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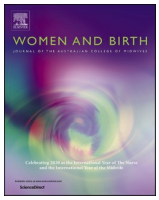

Women- and clinician- important outcomes and priorities regarding vasa praevia: An international qualitative study to inform development of a core outcome set☆

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ABSTRACT

Background: Many studies have reported interventions for women with vasa praevia to improve perinatal outcomes. However, which outcomes are important for women remains unclear.
Aim: To explore what outcomes are important for women with lived experience of vasa praevia and why, in order to inform the development of a core outcome set for studies on vasa praevia.
Methods: An international qualitative study was conducted with women and clinicians. Semi-structured interviews were audio-recorded, transcribed, and analysed taking an inductive approach.
Findings: Eighteen women and six clinicians (four obstetricians, two midwives) from the United States, United Kingdom, Canada, and Australia were interviewed. Participants identified 47 patient-important outcomes and experience measures, which were grouped under five themes: baby's survival and health, mother's physical health, mother's mental and emotional health, quality of health care delivery, and resource use and cost. While survival of the baby without short- and long-term morbidity remained the main priority, other important considerations included the physical, mental, social and financial wellbeing of families, future access to antenatal screening and diagnosis, information on management options and consequences, continuity of care, clear and effective communication, peer support and the appreciation of individual variations to risk tolerance, values and resource availability.
Conclusion: We have identified patient-important outcomes and experience measures that have been directly fed into the development of a core outcome set on vasa previa. Incorporating these considerations into both clinical practice and future research studies has the potential to improve outcomes and experiences for women with vasa praevia.

Statement of Significance

Problem or issue: Despite the profound impact of vasa praevia on the pregnancy and birth outcomes, little is known about what outcomes are important for women who have experienced this condition.

What is already known: Understanding women's perspectives, experiences and needs is pivotal to provision of high-quality maternity care, particularly for those with a high-risk pregnancy.

What this paper adds: Forty-seven outcomes and experience measures considered important for women with a lived experience of vasa praevia and clinicians were identified, which informed development of a core outcome set. The findings may facilitate

Abbreviations: NICU, Neonatal Intensive Care Unit.

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designing meaningful interventions and measuring the success of care.

1. Introduction

Vasa praevia, a condition in which unprotected fetally-derived vessels traverse through the amniotic membranes, in close proximity to the internal cervical os, renders the fetus at high risk of hypovolemic shock at birth if these vessels rupture at birth, and therefore is associated with a high risk of perinatal mortality and morbidity [1,2]. Vasa praevia is estimated to affect 1 per 2200 pregnancies, and its prevalence is increasing. [3] Antenatal diagnosis together with planned caesarean birth to bypass attempted vaginal birth is highly-effective in mediating safe birth and improving perinatal outcomes for this condition. [1,2] A systematic review looking at outcome reporting in studies on vasa praevia identified that not only were these outcomes inconsistently reported and defined, but also outcomes related to life impact, delivery of care or resource utilisation were only reported in 2% (3/160) of the studies. [4]

The inclusion of outcomes that are important for women in clinical studies is essential to enhance woman-centred care [5,6]. Qualitative research provides an opportunity to both identify and amplify women's voices and thereby make pregnancy research more relevant as perceived by women and their families [7,8]. Core outcome sets [9,10] – defined as standardised, essential, minimum sets of outcomes required to be reported in all studies on a particular condition – are useful in this context, and encourage the conduct of qualitative studies to identify and incorporate the outcomes important for women and clinicians into mainstream clinical research [8].

The aim of our study was to identify the outcomes that matter to women with lived experience of vasa previa, to thereby inform the development of a Core Outcome set for studies on Vasa Praevia (COVasP) [9,10], and gain a deeper understanding of why these outcomes are important for women, clinicians, and researchers.

2. Materials and methods

An international qualitative study was conducted with women with lived experience of vasa previa and clinicians with expertise in caring for this population. We first created a sampling matrix to ensure diversity in relation to context, experience and outcomes, and used purposive sampling to provide maximum variation among participants and their experiences, as indicated in our study protocol [9]. Recruitment of women was facilitated through the global network of the International Vasa Previa Foundation (https://vasaprevia.com) and the Vasa Praevia Raising Awareness (https://vasapraevia.co.uk/) in the UK. Clinicians were recruited through authors' professional networks based on location, and professional role at clinical/research/policy level.

