



What matters most to pediatric rheumatologists in deciding whether to discontinue biologics in a child with juvenile idiopathic arthritis? A best-worst scaling survey

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Abstract

Objectives Care for JIA patients has been transformed in the biologics era; however, biologics carry important (although rare) risks and are costly. Flares after biological withdrawal are seen frequently, yet there is little clinical guidance to identify which patients in clinical remission can safely have their biologic discontinued (by stopping or tapering). We examined what characteristics of the child or their context are important to pediatric rheumatologists when making the decision to discuss withdrawal of biologics.

Methods We conducted a survey including a best-worst scaling (BWS) exercise in pediatric rheumatologists who are part of the UCAN CAN-DU network to assess the relative importance of 14 previously identified characteristics. A balanced incomplete block design was used to generate choice tasks. Respondents evaluated 14 choice sets of 5 characteristics of a child with JIA and identified for each set which was the most and least important in the decision to offer withdrawal. Results were analyzed using conditional logit regression.

Results Fifty-one (out of 79) pediatric rheumatologists participated (response rate 65%). The three most important characteristics were how challenging it was to achieve remission, history of established joint damage, and time spent in remission. The three least important characteristics were history of temporomandibular joint involvement, accessibility of biologics, and the patient's age.

Conclusions These findings give quantitative insight about factors important to pediatric rheumatologists' decision-making about biologic withdrawal. In addition to high quality clinical evidence, further research is needed to understand the perspective of patients and families to inform shared decision-making about biologic withdrawal for JIA patients with clinically inactive disease.

Key Points

- *What is already known on this topic—there is limited clinical guidance for pediatric rheumatologists in making decisions about biologic withdrawal for patients with juvenile idiopathic arthritis who are in clinical remission.*
- *What this study adds—this study quantitatively examined what characteristic of the child in clinical remission, or of their context, are most important to pediatric rheumatologists in deciding whether to offer withdrawal of biologics.*
- *How this study might affect research, practice or policy—understanding of these characteristics can provide useful information to other pediatric rheumatologists in making their decisions, and may guide areas to focus on for future research.*

Keywords Arthritis, Juvenile · Biological therapy · Economics

Introduction

The advent of biologic treatments for juvenile idiopathic arthritis (JIA) has changed the nature of living with JIA with early diagnosis and appropriate treatment enabling improved

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outcomes for children. These include prevention of permanent joint damage and longer-term disability and attaining clinical remission/inactive disease [1, 2]. However, these treatments are not without risks and they are costly whether from a system or individual perspective [1]. Children who have achieved clinical remission could potentially discontinue biologic therapy, but with the risk of flare of disease and the risk of not being able to recapture inactive disease with the same medication [3, 4]. A recent systematic review concluded that there is limited clinical evidence to guide discontinuation of biologics (either by stopping medication or tapering medication over time) in children with JIA and how to balance the associated risks and benefits [1]. In the absence of clinical evidence, the decision to offer withdrawal is guided by pediatric rheumatologists based on their clinical experience about when it would be appropriate, and clinical practice and approaches vary [5–7]. Our previous qualitative research identified factors important to pediatric rheumatologists making this decision including clinical factors like time spent in remission, history of flares, history of joint damage, comorbidities, and contextual factors such as willingness of the patient/family to discontinue and access to biologics [5].

This paper examines the relative importance of these patient and contextual factors to pediatric rheumatologists in their decisions to offer discontinuation (tapering or stopping) of biologics to patients in clinical remission.

Materials and methods

We investigated the views of pediatric rheumatologists on factors (attributes) affecting their decision to offer discontinuation for a child on biologics using a survey. The survey included a stated preference ranking methodology that is now used widely in health care,

including in rheumatology [8, 9]. The best-worst scaling case 1 (object case) was selected as a method to rank attributes in order of importance because avoids the known methodological limitations of rating scales or direct ranking exercises [10] and creates simpler ranking tasks for the respondent. BWS also is not influenced by scale-related biases and "... provides insight into trade-offs which reflect actual decisions in daily clinical practice [8, 10]." Fourteen attributes (see Table 1) were identified in formative qualitative research [5], following the best practices for the development of the best-worst scaling and other preference studies [11].

