Journal of Community Genetics (2025) 16:167–181
https://doi.org/10.1007/s12687-025-00768-5

RESEARCH

Perceptions of patients and stakeholders on a prenatal sickle cell disease screening and its results among tribal populations of Gujarat: a participatory mixed-method research

Apurvakumar Pandya[1] · Kapil Dave[2] · Shrey Desai[2] · Gayatri Desai[2]

Received: 25 June 2024 / Accepted: 9 January 2025 / Published online: 21 January 2025

Abstract
Prenatal screening (PNS) for Sickle Cell Disease (SCD) offers a potential avenue for informed reproductive choices and the sickle elimination initiative of the Government of India. The objective of the study was to explore perceptions and ethical dilemmas surrounding prenatal screening for sickle cell disease and subsequent termination of pregnancy among pregnant women and key stakeholders from the tribal region of Gujarat. The study employed sequential mixed-methods research, embedding a participatory research approach. Pregnant women and key stakeholders from Jhaghadia, Bharuch District of Gujarat were interviewed. The qualitative data was analyzed using a thematic analytic framework, and the quantitative data was presented with descriptive statistics. Findings revealed key themes such as knowledge about SCD and its management, knowledge about PNS, barriers to PNS, acceptance of PNS and ethical dilemmas related to the termination of pregnancy (TOP). The results show a mixed preference for prenatal screening from both qualitative and quantitative data, primarily due to the complex decision-making process. Couples with SCD reported a high preference for PNS and TOP if the foetus had sickle cell disease whereas couples living with SCD trait were assigned lowest priority and preference to under-go PNS and TOP. Decisions were influenced by limited knowledge, lack of lived experience of SCD, family pressure, cultural and religious beliefs, and social stigma. A comprehensive behaviour change communication strategies that empower couples and key stakeholders through culturally sensitive SCD & PNS literacy, and genetic counselling is essential to promote informed decision-making within the cultural context of tribal communities.

Keywords Sickle cell disease · Pre-natal screening · Termination of pregnancy · Genetic counselling · Sickle cell elimination mission

Introduction

Sickle cell disease (SCD) is a monogenic condition where severely malformed red blood cells are formed (Platt, Dornelas, & Shackleton 2017). SCD is caused by a point mutation in the gene coding for β-globin (Hardouin et al. 2023). Sickle cell trait is a genetic condition that arises when an individual inherits one gene for normal haemoglobin (A) and one gene for sickle haemoglobin (S), resulting in the genotype (AS). In contrast, sickle cell disease occurs when a person inherits two abnormal sickle genes (SS) (Ashorobi et al. 2023).

People with SCD can suffer from anaemia, painful episodes, susceptibility to infection, stroke, and chronic organ damage particularly, kidneys, lungs, heart, and brain are more susceptible (National Heart, Lung, and Blood Institute, n.d.). There is currently no cure available for SCD, but the condition can be managed using a variety of therapies (Sickle Cell Foundation 2023). Therapies can extend life expectancy to about 45 years (Platt, Dornelas, & Shackleton 2017). SCD is a significant public health concern in tribal communities across the globe, especially tribal regions in India. The sickle gene is widespread among many tribal population groups in Gujarat, a western state of India (Colah et al. 2015; Kishore, Gupta, & Gupta 2023). Saxena and his team in 2017 estimated to have at least 9,00,000 sickle cell trait and 70,000 SCD patients (Saxena et al. 2017).

✉ Apurvakumar Pandya
apandya@iiphg.org

[1] Indian Institute of Public Health, Gandhinagar, Gujarat, India

[2] SEWA Rural, Jhagadia, Gujarat, India

Based on a modelling exercise conducted by Frédéric Piel and colleagues one decade ago, it has been estimated that, between 2010 and 2050, about 14.2 million babies will be born with sickle cell anaemia (Piel, et al 2013). The prevalence of the sickle cell gene is as high as 5%–34% in various scheduled tribes (STs), who are socioeconomically disadvantaged (Hockham, et al 2018; Patel, et al 2013; Saxena et al. 2017). Numerous community-based studies have documented a high prevalence of sickle haemoglobin (HbS) ranging from 2.3% to 31% (Chavda, Goswami, & Goswami 2015; Serjeant et al. 2016; Shiladaria, Patel, & Oza 2013).