Semi-structured, one-on-one interviews were conducted by the first author over the telephone, between August and December 2019. Data collection continued until no new information emerged from two successive interviews [11,12]. The 27–62 (average 44) minute interviews were audio-recorded and transcribed verbatim.

2.1. Data analysis

A descriptive-interpretive data analysis [13] was conducted iteratively, taking an inductive approach. [14] First, an inductive thematic analysis [15] was employed to analyse the interview data. The transcripts were read and coded line-by-line by the first author to identify initial themes and important outcomes. Taking the same approach, 50 % of the data were analysed independently by a second researcher. The codes, outcomes, and themes that emerged were then discussed by the authors to resolve any differences. A constant comparative method of data analysis was used to compare, contrast, and combine the major codes, themes, and outcomes.

Subsequently, the data-driven codes, themes, and outcomes generated by the inductive analysis were deductively grouped according to the taxonomy of outcomes in medical research [16]. The process of data analysis was iterative, as the authors met regularly online during data analysis to develop, discuss, combine, refine, and finalise the major codes, outcomes, and themes. Following the full analysis of the women's transcripts, clinicians' transcripts were analysed. Results were compared by time of diagnosis (antenatal vs postnatal) and patient/clinician role. Any discrepancies were discussed and resolved by the first and last author.

2.2. Ethics statement

The study protocol was approved by the Mount Sinai Hospital Research Ethics Board in Canada (18–0173-E;05/09/2018), and the University of Of Technology Sydney Ethics Committee, Australia (ETH19-3718;30/07/2019). We followed the Standards for Reporting Qualitative Research checklist to report our study findings [17].

3. Results

We interviewed 18 women and six clinicians from the USA (n=9), UK (n=6), Australia (n=5) and Canada (n=4) whose characteristics are summarised in Table 1. Most women were Caucasian (72 %, n=13), had a university or college degree (78 %, n=14), and were in a paid employment (89 %, n=16) at the time of interview. Women had a mean age of 33 and had conceived naturally (89 %, n=16) in the pregnancy complicated by vasa praevia. All nine in whom vasa praevia was diagnosed antenatally had live births by caesarean. Of the nine in whom the diagnosis was not made antenatally, the outcomes included one intrapartum stillbirth at 40 weeks, four early neonatal deaths, and four neonatal near-miss events resulting in neonatal intensive care unit (NICU) admissions ranging from 2 to 10 weeks' duration (Table 2).

Six clinicians participated in interviews, including four obstetricians

Table 1
Participant characteristics (n=24).

Women	N=18 n
Country	
USA	8
UK	5
Australia	3
Canada	2
Age at diagnosis of vasa previa (*y*)	33* (26–40)
25–29	5
30–35	7
35 or older	6
Ethnicity	
Caucasian	13
African American	1
Unknown	4
Education	
High School	4
College	5
University	9
Clinicians	**N=6** n
Country	
Australia	2
Canada	2
United Kingdom	1
United States of America	1
Age at interview (*y*)	56* (49–62)
Profession	
Obstetrician	4
Midwife	2

* Mean (range).

Table 2
Clinical characteristics of women with lived experience of vasa praevia (N=18).

ID	Year of diagnosis	Parity at diagnosis (interview)	Gestational age at hospitalisation (duration) in weeks	Actual and (planned) time of birth in weeks	Mode of birth	Perinatal outcome	Admission and Length of stay at NICU/SCN
Antenatal diagnosis (n=9)							
16	2019	0 (1)	NA *	35^{+4}	Spontaneous vaginal birth	Livebirth	No
11	2018	0 (2)	27 (5)	32^{+4} (36)	Emergency caesarean	Livebirth	34 days
4	2017	0 (1)	35 (2)	36^{+5} (34)	Planned caesarean	Livebirth	48 hours
13	2017	0 (1)	28 (2)	30^{+2} (35)	Emergency caesarean	Livebirth	5 weeks
12	2016	1 (2)	NA#	37^{+3} (37)	Planned caesarean	Livebirth	No
18	2016	1 (2)	33 (3)	36^{+1} (36)	Planned caesarean	Livebirth	1 day
7	2015	0 (1)	30 (5)	35 (35)	Planned caesarean	Livebirth	10 days
1	2011	0 (1)	32 (3)	35 (37)	Emergency caesarean	Livebirth	No
10	2007	0 (1)	26 (4)	30 (32)	Emergency caesarean	Livebirth	7 weeks
Postnatal diagnosis (n=9)							
17	2019	2 (3)	NA	41^{+2}	Instrumental vaginal birth	Neonatal death	Yes
9	2017	1 (1)	NA	38	Emergency caesarean	Neonatal death	Yes
5	2017	1 (1)	NA	37^{+5}	Emergency caesarean	Neonatal near-miss	3 weeks
15	2016	1 (3)	NA	39	Emergency caesarean	Neonatal near-miss	4 weeks
3	2016	1 (2)	NA	39	Emergency caesarean	Neonatal near-miss	2 weeks
8	2006	1 (4)	NA	41	Emergency caesarean	Neonatal death	Yes
2	2006	1 (4)	NA	40	Spontaneous vaginal birth	Intra-partum stillbirth	N/A
14	2004	6 (7)	NA	28	Spontaneous vaginal birth	Neonatal near-miss	10 weeks
6	1996	4 (5)	NA	40	Emergency caesarean	Neonatal death	Yes

