

Qualitative Patient Interviews to Assess Content Validity of the Modified Dysphagia Symptom Questionnaire

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INTRODUCTION

- Eosinophilic esophagitis (EoE) is an atopic inflammatory disease that is characterized by the significant presence of eosinophils, and damage to and chronic inflammation of the tissue of the esophagus.^{1,2}
- The current prevalence of EoE in North America is estimated to be approximately 57 per 100,000 persons;³ however, there is high variability in the prevalence estimates due to the limited, but growing, awareness of the disease and use of diagnostic codes in routine practice.^{3,4}
- EoE has been shown to cause significant health-related quality of life burden to patients and their families.⁵ EoE is also associated with urgent care, emergency room visits, or inpatient visits for procedures such as dilation and esophagogastroduodenoscopies. In addition, proton-pump inhibitors, topical steroids, elemental diets, and food substitutions are needed to manage the condition.⁵
- The predominant symptom among adolescent (aged 12–17 years) and adult patients with EoE is dysphagia or difficulty swallowing. Some patients may also experience food getting stuck or food impaction.³
- The Dysphagia Symptom Questionnaire (DSQ) is a daily patient-reported outcome (PRO) measure that was developed and validated to assess the frequency and severity of dysphagia related to EoE.^{6,7}
- Content validity of the DSQ has been previously documented: qualitative interviews with adults and adolescents with EoE informed its development, relevance, and ease of understanding.⁶
- The daily DSQ score ranges from 0 (no dysphagia) to 6 (dysphagia requiring medical attention to get relief); the daily score is considered missing if the patient has not eaten any solid food since waking up in the morning. The total DSQ score is estimated over a 14-day period. It is calculated by using the sum of a minimum of eight daily DSQ scores across the 14 days, and divided by the number of non-missing daily DSQ scores multiplied by 14; total scores can range between 0–84.
- In the original DSQ,⁶ participants' response of "no" to question 1 ("Since you woke up this morning, did you eat solid food?") resulted in the participants terminating the DSQ for that day without any score assignment. As a response of "no" to question 1 may result in data that may not be missing at random, a new follow-up question (Q1A) was added to assess whether patients did not eat solid food since waking up in the morning because of their EoE (Table 1).

Question number ^a	Response option	Scoring
1. Since you woke up this morning, did you eat solid food?	No	No score assigned
	Yes	
1a. Please select the reason for not eating solid food since you woke up this morning	Because of your problems with swallowing solid food	To be informed by qualitative patient interviews
	Because of a reason NOT related to your problems with swallowing solid food	
2. Since you woke up this morning, has food gone down slowly or been stuck in your throat?	No	0
	Yes	2
3. For the most difficult time you had swallowing food today (during the past 24 hours), did you have to do anything to make the food go down or to get relief?	No, it got better or cleared up on its own	0
	Yes, I had to drink liquid to get relief	1
	Yes, I had to cough and/or gag to get relief	2
	Yes, I had to vomit to get relief	3
	Yes, I had to seek medical attention to get relief	4

^aQuestion 4 ("What was the worst pain you had while swallowing food over the past 24 hours?") measures pain related to swallowing and is not included in the daily DSQ score calculation. DSQ, Dysphagia Symptom Questionnaire. Modified from DSQ v 4.0. J. Patient-Reported Outcomes (2017) 1:3 © S. Hudgens, C. Evans, E. Phillips and M. Hill, licensed under Creative Commons 4.0 <http://creativecommons.org/licenses/by/4.0>.