The pediatric rheumatologists from Canada and the Netherlands who are part of the "Canada-Netherlands Personalized Medicine Network in Childhood Arthritis and Rheumatic Diseases" (UCAN CAN-DU) were invited to participate in this online survey administered using the Qualtrics survey platform. Consent to participate was indicated by the action of advancing through to the survey from a consent page, and the study was approved by both the University of Calgary Conjoint Health Research Ethics Board (REB19-0360) and University of Twente (the Netherlands) BMS Ethics Committee (#20071). There was no direct patient involvement in this component of the research.

A balanced incomplete block design was used to generate 14 choice sets each including 5 different attributes [10]. Each choice set asks the respondents to consider a patient with JIA currently taking biologic medications who has achieved clinically inactive disease (according to the Wallace criteria [12]) and to identify which attributes are the most and least important to them in deciding whether to offer to withdraw (stop or taper) the child's biologic medication. Figure 1 shows an example choice task.

In addition to the BWS questions, a direct question elicited the most and least important of the 14 attributes and also asked if any attributes of the decision were missing that respondents felt were important. Demographic information and information

Table 1 Patient or contextual characteristics

Attribute 1: history of joint damage or erosive disease
Attribute 2: how challenging it was to achieve remission (e.g., number of medications or time needed to achieve remission)
Attribute 3: time spent in remission
Attribute 4: JIA subtype
Attribute 5: history of uveitis
Attribute 6: history of spine/SI involvement
Attribute 7: history of temporomandibular joint (TMJ) involvement
Attribute 8: history of comorbidities (e.g., inflammatory bowel disease (IBD), psoriasis)*
Attribute 9: patient age
Attribute 10: patient willingness to taper/stop
Attribute 11: parent willingness to taper/stop
Attribute 12: history of flares
Attribute 13: continuity of care and ability to access follow-up care (e.g., geographical limitations)
Attribute 14: accessibility of biologics

*The patient meets criteria for diagnosis of JIA and has a history of other comorbidities

on current practice for discontinuation of biologics (e.g., stop immediately, approaches to tapering) were also collected.

The raw best and worst counts as well as best-minus-worst counts are reported for each attribute. We conducted a conditional logit analysis of the best and worst choice data resulting in odd ratios (with confidence interval) relative to the reference attribute (accessibility of biologics), which provide ordering of importance. In exploratory analysis, we examined whether preferences differ between the two countries by estimating the conditional logit estimates per country.

Results

Participant characteristics (Table 2)

In total, 51/79 (65%) of participants completed the survey. Response rate was 63% (40/63) in Canada and 69% (11/16) in the Netherlands. Participants were 69% females, 76% were over 40 years of age, 78% from Canada, 56% were in practice between 6 and 20 years and spent on average 65% of their time in clinical work.

Current tapering approaches used by participants

The most common approach reported by 59% of participants was to increase the interval between medication doses until eventually deciding to stop. Only 8% would stop immediately. Another 12% would reduce dose and increase interval and maintain the new dose/interval, 8% would reduce dose and increase interval to eventually stop, and 4% would increase the interval and maintain the longer interval. Ten percent indicated “other” approaches which included different approaches depending on the biologic (infusions versus injections), as well as an approach to maintaining dose for younger kids as they grow and gain weight. The most common time-period over which tapering was implemented was 6–12 months (37% of respondents), and another 31% would taper over 2–6 months. Ten percent would taper over 12–24 months, and 14% selected “other” approaches. Stopping immediately was reported by 10% of the Canadian respondents, but none of those in the Netherlands reported this approach. In terms of both approach to withdrawal of biologics

Table 2 Survey respondent characteristics (n = 51)

	Overall (n = 51)	Canada (n = 40)	Netherlands (n = 11)
Gender			
Female (n, %)	35 (69%)	28 (70%)	7 (64%)
Age category (years)			
≤30	0 (0%)	0 (0%)	0 (0%)
31–40	12 (24%)	10 (25%)	2 (18%)
41–50	22 (43%)	6 (15%)	7 (64%)
>50	17 (33%)	15 (38%)	2 (18%)
Country			
Canada	40 (78%)		
Netherlands	11 (22%)		
Years in practice			
≤5 years	9 (18%)	6 (15%)	3 (27%)
6–10 years	14 (27%)	12 (30%)	2 (18%)
11–20 years	15 (29%)	11 (28%)	4 (36%)
21–30 years	8 (16%)	6 (15%)	2 (18%)
>30 years	5 (10%)	5 (13%)	0 (0%)
Percentage of time for clinical work (median, range)	65% (0–100%)	65% (0–100)	70% (40–80%)

and to the time interval for tapering, there was similarities in the most common approach between the two countries, and there was less variation in the Netherlands compared to Canada.