SCN , special care nursery; NA, not applicable.
* Vasa praevia was resolved,
# Hopsitalisation was not offered. Patient lived 10 minutes from the hospital.

sub-specialised in maternal fetal medicine and two midwives, who were practising in Canada (n=2), Australia (n=2), UK (n=1) and USA (n=1). The obstetrician's extensive experience in vasa praevia screening, diagnosis and management, research, policy and advocacy together with the midwives' experience in caring for pregnant women with vasa praevia provided substantial breadth and depth. Interviews generated a total of 47 unique important outcomes or experience measures, which in turn were grouped under five themes: baby's survival and health; mother's physical health; mother's mental and emotional health; delivery of care; and resource utilisation and cost (Fig. 1). The themes, subthemes, and outcomes or experience measures considered important for women are presented in Table 3.

3.1. *Baby's survival and health*

All participants reported baby's survival and health as the most important outcome. In addition to fetal/neonatal death from exsanguination, survival, palliation, and withdrawal of care after birth, many referred to short- and long-term health outcomes as important to consider. For example, one woman stated:

> I think overall they [babies] need to be followed to see, when were they born…Developmentally did they hit milestones? Health-wise, do they have any long-lasting issues or is there a trend among these children (W10)

Timing of birth was discussed by all clinicians and women with antenatally-diagnosed vasa praevia. They discussed the trade-offs needed to be considered between an earlier caesarean to prevent serious adverse neonatal outcomes and the risk of the long-term implications of prematurity. Some women felt that their obstetricians recommended very early caesarean because they were *'conservative'* and *'did not want to take the risk'*, even when there was no sign of preterm

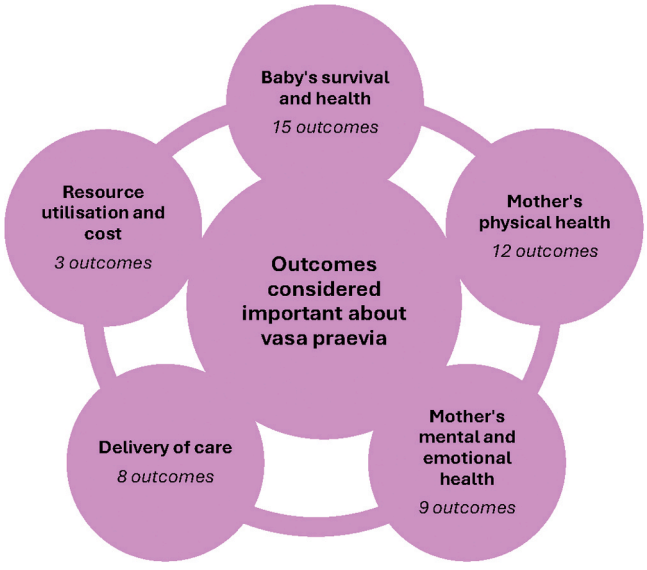

Fig. 1. Overview of thematic analysis describing outcomes and priorities considered important by women and clinicians.

Table 3
Themes, sub-themes, outcomes and experience measures related to pregnancies complicated by vasa praevia.