Patients with SCD experience considerable morbidity from both acute and chronic complications that lead to end-organ damage and reduced life expectancy (Brousse & Rees 2021). In addition, disease-related complications starting in infancy significantly impair physical, mental, and psychosocial aspects of health-related quality of life (Panepinto 2012). Common SCD-related complications among paediatric patients include acute pain, severe anaemia, stroke, splenic sequestration, acute chest syndrome, and meningitis (Beck et al. 2022). As per an Indian Council of Medical Research survey, about 20% of children with sickle cell disease die by the age of two, and 30% of children with SCD in the tribal community die before they reach adulthood (Tewari, & Rees 2013). SEWA Rural organization, which implements comprehensive care for SCA, documented a high prevalence of pain crises, hospitalizations, and blood transfusions among the patient population (Desai et al. 2016).

Prenatal screening (PNS) for sickle cell disease

Prenatal screening is a routine aspect of antenatal care and is offered based on informed reproductive choices and potentially mitigates the future burden of the disease (Patel, et al 2013). Early detection allows for informed family planning (Desai et al 2016; Linton, et al 2023; Patel, et al 2013). Figure 1 explains the flowchart of the prenatal SCD screening process.

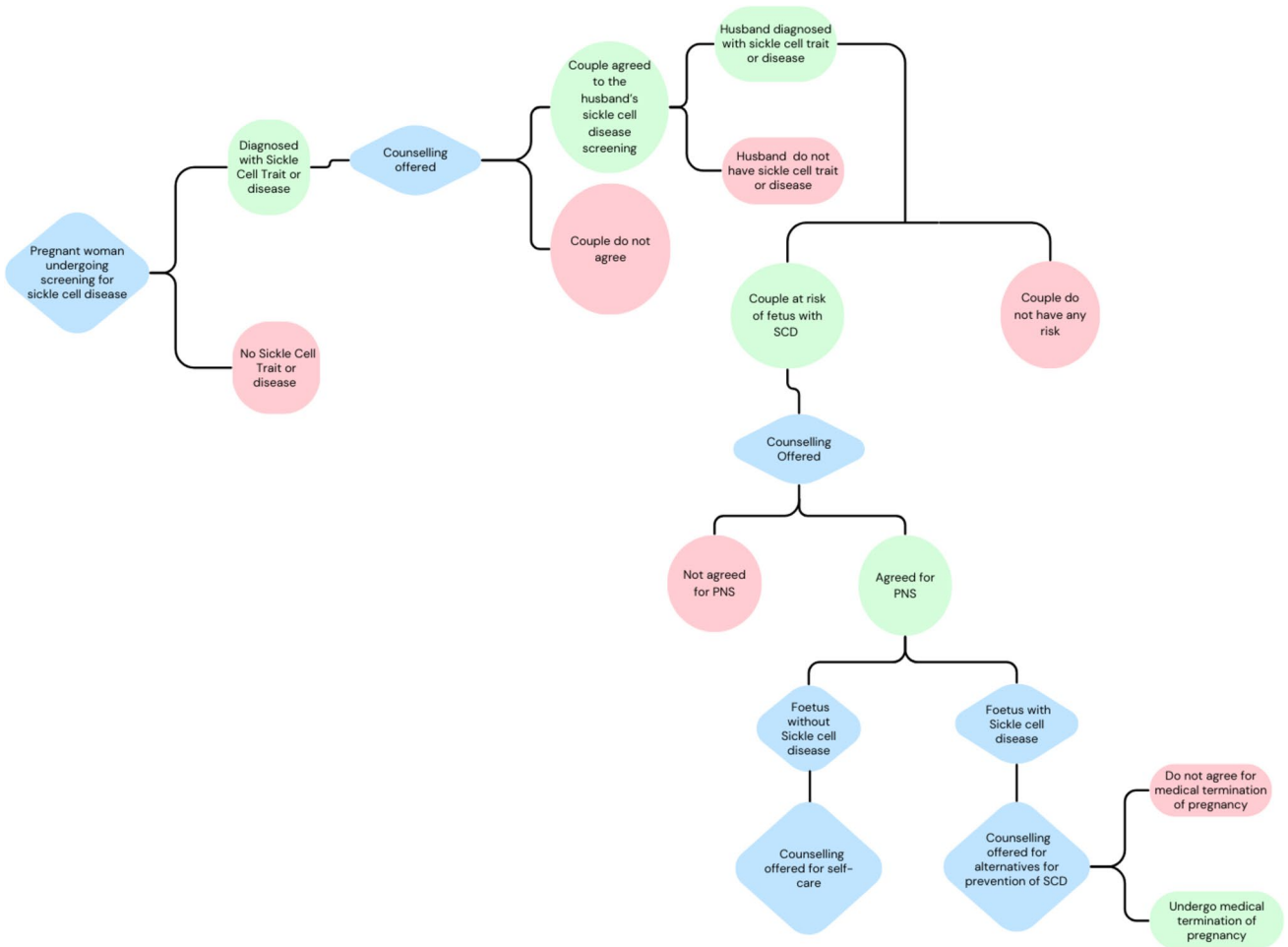


Fig. 1 Flow diagram of the PNS process

[The flow diagram explains the prenatal screening process. Pregnant women testing for sickle cell disease. If she is found with a trait or disease, her husband is motivated for sickle cell testing. If a couple is found with either sickle cell trait or disease, they are counselled for pre-natal screening of the foetus. If a foetus is found with sickle cell disease, couples are counselled to make informed decisions about pregnancy. If couples agree, the foetus is terminated. If couples do not agree, antenatal and post-natal services are offered continued being offered.]