Analysis of the best-worst scaling choice tasks (Table 3)

The attributes have been ordered by their parameter coefficients (odd ratios) indicating their order of importance. All coefficients are significantly different from the reference category of accessibility of biologics except for history of TMJ. The most important three attributes were [1] how challenging it was to achieve remission, [2] history of joint damage, and [3] time spent in remission with much larger odd ratios than the other attributes. The odd ratios for most important attribute is statistically different from the second and third; however, the second and third are not statistically different from each other. The third most important is different from the fourth and all the other attributes. The least important three are history of TMJ, accessibility

Fig. 1 Example choice task for the BWS survey

Please indicate what is the most important and least important patient or contextual characteristic in your decision to taper or stop biologic medications?

MOST important (select only one)	Patient or contextual characteristic	LEAST important (select only one)
	Accessibility of biologics	
	History of joint damage or erosive disease	
	Time spent in remission	
	JIA subtype	
	Spine/SI involvement	

of biologics (reference category) and age, and age is statistically different from the others. The top three and bottom three are in alignment with the direct questions about the most important and least important attributes (final two columns in Table 3).

Exploring differences in attribute coefficients across countries

The number of respondents is too small for the Netherlands sample, to allow for a full analysis of the differences in attributes across countries. However, we did examine these differences in an exploratory only analysis. The coefficients by country are reported in Table 4. Figure 2 plots the coefficients estimated from the two countries separately and show that while many coefficient estimates between the countries are similar, there are some areas of divergence (history of uveitis, continuity of care/accessibility of follow-up and history of comorbidities).

Other factors that were important

Respondents were asked and identified other factors important to the decision to discontinue biologics. Nineteen of the 51 participants did identify other factors. These were grouped into the following themes: prior macrophage activation syndrome (for systemic JIA), whether the child was RF+ (which determined whether subtype mattered), side effects, adherence or difficulties with medications, and timing of considering withdrawal (around transition to adult care or to a new provider, life events or recreational goals).

Discussion

This is the first study to quantify what is most important in pediatric rheumatologist decisions to withdraw biologic treatment. Some key clinical components, such as difficulty in achieving remission, history of joint damage, and time spent in remission were important to the pediatric rheumatologist

Table 3 Order of importance of patient and contextual factors when deciding whether to withdraw biologics

	Best count ¹	Worst count ¹	B-W count ¹	Odd ratio (95% CI)	Chosen best in direct question n, (%) ²	Chosen worst in direct question n, (%) ³
How challenging to achieve remission	160	1	159	65.3 (44.4–96.2)	19 (38%)	0 (0%)
History of joint damage	131	2	129	34.4 (23.4–50.5)	8 (16%)	0 (0%)
Time spent in remission	114	9	105	26.1 (17.8–38.3)	16 (32%)	0 (0%)
History of flares	73	9	64	13.9 9.5–20.3	3 (6%)	0 (0%)
History of uveitis	57	18	39	9.1 6.3–13.3	0 (0%)	0 (0%)
Patient willingness to stop	49	27	22	5.5 3.9–7.9	2 (4%)	1 (2%)
History of spine/SI involvement	41	36	5	4.5 3.1–6.4	0 (0%)	0 (0%)
History of comorbidities	20	46	–26	3.4 2.4–4.8	0 (0%)	1 (2%)
Parent willingness to stop	19	33	–14	3.1 2.1–4.4	0 (0%)	2 (3.9%)
JIA subtype	31	71	–40	2.9 2.1–4.2	1 (2%)	4 (7.8%)
Continuity of care/access to followup	16	71	–55	1.9 1.3–2.7	1 (2%)	5 (9.8%)
History of TMJ	3	108	–105	1.1 0.8–1.6	0 (0%)	5 (9.8%)
Accessibility of biologics*	0	92	–92	1 (n/a)	0 (0%)	0 (0%)
Patient age	0	191	–191	0.27 0.19–0.38	0 (0%)	28 (54.9%)

*Reference category, all odds ratios are relative to this category, and represent how important the attribute is relative to the omitted category

¹Best-Worst count is the difference between the number of times the Attribute was chosen as best, less the number of times it was chosen as worst in the best worst scaling 14 questions

²The number of times the Attribute was identified as most important in the direct question (note there is one missing response)

³The number of times the Attribute was identified as least important in the direct question

while deciding if it was the right time to discontinue a biologic. The JIA subtype was not very important, but this could have been due to not mentioning RF status in the attributes used for the best-worst scaling. The age of the patient was the least important factor in the decision to discontinue biologics.