Theme	Sub-theme	Patient-important outcomes or experience measures
Baby's survival and health	-Survival	Livebirth/stillbirth
		Neonatal death
	-Fetal growth and wellbeing	Damage to/rupture of the fetal vessel
		Fetal growth concerns
	-Adverse events at/following birth	Gestational age at birth
		Wellbeing at birth
		Need for immediate/early neonatal interventions
		Early neonatal complications
		Admission to NICU
		Length of hospital stay
	-Mother and baby bonding	Skin-to-skin contact
	-Infant feeding	Breastfeeding
	-Baby's long-term outcomes	Physical or structural development
		Neurodevelopmental and cognitive
		Requirement for ongoing health care monitoring and testing
Mother's physical health	-Adverse antepartum events	Antepartum haemorrhage
	-Antenatal care	Prophylactic antenatal interventions for preterm birth
		Preterm birth monitoring
		Antenatal hospitalisation
		Duration of antenatal hospitalization
	-Labour and birth	Intrapartum haemorrhage
		Labor analgesia and anaesthesia
		Mode of birth
	-Postnatal issues	Postnatal care
		Length of stay in hospital
		Wound infection
		Breastfeeding complications
Mother's mental and emotional health	-Mental and emotional health concerns	Anxiety
		Depression
		Blame (Self and/or other)
		Emotional invalidation
	-Long-term impact of perinatal mortality and morbidity	Post-traumatic stress disorder
		Relationship breakdown
		Emotional impact on children
		Subsequent pregnancy planning
		Subsequent pregnancy management
Delivery of care		Autonomy in decision making
		Care satisfaction
		Care continuity
		Respect
		Availability and accessibility
		Acceptability of care plan
		Acceptance of change in antenatal diagnosis
		Adherence and compliance to antenatal care
Resource utilisation and cost		Cost to the health care system
		Cost to families
		Loss of family income

labour.

> "She [obstetrician] was just more conservative. She was maybe scared and I understand it. She didn't want to take the risk. She said 34 weeks is the maximum…I didn't want to get the C-section at 33 or 34 weeks. I wanted to keep her in as long as possible, but obviously I was looking also if it's safe for her or not" (W18)

Obstetricians explained the need for caesarean before the woman goes into labour but highlighted the need to balancing the risk of perinatal mortality and morbidity due to ruptured vasa praevia vessel versus iatrogenic preterm birth and its consequences.

> "Some units deliver even at 34 weeks for vasa praevia and again their worry stems from the inability to be absolutely sure that somebody will not go in labour…we use cervical length, fetal fibronectin, so we try to be clever but still you cannot be sure. So we have a core consensus that we deliver somewhere around 36 weeks. (OB2)

Women and midwives highlighted the importance of mother-baby bonding and breastfeeding even after emergency caesarean and/or when the baby was in a NICU, sometimes in a different hospital, within the context of highly medicalised care.

> "Every baby should go skin-to-skin after birth. I think skin-to-skin in an OR setting is much more of a challenge…It requires parent buy in for sure, but nursing buy in, anaesthesia buy in, obstetrician buy in… If we can get a baby that's born early to breastfeed, the long-term outcomes for that baby are known to be better" (M1)

> "As soon as she was born, I had lots of support really, and I actually expressed milk every couple of hours and they kept it in a freezer in the fridge and that's what they fed her with until … she could start to feed herself, and then she had a bit of both, and then moved on to full breastfeeding" (W13)

This theme demonstrates that women's needs and priorities goes beyond baby's survival and highlights the importance of long-term neonatal outcomes.

3.2. Mother's physical health

When the diagnosis of vasa praevia was made antenatally, women reported maternal physical health implications related to limited physical activity, bed rest and sometimes side effects from medications such as antenatal corticosteroids, terbutaline and magnesium sulphate.

> "I was on kind of modified bedrest…at 30 weeks I went inpatient. I was extremely happy with my care…We live out in the country, if anything were to happen it would take me a while to get to the hospital. So that was probably, I felt like that was the safest place." (W7)

The importance of outpatient management for women with antenatal diagnosis of vasa praevia including screening for risk of preterm birth was highlighted by the clinicians. It was noted that pregnancy should not be overmedicalised for women during antenatal hospitalisaton.

> "There is the risk of medicalising them…Unless she is contracting, she's feeling quite well, and she's being hospitalised and separated from her family and restricted in her ability to do things and her ability to move around…Unless there are comorbidities they're feeling well and they're wanting to be mobile and up and doing their normal daily thing." (M1)

> "If the women are admitted to the hospital…they're not sick, they're healthy. I mean the baby is healthy as well until something happens. It can be sometimes difficult in terms of we are medicalising this, and then it might create more anxiety…It's a good point not to overmedicalise it." (OB3)

Maternal physical health was severely affected by emergency caesarean when diagnosis was not made antenatally. Women reported unanticipated postoperative pain, reduced mobility, and prolonged recovery. Despite severe pain, some women reported discharging themselves early from hospital, following an emergency caesarean, to care for their babies who had been transferred to other hospitals, despite knowing that this could delay recovery and increase wound complications.

