: Afifi TD, Afifi WA, editors. *Uncertainty, information management, and disclosure decisions: theories and applications*. New York: Routledge/Taylor & Francis Group; 2009. p. 45–66.
32. Barbour JB, Rintamaki LS, Ramsey JA, Brashers DE. Avoiding health information. *J Health Commun* 2012;17:212–29.
33. Parsons S, Thomson W, Cresswell K, Starling B, McDonagh JE. Barbara Ansell National Network for Adolescent Rheumatology. What do young people with rheumatic disease believe to be important to research about their condition? A UK-wide study. *Pediatr Rheumatol Online J* 2017;15:53.
34. Kerr AM, Haas SM. Parental uncertainty in illness: managing uncertainty surrounding an "orphan" illness. *J Pediatr Nurs* 2014;29:393–400.
35. Kerr AM, Harrington NG, Scott AM. Uncertainty management and decision making: parents' experiences during their first visit to a multidisciplinary clinic for their child's vascular anomaly. *J Pediatr Nurs* 2020;52:18–24.
36. Hoefel L, Lewis KB, O'Connor A, Stacey D. 20th anniversary update of the Ottawa Decision Support Framework: Part 2 subanalysis of a systematic review of patient decision aids. *Med Decis Making* 2020;40:522–39.
37. Fedele DA, Hullmann SE, Chaffin M, Kenner C, Fisher MJ, Kirk K, et al. Impact of a parent-based interdisciplinary intervention for mothers on adjustment in children newly diagnosed with cancer. *J Pediatr Psychol* 2013;38:531–40.
38. Mullins LL, Molzon ES, Suorsa KI, Tackett AP, Pai AL, Chaney JM. Models of resilience: developing psychosocial interventions for parents of children with chronic health conditions. *Family Relations* 2015;64:176–89.
39. Hoff AL, Mullins LL, Gillaspay SR, Page MC. An intervention to decrease uncertainty and distress among parents of children newly diagnosed with diabetes: a pilot study. *Families Systems & Health* 2005;23:329–42.
40. Macfadyen A, Swallow V, Santacroce S, Lambert H. Involving fathers in research. *J Spec Pediatr Nurs* 2011;16:216–9.

Appendix A: Interview topic guide

Thank you very much for your interest in the project and giving your time to take part. The interview will ask about your experiences as a parent of someone with JIA and what you understand and think about JIA. Do you have any questions before we start? I just want to emphasise that you can skip any questions you would prefer not to answer or end the interview at any time you wish. Are you happy to begin?

Can I begin by asking your child's age and which type of JIA he/she has? (prompt with names of the different types of JIA if necessary).

Symptoms and Diagnosis

Could you tell me about when s/he first started showing symptoms?
How was s/he diagnosed with JIA?
What kind of symptoms has s/he experienced since then?
Prompts Is it clear which symptoms are related to JIA? Are symptoms predictable?

Treatment

What treatment is your child receiving for their JIA? (prompt with different types of treatment e.g. medication, joint injections, physiotherapy).
Prompts Understanding of how the treatment manages their JIA? Any concerns about the treatment/healthcare they receive?

Impact

How do you think JIA has affected your child? Might it change in the future?
Prompts Physically, socially, emotionally? (school, friendship groups, family relations, growth, ability to participate in sports, activities, mood)
How has your child's JIA affected you? Might it change in the future?

Self-management

[Depending on age of child] What kind of things can your child do to help manage their JIA themselves? Are there things they should avoid doing?
Can you tell me about your role in caring for a child/young person with arthritis?
Prompts How easy/difficult do you find it? How confident do you feel to make decisions about your child's care?

Social support

Do you receive support from others? Is it reliable?

Do you talk about your child's JIA to others/friends/family?

How did your family respond to your child being diagnosed with JIA?

Information and understanding

Where have you obtained most information about JIA? Do you feel you receive enough information? Are there aspects that you feel uncertain about?

Overall, how well do you feel that you understand your child's arthritis?
...and their treatment?

That is all of the questions I had for you – are there any other aspects of having a child with JIA that we haven't discussed that are important?

Thank you very much for taking part.