Examining the Relevance of the SF-36v2 Acute for Assessment in HDFN- or FNAIT-affected Pregnancies: Results of a Landscape Assessment and Clinician and Patient Interviews

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BACKGROUND

- Hemolytic disease of the fetus and newborn (HDFN) and fetal and neonatal autoimmune thrombocytopenia (FNAIT) are rare alloimmune conditions in pregnancy caused by antigen incompatibility in blood components between the pregnant individual and their fetus[1,2]
- While the pregnant individual is asymptomatic, both HDFN and FNAIT can lead to extensive fetal and neonatal morbidity if left untreated
- Patient-reported outcome (PRO) instruments are increasingly recognized by regulatory agencies and health technology assessors as important tools to evaluate how patients feel and function as well as the overall impact of a health intervention[3-5]
- A measurement gap exists as there are no disease-specific PRO instruments for assessing the experience of individuals diagnosed with HDFN and FNAIT, and few PROs exist to evaluate asymptomatic high-risk pregnancies
- General health-related quality of life (HRQoL) PRO instruments provide an opportunity to evaluate function and well-being in physical, emotional, and social domains of life in patients with different diseases

OBJECTIVE

- To describe the patient experience in HDFN and FNAIT and to evaluate whether a general HRQoL instrument, the Short Form-36 Health Survey v2 Acute (SF-36v2 Acute), is suitable to measure areas identified as meaningful in these patient populations

METHODS

- A targeted literature review was conducted to identify concepts of importance and to inform qualitative interview guide development
  - A total of 33 records related to HDFN or FNAIT were included in the final literature reviewed
  - Literature insights relied primarily on publicly available blog posts (n = 30) written by individuals who experienced an HDFN- or FNAIT-affected pregnancy
  - In addition, 2 peer-reviewed articles and 1 conference abstract were identified
- Qualitative, semistructured interviews were conducted with maternal-fetal medicine (MFM) specialists who had experience treating HDFN- and FNAIT-affected pregnancies (n = 4) and with participants who had ≥1 pregnancy diagnosed with either HDFN (n = 10) or FNAIT (n = 8; **Table 1**)
- All participants were recruited from the United States via a research partner following institutional review board approval and confirmation of written consent. Interviews were audio recorded and transcribed verbatim. Concept-elicitation data were analyzed using thematic analysis to identify and discover any themes or patterns within the data
- Qualitative insights with MFM specialists and participants describing their experiences during an affected pregnancy were mapped to the items of the SF-36v2 Acute to evaluate conceptual coverage
- Qualitative data were analyzed using NVivo v1.6 (or higher)

TABLE 1: Demographic characteristics

	HDFN (n = 10)	FNAIT (n = 8)
Sex, n (%)		
Female	10 (100)	8 (100)
Age, years		
Mean (SD)	35.5 (4.58)	41.0 (4.84)
Median (Q1, Q3)	35.5 (32.0, 37.0)	40.5 (38.0, 43.5)
Minimum, maximum	30.0, 46.0	34.0, 50.0
Race/ethnicity, n (%)		
Non-Hispanic White or Caucasian	7 (70)	8 (100)
Black or African American	1 (10)	–
Asian or Asian American	2 (20)	–

FNAIT, fetal and neonatal autoimmune thrombocytopenia; HDFN, hemolytic disease of the fetus and newborn; Q1, first quartile; Q3, third quartile; SD, standard deviation.

RESULTS

- Literature insights relied primarily on publicly available blog posts written by individuals who had experienced an HDFN- or FNAIT-affected pregnancy
- Concepts related to the impacts on emotional well-being, physical well-being, and work reported during interviews with MFM specialists and individuals with HDFN- or FNAIT-affected pregnancies mapped most directly to the items of the SF-36v2 Acute (**Figure 1**)

FIGURE 1: Exemplary quotes from interviews with MFM specialists and participants[a]

Emotional impacts:

"I think it's a higher anxiety for the parents because the, um, risks that they face in their fetus is something that could be devastating." (CLIN-004)

"We worried whether the baby would survive." (H-101-NT)

"I just had this, this overwhelming feeling that I'm not going to end this, like this pregnancy is going to end with a stillborn. Like I just…I knew it in my, in my soul that I was not going to have a baby at the end of this. And it was, it was gut wrenching and terrifying. And I kept thinking like he's going to die." (F-104-T)

Physical functioning after treatment:

"…if you're getting, IVIg once a week and it not only knocks out most of that day, but knocks out the next day as well…It's gonna make it hard for you to work and keep a job." (CLIN-003)

"…there's so much going on at once that they find it overwhelming. And that's where…you could get into more extreme fatigue." (CLIN-004)

"You know, um, the days that I got the IVIg, I would just come home and try to wr- wrap up my work day and rest, and then those days were usually the worst days just in terms of just physically and mentally just feeling so tired, um, so tired from it." (H-108-T)

"Um, yeah. So my treated pregnancy, um, I was definitely…The IVIg definitely took a lot out of me. Like, it, it was, it w- it was, it was tough. You know? I was, I was exhausted after my treatments." (F-103-T)