OBJECTIVES

- Objective 1:** To confirm that the newly added follow-up question (Q1A) is relevant to, and well understood by, participants with EoE.
 - Objective 2:** To assess whether participants' response to Q1A, indicating that they did not eat solid food because of their problems with swallowing food, can be interpreted as a proxy for EoE disease severity.
 - Objective 3:** To inform scoring of question Q1A on days when participants respond by indicating that they did not eat solid food because of their problems with swallowing solid food.
- ## METHODS
- Cognitive debriefing interviews were conducted using a semi-structured interview guide.
 - Institutional Review Board approval was granted for the study; participants completed an informed consent form and interviews were conducted either in-person or via telephone. The cognitive interviews were recorded and transcribed for analysis.
 - Participants were identified from three sites in the United States.
 - Eligible participants (aged 12 years and above) had diagnosed EoE and ≥1 dysphagia event during the past 2 weeks (at the time of screening) that at least required drinking liquids, coughing or gagging, vomiting, or medical attention to get relief.
 - The cognitive debriefing interview guide included probing questions on the DSQ items and newly added Q1A.
 - In assessing participants' understanding and relevance of Q1A (Objective 1), participants were also asked about the recall period on the questionnaire and any suggestions for changes to the wording of the questions.
 - In assessing patient experience and association of Q1A to disease severity (Objective 2), participants were asked to describe the situations in which they had avoided solid food due to their problems with swallowing.
 - To inform scoring of Q1A (Objective 3), two approaches were used to understand patients' experience of not eating solid food:
 - Approach 1: participants were asked whether they would avoid solid food if they anticipated that the solid food would get stuck in their throat, requiring them to drink liquids, cough/gag, vomit, or seek medical attention to get relief. As each response option for Q3 on the DSQ represents increasing levels of dysphagia severity, the goal was to understand at what point participants say that they would prefer to avoid solid food entirely rather than having to take action to get relief from dysphagia.
 - Approach 2: participants were asked to recall an episode when they avoided eating solid food and were then asked how long it took for them to resume eating solid foods. This information provided insights on how long participants typically go without eating solid food because of EoE.

RESULTS

- Twenty-four participants with EoE were interviewed, including six adolescents (mean age 14.5 [±2.1] years) and 18 adults (mean age 28.3 [±9.0] years); 54.2% of the full sample were female (Table 2) and 79.2% were White.
- Mean number of dysphagia episodes in the past 2 weeks were 5.7 (±3.1) for the adolescent respondents and 7.3 (±4.3) for the adult respondents, suggesting that participants were highly symptomatic.

Understanding and relevance of Q1A

- All participants reported that they understood what Q1A was asking and most participants said that Q1A was relevant to their experience of EoE (Table 3).
- No major issues were identified with the response options or recall period.

Association of Q1A with disease severity

- Participants mentioned that they may not eat solid food on some days, either due to fear of potential food getting stuck/impaction, and/or during recovery from a recent stressful dysphagia event (Table 3).
- Participants indicated that they generally avoided solid food because of a recent dysphagia event that made it uncomfortable (eg, nausea, throat felt tight) for participants to eat solid food (Table 3).

Table 2. Demographic and clinical data of respondents

Demographic variable	Adolescents 12–17 years (n=6)	Adult ≥18 years (n=18)	Total sample (n=24)
Age, years, mean (SD)	14.5 (2.1)	28.3 (9.0)	24.9 (9.9)
Gender, n (%)			
Female	5 (83.3)	8 (44.4)	13 (54.2)
Male	1 (16.7)	10 (55.6)	11 (45.8)
Enrolled in school, n (%)			
Middle/junior high	1 (25.0)	0	1 (25.0)
High school	3 (75.0)	0	3 (75.0)
Education, n (%)			
Did not complete high school	1 (50.0)	0	1 (5.0)
High school diploma	0	3 (16.7)	3 (15.0)
Some college or certification program	0	4 (22.2)	4 (20.0)
College or university degree	0	6 (33.3)	6 (30.0)
Graduate degree	0	4 (22.2)	4 (20.0)
Other	1 (50.0)	1 (5.6)	2 (10.0)
Dysphagia episodes in past 2 weeks, mean (SD)	5.7 (3.1)	7.3 (4.3)	6.8 (3.9)

SD, standard deviation.