In exploratory analysis, we found that, overall, both the Canadian and Dutch pediatric rheumatologists showed agreement on most clinical features that drive decision making surrounding discontinuing biologics. There may be some differences between Canada and the Netherlands in the rank order of importance of the three factors: history of uveitis or the presence of co-morbidities, and continuity of care/access of follow-up care. These differences in relative importance of the factors may be explained by differences in clinical practice patterns, or in health-care system and geographical differences. These comparative results are exploratory only.

Previous studies have used rating scale methods to look at what is most important to pediatric rheumatologists in decisions to discontinue biologic treatments in children with non-systemic and systemic JIA, respectively [13, 14]. In common with our findings, time spent in remission was a very important for both non-systemic and systemic JIA. Both these previous studies identified a number of other factors that were very important, which is a common challenge with more direct ranking techniques where when asked to simply identify importance in a Likert scale, many factors can be identified as very important.

The strength of this study is the study design using the best-worst scaling methodology which provides a quantitative assessment of the order and the extent of importance to the pediatric rheumatologist of the patient or contextual characteristics. Inclusion of not only patient's clinical attributes but also other factors associated with influencing the decision to discontinue provides broader insight into important drivers of the decision process. Inclusion of the two countries makes the results more transferable. The comparisons between countries are exploratory only as the numbers were limited by the number of pediatric rheumatologists in the Netherlands. Although both Canada and the Netherlands have a comparable universal health care system, there were small differences noted in the exploratory analysis above which is suggestive that local clinical drivers can be important. It is not an unexpected finding that different groups (in this case the two countries) have different rank orderings of the factors. Although the differences make sense in terms of being explainable by differences in the countries, the sample size is too small to fully explore this. The survey was administered online and the response rate may have been affected by the ongoing COVID pandemic and associated overwhelmed health care system, although overall response rates were quite high. This methodology does also have limitations. The BWS methodology identifies the most important characteristics in the decision to withdraw biologics but does not allow one to comment on whether the clinician would or would not discontinue the medication for a specific scenario. The BWS choice tasks responses were hypothetical stated preferences which can be different from revealed preferences (i.e., actual discontinuation

Table 4 Exploratory analysis by country: coefficients for BWS analysis¹

	Canadian coefficient (Std. error)	Dutch coefficient (Std. error)
Attribute 2: how challenging to achieve remission	4.12 (0.22)	5.35 (0.52)
Attribute 1: history of joint damage	3.21 (0.22)	5.60 (0.52)
Attribute 3: time spent in remission	2.94 (0.22)	5.29 (0.52)
Attribute 12: history of flares	2.40 (0.22)	4.21 (0.51)
Attribute 5: history of uveitis	1.64 (0.21)	4.96 (0.51)
Attribute 10: patient willingness to stop	1.42 (0.20)	3.40 (0.50)
Attribute 6: history of spine/SI involvement	1.32 (0.20)	2.65 (0.50)
Attribute 8: history of comorbidities	0.88 (0.20)	3.21 (0.54)
Attribute 11: parent willingness to stop	0.90 (0.21)	2.32 (0.50)
Attribute 4: JIA subtype	0.77 (0.20)	2.75 (0.52)
Attribute 13: continuity of care/access to followup	0.88 (0.21)	-0.20 (0.41)
Attribute 7: history of TMJ	-0.07 (0.20)	1.23 (0.46)
Attribute 14: accessibility of biologics ²	Reference	Reference
Attribute 9: patient age	-1.58 (0.21)	-0.53 (0.37)

¹Note that here we are reporting the coefficients from the conditional logit regression for each country but are not reporting the odds ratios by country because the sample size is too small for the odds ratios to be meaningful for the Dutch sample

