Mixed Methods Research to Understand Disease Burden and Unmet Need in HDFN and FNAIT

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BACKGROUND

- Hemolytic disease of the fetus and newborn (HDFN) is a rare and potentially life-threatening condition that occurs when the blood types of the pregnant adult and the fetus are incompatible and the pregnant adult develops alloantibodies against an antigen on fetal red blood cells. During pregnancy, the maternal alloantibodies cross the placenta and bind to fetal red blood cells, causing destruction and anemia in the fetus or the newborn¹
- Fetal and neonatal autoimmune thrombocytopenia (FNAIT) is a rare disease resulting from human platelet antigen (HPA) incompatibility between a pregnant adult and their fetus. In FNAIT, maternal immunoglobulin G antibodies against fetal HPA cross the placenta and destroy fetal and neonatal platelets, resulting in thrombocytopenia²
- Both HDFN and FNAIT are associated with significant morbidity and mortality for developing fetuses and neonates; however, they are asymptomatic for mothers
- There is a lack of available literature from the patient perspective to describe the disease burden and unmet need of these 2 rare pregnancy-related alloimmune conditions

OBJECTIVE

• To advance the understanding of the impacts associated with the experience of a pregnancy affected by HDFN or FNAIT from the patient perspective

METHODS

• Qualitative, semistructured interviews were conducted with mothers who had ≥1 pregnancy diagnosed with either HDFN (n = 10) or FNAIT (n = 8; **Table 1**)

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

- All participants were recruited from the United States via a research partner following institutional review board approval
- Eligible participants had ≥1 previous pregnancy with a diagnosis of HDFN or FNAIT, were able to complete the interview in English, and were willing and able to give informed consent and to comply with study procedures; there were no exclusion criteria
- The Mother-Generated Index (MGI) was mailed to participants to complete and return in their own time. The MGI is a 3-step patient-reported outcome measure that assesses antenatal and postnatal quality of life. Mothers are asked to record, in a free-text field, up to 8 areas of their lives that have been impacted by having a baby. For the purposes of this study, mothers were instructed to complete the MGI while thinking about their most recently affected pregnancy with HDFN or FNAIT
- Interviews were audio recorded and transcribed verbatim. Transcripts and MGI data were analyzed using thematic analysis methods

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RESULTS

- Participants reported an HDFN- or FNAIT-affected pregnancy encompassed several impacts, including those associated with diagnosis, making time for and tolerating treatments, and uncertainties during the neonatal period
- The most frequently reported impacts of HDFN or FNAIT experienced during pregnancy included negative impacts on emotional well-being (HDFN: n = 10/10, 100%; FNAIT: n = 8/8, 100%), such as anxiety, worry, or fear; physical well-being (HDFN: n = 8/10, 80%; FNAIT: n = 5/7, 71%), such as fatigue, tiredness, and reduced energy levels; and social well-being (HDFN: n = 8/10, 80%; FNAIT: n = 5/7, 71%), with respect to relationships with family and friends (Figures 1-3)
- Concept-level findings for FNAIT were based on a total of 7 participants as 1 participant was diagnosed after giving birth and, therefore, was not able to report impacts during pregnancy

FIGURE 1: Combined disease experience visualization: impacts during pregnancy FIGURE 3: Exen

HDFN and FNAIT: Key impacts during pregnancy				
Emotional well-being	Physical well-being	Future conception planning	Work	
 Anxiety and worry Fear Sadness Helplessness Uncertainty Anger Systeme distance 	 Difficulty with normal exercise routines and activities (moderate/ vigorous) Fatigue, tiredness, and reduced energy levels Bodily pain and/or disconfort 	 Decisions about future pregnancies (both positive and negative) Advice from medical professionals (both positive and negative) 	 Missed work Support at work (HDFN)^a Negative changes in performance at work (HDFN)^a 	
 Guilt Guilt Disconnection from baby Shock Depression Confusion (HDFN)* 	 Difficulty with physical functioning and mobility Difficulty with self-care (HDFN)* 	Impact on family Impact on relationship with partner (both positive and negative) Impact on relationship with other children (both positive and negative) Impact on extended family (negative; HDFN)	Social well-bein - Impact on relationships with family and friends (both positive and negative)	

FNAIT, fetal and neonatal autoimmune thrombocytopenia; HDFN, hemolytic disease of the fetus and newborn Red indicates the concept only emerged during HDFN interviews.

FIGURE 2: Greatest emotional well-being impacts during an HDFN- or FNAIT-affected pregnancy

Greatest impact Participants reported the long-term, persistent impact on emotional well-being, including poor mental health such as uncertainty and stress
Overall impact The impact of HDEN and FNAIT on participants' emotional well-being during regnancy was discussed at length during the ualitative interviews and provided evidence for the importance of this domain

FIGURE 3: Exemplary quotes from participants with HDFN and FNAIT^a

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Anxiety	and worry:	Depress	sion:		
* }	"We were worried about losing the baby at first it was a miscarriage, and then it was second trimester loss, and then it was, 'Oh my gosh, what if she's stilloorn? What if she, uh, becomes anemic or has hydrops, or something like that?" (H-110-NT)	٩	"I mean, I think I got really depressed during the treatment because, you know, it's not like you have family and friends and stuff to come and visit you during that time and it's very lonely and some of the nurses are jerks" (F-107-T)		
Helples	sness:	Grief:			
Soft	"No matter how much knowledge and power you, you have, you have no control over the ultimate outcome." (F-102-T)	:	"I think, yeah it took me over a year, or a year and a half to come out of that grief, um, mentally and, like, you know, emotionally." (H-102-T)		
Extreme distress:		Future conception planning:			
	"I guess, for me, it [extreme distress] meant, like, feeling so overwhelmed that I couldn't really be myself or do my regular, daily duties." (H-105-NT)		"[M]y maternal fetal specialist had pretty much told me it probably wasn't a good idea [to have another baby]. So I don't think I ever really seriously thought about it." (F-105-T)		

FNAT, feal and neonatal autoimmune thrombocytopenia; HDFN, hemolytic disease of the fetus and newborn. "Quotes are represented by "H" for participants with HDFN, "F" for participants with FNAT, "T" for participants who received treatment during pregnancy, and "N" for participants who did not receive treatment during pregnancy.

- Postnatally, participants reported they also face long-term challenges as a result of their experience with HDFN or FNAIT, including decisions regarding whether or not to pursue future pregnancies and persistent emotional well-being impacts
- Participants recorded MGI areas that were important to them, allowing for a deeper understanding of
 what is relevant to pregnant individuals impacted by HDFN or FNAIT. MGI data were consistent with
 and confirmed the results generated by the qualitative interviews, with social well-being and emotional
 well-being impacts indicated as the most relevant to the experiences of participants (Figure 4)

FIGURE 4: Important areas of life affected by HDFN and FNAIT as recorded on the MGI



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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.

KEY TAKEAWAY

Primary research with individuals who had either an HDFN- or FNAIT-affected pregnancy demonstrated the profound impact diagnosis and treatment have on maternal emotional, social, and physical well-being

CONCLUSIONS



The results of this study contextualize significant impacts that mothers experience during and after an HDFN- or FNAIT-affected pregnancy



The qualitative interviews with mothers indicated that several areas of well-being were affected, which were affirmed by MGI data



This research provides important insights into disease areas with limited patient-generated information and direction for future studies on the assessment of health and well-being in these high-risk pregnancies

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DISCLOSURES

SP, MLT, and KS are employees of Janssen Global Services, LLC. PM, SH, and JR 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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