Table 3. Excerpts from participant interviews

Understanding and relevance of Q1A	
Understanding: <i>"Uh the question's asking, it wants clarification for, for why you didn't eat solid food. And it wants to know whether it, it was because you have trouble swallowing, like related, uh related to the EoE, or if it was something unrelated, like if you were fasting or something, really busy and you didn't get to just eat or something". (Participant 01-007-A)</i> <i>"Um, it just ... if you selected no it was just trying to figure out why you hadn't tried to eat solid foods that day". (Participant 01-001-P)</i> <i>"It's asking you if you haven't eaten solid food since you've woken up, why?... I think the last answer covers a lot of ... the rest, because something not related, which is like, everything else. Probably". (Participant 01-009-P)</i> <i>"Um, if the problem of swallowing food itself is the reason why I haven't eaten solid food or if there is a separate reason altogether". (Participant 05-004-A)</i>	Relevance: <i>"Um, did your symptoms keep you away from eating food or just for other reason not to eat food when you woke up?; this question is a little bit important, a little bit more important than the first question. Like, if I said no, I'm glad I had so much to elaborate". (Participant 01-002-A)</i> <i>"For me personally, I think it's [referring to Q1A] pretty good because if I selected no for the first answer, I think it's good for you guys to know why". (Participant 01-009-P)</i> <i>"For me personally, um, well I usually do eat solids, so there are very few days that I cannot eat solids, so for me it probably wouldn't be super relevant, but for a lot of people I think". (Participant 01-008-A)</i>
Association of Q1A to disease severity	
<i>"I would say like I eat solid food uh almost every day, well every day, but there were, there was a point where I would, had really bad eating... That like for about a week and a half straight I didn't eat solid food". (Participant 01-007-A)</i> <i>"Sometimes I get nauseous in the thought of food when I wake up, and other times it's just like I can't handle it because I had a rough, a rough night with my EoE beforehand... So sometimes you just want to skip (solid food) and try to give your, give your stomach time to reset, you know". (Participant 01-002-A)</i> <i>"For whatever reason, the day ... like the day before, I just had a hard time swallowing and it had gotten stuck really bad, and so the next day I just took it easy and didn't eat solid foods". (Participant 05-009-A)</i>	<i>"And some people avoid solid food because they're afraid that something's going to happen. Some people just can't get it down, some people, you know ... I don't know. There are lots of different reasons". [The interviewer asked participant if he/she had been in a situation when they avoided solid food]. "Yes, yes, yes. If I'm in a flare, and I know it's going to get stuck, I will not [eat solid food]". (Participant 01-008-A)</i> <i>"I actually have avoided eating solid food because either my throat feels tight or maybe I tried to eat it in the morning and it did go down slowly, and I'm ... It's a scary situation, being able to feel your food go from here down". (Participant 01-006-A)</i> <i>"It'd get stuck". (Participant 05-004-A who was experiencing heightened symptoms on the day of the interview, and when asked what would happen if s/he ate solid food)</i>
Scoring of Q1A	
Approach 1 ("Would you eat solid food if you had to drink liquids, cough, vomit, seek medical attention to get relief?"): <i>"... I used to always have to drink things to get it down". (Participant 01-004-P)</i> <i>"Um, I've never avoided foods if I knew [I would] have to drink liquid to get relief because that's pretty much everything". (Participant 01-011-A)</i> <i>"Probably if I had to vomit [to get relief from dysphagia], then I would avoid eating solid foods". (Participant 01-004-P)</i> <i>"Oh, um, I would draw the line at coughing and-or gagging. Like, that's kind of the point that it's not worth it anymore to try and eat solid food". (Participant 01-006-P)</i> <i>"If I thought I was going to vomit then I would [avoid solid food]". (Participant 01-010-A)</i> <i>"I'm not worried about choking. Um often times like I don't really think about it preemptively. Um it's just more like if I'm eating something and like I have a sev-, like a severe outbreak really quickly, then I would worry about it. So, but I definitely avoid foods that I know are going to give me that reaction". (Participant 01-007-A)</i>	<i>"Okay. Um, if things are going really badly, I'd be really afraid I'd have ... I'd end up with an impaction that I'd have to go to the hospital for... So that would probably be the biggest one...I'd probably just avoid it mostly because I'd be afraid I'd end up in the hospital. And I don't want to go". (Participant 01-008-A)</i> Approach 2 ("How long until you start eating solid food again?"): <i>"I mean, like maybe like 6 hours". (Participant 05-002-A)</i> <i>"...not for a couple hours at least". (Participant 05-003-A)</i> <i>"I started eating the next day ...it was probably like, 12 hours". (Participant 05-005-A)</i> <i>"Um it will take me 2, 3 hours". (Participant 05-006-A)</i>