"I signed myself out, less than 18 hours after he was born…I went there [hospital where baby was transferred to] as a visitor…not a patient…By the end of that day…I was at home in tears wondering why I had made the decision to go to one hospital when I could have gone to the other one and we would have been admitted in the same place." (W10)

Some women felt that they did not receive necessary postnatal care, because the focus of care was on the baby. One woman stated:

"It was a very traumatic time. I was poorly trying to recover from caesarean and trying to recover…The focus was all about her…We had lots of care for [Baby], but didn't really had that ongoing care for me." (W13)

"I feel like everyone just tended to the baby and they're like, 'Oh, you're fine.' So mom was always left in the back corner…I feel like the mom needs to be taken care of just as much as the baby… I fell into a very deep, dark depression." (W5)

Some anaesthetic considerations not directly related to maternal physical health included report of a stillbirth on account of delaying the caesarean due to unavailability of an anaesthetist. Women reported a preference for *'being awake for birth'*, although the importance of appropriate anaesthesia was recognised.

3.3. Mother's mental and emotional health

According to the participants, vasa praevia regardless of timing of diagnosis and overall outcome, had a considerable impact on women' mental health. When vasa praevia was diagnosed antenatally, women described feeling overwhelmed, anxious and scared about outcomes, such as sudden spontaneous rupture of membranes or bleeding, uncertainty about the maternal or fetal source of potential bleeding, not being able to get to the hospital in time for emergency caesarean, and the risks of premature births.

"He [doctor] said 'You are either in bed or on the couch, you can get up to get something to eat or to shower. But the rest of the time, I want you on the couch or in bed…You're on bed rest or if you don't follow this then you can lose this baby." (W 10)

"There wasn't really any acknowledgement or support for the fact that for two weeks I've been locked up in a hospital with people talking scary things at you 'This could happen, you could lose baby'. If someone talks like that for a long time, regardless of the outcome, you need help, don't you…Emotional help and support is important." (W13)

Nevertheless, all women with an antenatal diagnosis (even when it resolved at a later gestation) shared that they felt lucky vasa praevia was detected antenatally. Those who had received an agreed pregnancy and birth plan expressed satisfaction with their care and that they felt reassured and safe during pregnancy. Women acknowledged that anxiety is associated with diagnosis of any condition, but they preferred to know, and that the anxiety from having the diagnosis would probably be lower than that of having an adverse outcome from not knowing.

"A little bit of anxiety and stress is worth it for your baby to not be dead, because I guarantee you're going to have a whole lot more anxiety and maybe a whole lot more stressed out if your baby dies". (W 4)

In instances where the diagnosis was not made antenatally, and neonatal outcomes were poor, women described experiencing psychological trauma not only in the short-term from the emergency caesarean, but also a long-term impact on mental health of themselves and their partners, negatively affecting their relationship, and desire for future pregnancies. Some blamed clinicians and the healthcare system for a lack of antenatal diagnosis and described feeling angry and misled when told that adverse outcomes were *'unavoidable'* due to vasa praevia being an *'undetectable'* condition. They reported diagnoses of depression, anxiety, and/or post-traumatic stress disorder (PTSD). Some recognised that their focus on the baby and being in *'fight-mode'* may had led to a delay in self-care, self-advocacy and a diagnosis of depression, sometimes identifying their need for emotional support after one year. Many women felt that the focus of partners and clinicians on the baby sometimes led to feelings of social isolation.

Despite the low risk of recurrence of vasa praevia, women described anxiety related to traumatic births and adverse outcomes in subsequent pregnancies, leading to browsing online for pregnancy and birth complications, and requesting interventions, including extra tests, scans, hospital admissions, and early caesarean, even in the absence of vasa praevia. For example, one woman requested medically non-indicated preterm births in three subsequent pregnancies after enduring the mortality of her newborn due to vasa previa. Another explained:

"My mental state was shocking [during subsequent pregnancy]. I worried all the way through the pregnancy…went on maternity leave at 25 weeks because I was so nervous and anxious…spent most of my time on Google…and forums researching other things that might go wrong because if you've already had one thing go wrong that you didn't know about, how many other things are out there that could be managed if only you knew about them". (W10)

Clinicians also described the impact of vasa praevia on the mental health of women. They explained that stress and anxiety due to vasa praevia would be largely reduced by antenatal diagnosis, effective disclosure of the diagnosis, emotional support, and individualised and agreed upon care plan. Providing reassurance to women that perinatal outcomes are good with antenatal diagnosis and appropriate care were highlighted to be pivotal.