Impact on work:

"…[surveillance] takes a fair commitment and if somebody's working and then and she's gonna come for a weekly visit…that's a lot of time traveling, being in our office, going back to home or to work." (CLIN-001)

"For my ultrasounds, I thankfully didn't need to take off an entire day, but I was gone for at least 2 hours of my workday. Um, obviously nobody does ultrasounds after business hours, so (laughs)…Um, for my procedures, they were a full-day thing. And so I had to take an intermittent leave through, uh, the state that I live in, um, like paid family leave. Um, and so I took, uh, the day of procedures and the day after procedures off, um, to recover." (H-103-T)

"I wasn't sure how I was gonna react to the treatment. So, I wanted them to know if I was sick or something that was what was going on…It did cause me to have to change my schedule a bit regarding office hours for student meetings. Like I had, I had to cancel them and make them…at that time virtual, like this wasn't really an option…So I had to kind of tell them, 'Well, if you need me, we can make arrangements,' but I did have to cancel regular standing office hours." (F-102-T)

FNAIT, fetal and neonatal autoimmune thrombocytopenia; HDFN, hemolytic disease of the fetus and newborn; IVIg, intravenous immunoglobulin; MFM, maternal-fetal medicine.
[a]Quotes from MFM specialists are represented by "CLIN." Quotes from participants are represented by "H" for those with HDFN, "F" for those with FNAIT, "T" for those who received treatment during pregnancy, and "NT" for those who did not receive treatment during pregnancy.

- 25 items of the SF-36v2 Acute were directly supported by 10 concepts reported in interviews (**Table 2**)
- 5 items of the SF-36v2 Acute were supported indirectly by concepts reported during the interviews (eg, relationships with family and friends encompassed a wide range of experiences, both positive and negative, that could be captured by the items)
- Only 6 items, focused on general or overall health, were not discussed by participants or MFM specialists in the interviews

TABLE 2: SF-36v2 Acute item mapping

SF-36v2 Acute domain	SF-36v2 Acute item numbers	Qualitative concepts discussed in interviews	Reported by participants	Reported by MFM specialists
Physical functioning	3 a-j	Reduced physical functioning and mobility Difficulty with self-care	✓	✓
Role limitations, physical and emotional	4 a-d, 5 a-c	Missed work	✓	✓
		Negative changes in performance at work	✓	X
Bodily pain	7, 8	Bodily pain and/or discomfort	✓	X
Vitality	9 a, e, g, i	Fatigue, tiredness, and reduced energy	✓	✓
Social functioning	6, 10	Impact on relationships with family and friends	✓	✓
Mental health	9 b-d, f, h	Anxiety and worry Depression or sadness	✓	✓
General health	1, 2, 11 a-d	–	X	X

MFM, maternal-fetal medicine; SF-36v2 Acute, Short Form-36 Health Survey v2 Acute.

REFERENCES:

1. de Haas M, et al. *Vox Sang*. 2015;109(2):99-113. 2. Strong NK, Eddleman KA. *Clin Lab Med*. 2013;33(2):311-325. 3. US Food and Drug Administration. Patient-focused drug development: selecting, developing or modifying fit-for-purpose clinical outcomes assessments. Accessed April 9, 2024. https://www.fda.gov/regulatory-information/search-fda-guidance-documents/patient-focused-drug-development-selecting-developing-or-modifying-fit-purpose-clinical-outcome. 4. European Medicines Agency. Appendix 2 to the guideline on the evaluation of anticancer medicinal products in man. The use of patient-reported outcome (PRO) measures in oncology studies. Accessed April 9, 2024. https://www.ema.europa.eu/en/appendix-2-guideline-evaluation-anticancer-medicinal-products-man-use-patient-reported-outcome-pro-measures-oncology-studies-scientific-guideline. 5. Angelis A, et al. *Eur J Health Econ*. 2018;19(1):123-152.

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KEY TAKEAWAYS

- Patients with pregnancies affected by HDFN or FNAIT reported impacts across multiple HRQoL domains, including emotional, physical, and social well-being
- Although there are no available disease-specific PRO instruments to assess HRQoL in HDFN- and FNAIT-affected pregnancies, the SF-36v2 Acute captured key impacts mentioned by patients

CONCLUSIONS

- This study provides important context in HDFN and FNAIT, where very little patient experience data are available
- Evidence collected from this study demonstrates that items within the SF-36v2 Acute capture several relevant impacts experienced by individuals during an HDFN- or FNAIT-affected pregnancy
- Results of this study can be used to support the use of the SF-36v2 Acute in future studies to evaluate the impacts of HDFN or FNAIT on HRQoL for pregnant individuals

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DISCLOSURES

KS, MLT, and SP are employees of Janssen Global Services, LLC. AIB, JR, and SH are employees of Clinical Outcomes Solutions, which received funding from Janssen for this analysis. SD is a former employee of Janssen Global Services, LLC.

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