Scoring of Q1A

- A subset of participants (n=12) also spontaneously discussed or were asked additional probing questions to understand the situations in which they would skip eating solid food entirely and how long after a dysphagia episode would it take for participants to resume eating solid food.

Approach 1

- The line of questioning via this approach was difficult for some participants to understand, as it asked whether they would avoid eating solid food based on the anticipated outcome of dysphagia.
- However, additional insights were obtained:
 - Some participants mentioned that they routinely drink liquids to help solid food go down. Therefore, having to drink liquids to get relief from a dysphagia episode does not appear to be an anticipated outcome that may prompt patients with EoE to avoid eating solid food entirely (Table 3).
 - The anticipated outcomes of a dysphagia episode that would prompt participants to avoid eating solid food entirely were highly variable. While "felt like having to vomit to get relief from dysphagia" and "coughing and gagging" were mentioned as probable factors leading to food avoidance, no clear threshold that prompted participants to avoid eating solid food could be defined from the responses (Table 3).

Approach 2

- Participants reported that although they may avoid solid food, it was very rare for them to do so for prolonged periods of time because of their EoE. Previous dysphagia episodes may prompt participants to take a break from eating solid food; some participants mentioned that they were comfortable to eat solid food again as early as a couple of hours. In some cases, participants reported avoiding solid foods for longer durations, such as a few days.
 - All of these participants reported that it was generally for a short period of time. Previous dysphagia episodes had prompted participants to take a break from eating solid food, although three participants mentioned that they were back (feeling confident; not feeling worried) eating solid food again within a few hours or the next day.
 - In rare cases, participants reported avoiding solid foods for longer durations, such as a few days, due to fear of vomiting or hospitalization, which reflects the presence of very severe EoE symptoms.

CONCLUSIONS

- The newly added Q1A is relevant, understandable, and can provide useful information on days when patients with EoE do not eat solid food, by asking whether this is related to difficulty with swallowing.
- Through the qualitative interviews, it was noted that participants who were skipping solid food were either afraid of eating because of the expectation of dysphagia or were recovering from a recent dysphagia event. Nonetheless, even in situations when participants had a severe dysphagia episode that required multiple actions to get relief, they generally did not avoid eating solid foods entirely for prolonged periods of time.
- In the context of daily administration of the DSQ, if patients do not eat any solid food since waking up in the morning because of their problems with swallowing solid food, they are likely experiencing severe EoE disease burden, which is prompting them to avoid eating solid foods for prolonged periods of time; this can be inferred as the worst dysphagia severity level.
- Hence, empirical evidence based on qualitative interviews with participants with EoE supports scoring Q1A with the worst daily score (ie, score of 6) on days when patients indicate that they did not eat any solid food since waking up in the morning because of their difficulty with eating solid food.

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Conflicts of interest

Siddhesh Kamat and Jingdong Chao are employees of Regeneron Pharmaceuticals, Inc. and own stock and/or stock options in the company. Laurent Eckert and Isabelle Guillemain are employees of Sanofi and own stock and/or stock options in the company. Christopher Hartford and Tara Symonds are employees of Clinical Outcomes Solutions, a company that received research funding for the current study.