Some women perceived that clinicians may also experience stress while caring for the pregnant people with vasa praevia. Both clinicians and women discussed that clinician and/or patient anxiety may influence timing of antenatal hospitalisation and timing of birth.

3.4. Delivery of care

Discussions about delivery of care identified several related outcomes. Midwives and obstetricians recognised the importance of taking *'a holistic approach'* in providing care for women with vasa praevia. To improve quality of care, autonomy in decision-making was highly valued and linked to having access to information. Women described the importance of making informed choices and receiving adequate, evidenced based, clear and comprehensive information at the right time, regarding the risks and benefits of different care pathways to facilitate shared decision-making and development of a pregnancy and birth plan.

"It's about choice…the options…you'd rather be anxious and get it checked out…it's not just the baby. It's the placenta…that needs to be looked into more, but I didn't have that chance because I didn't know." (W17)

Obstetricians explained the need for a negotiated individualised pregnancy and birth plan based on women's other risk factors and risk tolerance, where the woman lives, access to emergency resources, family situation, and type of vasa praevia. The value of patient-reported outcomes and experience measures was noted.

"There needs to be obstetric, maternal, neonatal, fetal outcomes… There also needs to be patient-reported outcomes and patient-reported experience measures developed for this particular condition." (OB1)

Some women who did not have an antepartum diagnosis, and consequently experienced vaginal bleeding, felt that clinicians did not take their bleeding seriously to consider a diagnosis of vasa praevia and therefore expedite emergency caesarean, as their suspicion that the

bleeding was from ruptured fetal vessels was low. However, women with an antenatal diagnosis of vasa praevia reported trusting their care providers and being satisfied with their care, especially when there was open and effective communication and an agreed-upon plan for pregnancy and childbirth.

> "I got all the information; it was given to me by a very experienced doctor…We all collectively as a team decided that 37 weeks would be fine if I checked into the antepartum unit and had round the clock care". (W4)

Participants emphasised the importance of continuity of care with the primary care provider while having a team provide specialised care, because it facilitated trusting relationships between women and their care providers, which in turn, led to improved care satisfaction. The importance of continuity of care was also noted.

> "It was very, very important for me that I only see one and only doctor. He knows what's going on, we made a plan and I can trust him." (W18)

> "All my appointments at the doctor's surgery were with the same midwife…I had her in my first pregnancy as well. So it was quite nice cause I knew her…they know when you're nervous and your history and what's going on and things. It makes it nicer. Makes you feel more cared for". (W 12)

> "I think continuity of care, I think continuity of advice. I think it's important". (OB1)

Lack of continuity of care was perceived as a contributing factor for adverse outcomes. For example, one woman felt that her episodes of antenatal bleeding could have triggered the need for detailed ultrasound, if she had seen the same health care provider.

> "I fell through the system…I never saw the same midwife…it was a different midwife who didn't know me, didn't know my baby…I think if you've got one midwife, she gets to know you…you build trust." (W 17)

Availability of and access to high-quality ultrasound, clear evidenced-based information, a skilled and knowledgeable multidisciplinary team, emergency resources, emotional and peer support were considered important by all participants. Although women originally had planned to have a vaginal birth and minimal antenatal interventions, suggested interventions including antenatal hospitalisation and early caesarean were not only accepted by the women, but also made them feel safe. However, a decision to diverge from clinician's advice (non-adherence) was reported by some women, especially regarding timing of these interventions.

> "The standard protocol is to deliver a baby at 34 weeks for vasa previa, which they pushed for and I kind of pushed back." (W4)

The importance of safely delaying antenatal hospitalisation and elective caesarean (if there was no sign of preterm labour) was highlighted repeatedly in the interviews with the women who had antenatal diagnosis as well as the clinicians. For example, one woman changed her hospital and obstetrician because she was told at 19 weeks that she needed to have a caesarean at 34 weeks:

> "She [Obstetrician] said 'we are going to admit you to the hospital at 30 weeks. I am going to take the baby out at 34 weeks and I don't go any longer'. I didn't want to get the C-section at 33 or 34 weeks. I wanted to keep her in as long as possible…She didn't want to take the risk…He [second obstetrician] wasn't that scared…I ended up seeing him." (W18)