²Accessibility of biologics is reference category

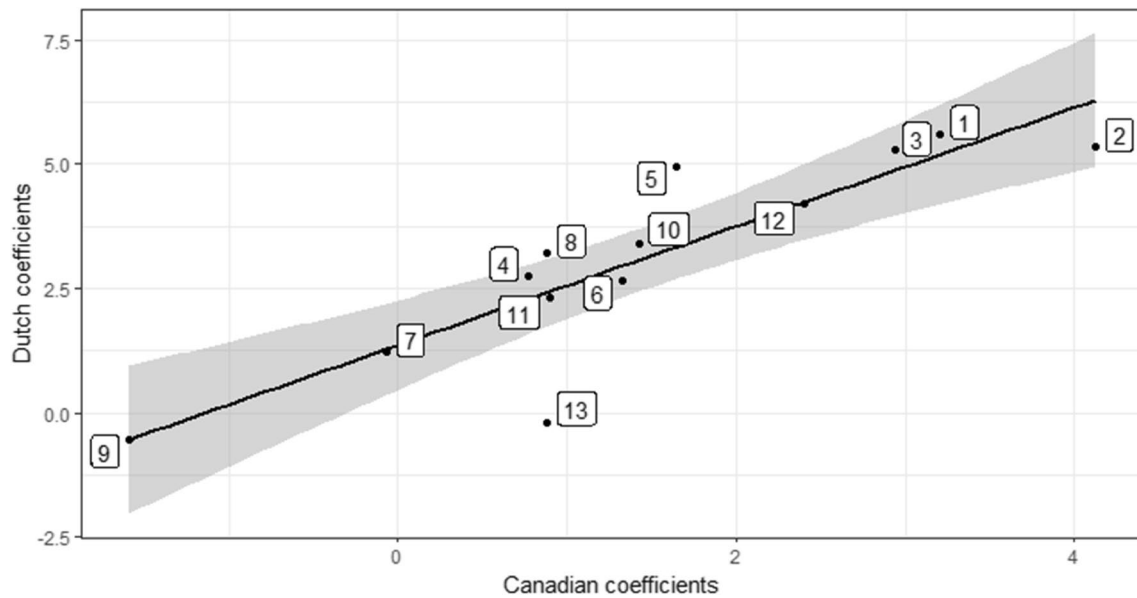


Fig. 2 Comparison of the Canadian and Dutch coefficients for BWS analysis. *Shaded area represents confidence limits. Legend: Attribute 2, How challenging it was to achieve remission; Attribute 3, Time spent in remission; Attribute 4, JIA subtype; Attribute 5, History of uveitis; Attribute 6, History of spine/SI involvement; Attribute 7,

History of TMJ involvement; Attribute 8, History of comorbidities; Attribute 9, Patient age; Attribute 10, Patient willingness to taper/stop; Attribute 11, Parent willingness to taper/stop; Attribute 12, History of flares; Attribute 13, Continuity of care and ability to access follow-up care; Reference category (Attribute 14), Accessibility of biologics

choices that clinicians make in real-life). It also requires that respondents be attentive to the choice tasks and account for the information provided in making their choices. These limitations are mitigated by the standardized and rigorous design and enable a quantitative assessment of preferences that is not possible to always otherwise observe in a real-world setting [10].

In this study, patients and families' views were not assessed. Given the findings about what type of patients that clinicians would offer discontinuation of biologics to, the next step is to examine what approaches to discontinuation would be acceptable to patients and families in the decision to stop or continue biologic medication. Understanding patient/family specific drivers are needed to inform shared-decision making to determine how and when to discontinue a biologic in a patient with JIA in inactive disease.

In conclusion, this study identified the most important patient or contextual factors affecting the decision by the pediatric rheumatologist about whether to suggest discontinuing biologics. This reflects decision-making about biologic discontinuation in the context of current clinical uncertainty and gives pediatric rheumatologists information about how their colleagues are making these decisions. This may reduce variations in practice and inform future tapering strategies, as well as inform priorities for further research. Ultimately, more high-quality clinical evidence about what patients can safely discontinue biologics is needed to inform this decision. In addition, further study of the patient and

family perspective is also needed to help inform shared decision-making about discontinuing biologics and the risk-benefit trade-offs in patients with JIA.

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Author contribution GC and DM were involved in the conception and design of the study. CG-O and GC conducted the data analysis. All authors contributed to data interpretation. GC drafted the manuscript, and all others critically reviewed it. All authors read and approved the final manuscript.

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Data availability The ethics approval for this survey did not include sharing the data beyond the study team; therefore, we are unable to make this data available.

Declarations

Competing interests DM reports grants from Canadian Institutes of Health Research, Alberta innovates, and the Arthritis Society supports for attending meetings from Illumina and ISPOR, and consulting fees from Analytica. All are outside of the submitted work.

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All other authors declare that they gave no competing interests.

Patient involvement There was no direct patient involvement in this component of the research.

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