Similarly, self-advocacy was reported by another woman who was offered contrasting opinions. Although maternal-fetal medicine specialists had confirmed vasa praevia at 28 and 32 weeks, the woman was told two days before her planned caesarean (at 36^{+5} weeks) by a general obstetrician (based on a repeat ultrasound at 36 weeks), that she no longer had vasa praevia and could have a vaginal birth. She self-advocated and decided to have a specialised ultrasound the following day, which confirmed the presence of vasa praevia, following which she had a caesarean. Vasa praevia was confirmed at birth, which led to a case review and a patient-care conference. In contrast, another woman accepted that her vasa praevia had resolved and gave birth vaginally, after ultrasounds at 28 and 30 weeks confirmed no vasa praevia, with none visible at birth. These experiences highlight the essential need for accurate diagnosis, which may be more challenging in the third trimester.

3.5. Resource utilisation and cost

Impact of vasa praevia on the healthcare system resource use, and cost to families were discussed by clinicians and all women, particularly those who did not have antenatal diagnosis of vasa praevia. Women described financial burden from paying for a funeral, medico-legal investigations, long-term neonatal monitoring, and tests and treatments after experiencing adverse perinatal outcomes. Some reported the financial burden from years of mental health counselling and treatments. The inability to go back to work after the loss of their baby was described by some women, while reporting that their partners focused more on work. Loss of income was also expressed by some women who had prolonged antenatal hospitalisation due to vasa praevia, although some reported being able to work from the hospital. Women who experienced adverse outcomes reported increased health service utilisation and spending more on their subsequent pregnancies, because their anxiety prompted them to choose having several ultrasounds and more interventions during their pregnancy and birth.

> "I still see a psychologist…Two years' worth of financial hardship, emotional hardship, mental hardship…the fact that I haven't been able to conceive over those two years, I've been through three failed rounds of IVF in those two years." (W 9)

Some women and clinicians linked lack of routine vasa praevia screening to the cost to the healthcare system. They perceived that women could have the screening at 18–20 weeks if they had been given the information and choice. For example, one woman explained that:

> "If I had gone to my midwife appointment and they said to me, all right we need to book you in for your 20-week ultrasound. You can have a normal abdominal ultrasound or you can have a transvaginal ultrasound and…explained the things that a transvaginal ultrasound can pick up compared to what a normal abdominal ultrasound can pick up, then the woman has the choice as to which ultrasound she wants to go with… I know that I would've picked the transvaginal ultrasound…I would have paid more." (W9)

> "If people know that this was something that could be investigated as a routine antenatal ultrasound, albeit with somebody with a very specialist skill…they would have wanted to have had that discussion with somebody to make a choice…they would have availed themselves of that opportunity and borne whatever cost was associated with it." (M2)

All participants highlighted the important of a detailed morphology ultrasound checking for the placenta, velamentous cord insertion and/or vasa praevia. Some stated the need for routine vasa praevia screening and perceived that *"because vasa praevia is something that they* [obstetricians] *rarely see, they don't have to worry about it. They downplay it.*" (OB1)

> "The argument against universal screening is, it is not cost effective, there could be false positives…That is not accurate, all the studies that have been done, have shown a very high accuracy". (OB4)

4. Discussion

We identified 47 outcomes and experience measures that were important for women, midwives, and obstetricians. These represented the broad themes of baby's survival and health, mother's physical health, mother's mental and emotional health, delivery of care, and resource utilisation and cost. In addition to identifying outcomes considered important for women to inform the development a core outcome set, our findings emphasise the interplay between clinical and non-clinical outcomes and experience measures as well as their impact on the short- and long-term health of families, which should be considered in clinical practice and future research studies.

Most studies on vasa praevia have focused on reporting perinatal mortality with limited reporting, if at all, on long-term neonatal outcomes following neonatal near-miss events and/or very early iatrogenic caesarean, resource considerations from the women's perspective, or the impact on the mental health of women. Indeed, perinatal mortality and morbidity due to vasa praevia have a devastating impact on families and their care providers [18,19], and was the most important outcome discussed by all participants. However, our study highlights that in addition to baby's survival without short- and long-term morbidity, women valued their own physical, mental, social and financial wellbeing, access to antenatal screening and diagnosis, information on management options and consequences, continuity of care through a primary care provider alongside specialised care, clear and effective communication, emotional support, and the appreciation of individual variations to risk tolerance, values and resource availability.

Core outcome sets – a minimal standardized set of outcomes to be reported in all future studies on a topic – often have a heavy clinical focus, and our study affirmed that women with lived experience of vasa praevia highly value clinical outcomes related to mother and baby. In addition to the outcome domains of mortality and clinical morbidity, participants identified as important, outcomes representing the three other broad outcome domains in healthcare research: functioning/life impact, adverse events, and resource use [16]. This finding is consistent in qualitative studies on pregnant women with other conditions such as obesity [20], venous thromboembolism [21] and heart disease, [22] which highlight how women with lived experience of these conditions value outcomes across all five major outcome domains, in contrast to the published literature that emphasises mostly mortality and physical morbidity. The 47 unique outcomes or experience measures identified in this study were integrated with the outcomes identified from literature review [4] to design the questions for an international Delphi survey to develop vasa praevia core outcome set. The results of the Delphi survey with 204 women, midwives and obstetricians will be published separately.

Participants reported an overwhelming impact of the diagnosis (or lack thereof) on women's mental health. They noted that the focus of the caregivers was often on the binary outcome of the pregnancy – survival vs. mortality of the baby, rather than a holistic view on the overall health and wellness of the family. Many women, particularly those not diagnosed antenatally felt that they were left out of participating in their own pregnancy care – either having information withheld, not having a choice in their care plan or not being listened to. Indeed, participants valued the importance of provision of clear, evidenced-based information, similar to what has been previously reported [19].

Not all statements generated through these interviews reflect 'measurements or observations utilised to evaluate the effectiveness or safety of interventions', which is how 'outcomes' are defined in the context of clinical trials [10]. However, from the women's point of view, outcomes are often difficult to explain, especially when used for preventive interventions such as ultrasound screening or inpatient management, and closely connected with experiences [8], making it important to elicit both. To this end, women expressed that several factors often altered their decisions and care pathways as well as the outcome and overall experience of their pregnancy and birth and are therefore important to consider and capture in studies on vasa praevia. These included autonomy in decision-making, respect, continuity of care, access to high-quality maternity care and emotional support.

Now that the perinatal mortality of vasa praevia has considerably decreased through antenatal diagnosis [1,23–25], the emphasis should also be on provision of woman-centred care and improving maternal emotional health, respect, shared decision-making and satisfaction with care, in addition to physical health [6,26]. Compassionate and high-quality, evidence-based care provided by specialised, multi-disciplinary teams may improve outcomes deemed important for women, alongside clinical outcomes [27].

5. Strengths and limitations

Designed and conducted by a multidisciplinary team of clinicians, researchers, and consumer representatives from two countries, with expertise in midwifery, obstetrics, maternal-fetal medicine, core outcome set development, qualitative research, and clinical/lived experience of vasa previa, the study provides a holistic, woman-centred view to guide clinical management of and future research studies on vasa praevia. To enhance the study rigour, careful consideration was given to ensuring a diverse representation of women in terms of geography, experiences, and clinical outcomes.

Although transferrable to clinical practice and research settings, as with all qualitative research studies, our findings cannot be generalised to all settings and is limited by selection- and reporting bias. Women were from high-income, English-speaking countries, whose views may not reflect those from low-income countries and/or other linguistic and cultural backgrounds. Given more than two-third of the women were highly educated, findings may represent views of women who are active in advocacy for themselves and other women, and not capture the views and experience of women with low health literacy. Nevertheless, as studied on vasa praevia continue to be published, especially those comparing alternate management approaches [24], this paper would enable researchers to study and report on outcomes important for women in addition to the clinical outcomes that have been traditionally reported.

6. Conclusion

This study identified outcomes and experience measures that were considered important for women and clinicians including baby's survival and health, mother's physical and mental health, and delivery of care. In addition to informing the development of a core outcome set, the findings may serve the important purpose of peer-education, creating awareness on what women want and value from interventions regarding vasa praevia. Including the outcomes considered important for women in future clinical research study designs will inform future care and research to be driven by outcomes that are important to the women we care for.

Author contributions

NJ, ND, JK and RDb conceived and designed the study. NJ collected data and drafted the manuscript. NJ and RDa analysed the data. All authors contributed to the analysis and interpretation of the data, critically revised the manuscript, and approved the final version of the manuscript.

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Ethical statement

The study protocol was approved by the Mount Sinai Hospital Research Ethics Board, Canada (18–0173-E), and the University of Technology Sydney Ethics Committee, (ETH19-3718).

Declaration of Competing Interest

Dr Natasha Donnolley is the Director and Vice President of International Vasa Previa Foundation. Other authors report no conflict of interest.

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