The most common method of PNS involves first identifying a couple who is at risk of having a fetus with sickle cell disease, counselling at-risk couple to undergo PNS, followed by taking a sample of amniotic fluid and genetic testing of the fluid to determine whether the fetus has SCD or not. In case the fetus has SCD, the couple is counselled so that they can make an informed decision about termination or continuation of pregnancy. However, navigating this pathway presents a complex terrain with barriers, ethical dilemmas, and diverse community factors. It is important to understand patients' and stakeholders' perspectives. The Government of India has initiated a campaign to eliminate sickle cell disease by 2047 (National Sickle Cell Elimination Mission 2023). To achieve this goal, prenatal screening, testing, and counselling services play a crucial role as there is no way to know how severe would be the disease in a given fetus after birth (Sickle Cell Anemia Control Program 2013).

The present study explored patients' and key stakeholders' perspectives and navigated perceived ethical issues on PNS for Sickle Cell Disease. Specific Objectives were exploring perceptions and perspectives of patients, couples, and stakeholders on pre-natal screening of SCD and termination of pregnancy as a preventive strategy; acceptability of pre-natal screening among eligible couples and stakeholders for pre-natal screening of SCD and termination of pregnancy as a preventive strategy, and explore ethical dilemmas in taking decisions about pre-natal screening and termination of pregnancy. The findings of the study may inform the Sickle cell elimination mission of the Government of India.

Methodology

Study area

The study was conducted in the Jhagadia block of Bharuch District, Gujarat, India, located approximately 20 kms from Bharuch City and 225 kms from Gandhinagar, the state capital (Census of India 2011). The district has a population of 1,551,019, with 31.5% being tribal (Census of India 2011). The selected block, Jhagadia, has a significantly higher tribal population, accounting for approximately 69% (Census of India 2011).

Study setting

The SEWA Rural, a voluntary organization, implements a comprehensive care model for SCD at its 250-bed charitable hospital in Jhaghadia block, Bharuch in Gujarat since 2014. This comprehensive care model for SCD includes screening, standardized treatment as per inpatient and outpatient protocols, and health education of the community. The organization started providing PNS service in 2022.

Research design

This study employed a participatory sequential mixed-methods design (qual-quant). Participatory research involves collaboration between researchers and participants to understand a specific issue and work towards positive change. In this context, participatory action research principles were utilized, emphasizing active participation and action by community members directly affected by the issue (Baum et al. 2006; Vaughn, & Jacquez 2020).

In order to ensure community ownership and relevance, the first author facilitated a participatory workshop with representatives of the research participants. This collaborative process involved participants in the development of the research design and strategies, fostering a deeper understanding of their perspectives and concerns. These members were part of the pilot exercise as well, providing valuable feedback on the research instruments and data collection procedures before their application to the main study sample.

Research positionality

This study prioritizes health equity by focusing on tribal communities in Western Gujarat, India, historically marginalized groups facing significant health disparities. The first author, a native of Valsad district, a tribal region in Western Gujarat, completed his secondary education there. While his higher education took place in Vadodara, a city in Central Gujarat, he maintains a deep understanding of the tribal communities of Bharuch district, also located in Western Gujarat. His research training emphasizes qualitative methodologies, particularly grounded theory and participatory research. He has extensive experience working with diverse marginalized populations, including women, LGBTQ + individuals, and children in vulnerable situations. Furthermore, his doctoral research, grounded in Charmaz's Constructivist Grounded Theory (Charmaz 2006), underscores an interpretivist paradigm that recognizes how socio-cultural and educational experiences shape individual realities. This perspective aligns with the understanding that systemic inequalities, driven by factors such as power dynamics and social structures (Bourdieu 1986), deeply influence health outcomes. These inequalities are often reflected in research

and programme design, where beneficiaries are often treated as passive recipients of externally imposed solutions, hindering health promotion (Wallerstein 1979).

Present study challenges inherent inequalities by embracing participatory research methodologies that prioritize the perspectives of marginalized communities as crucial knowledge sources (Creswell 2013). This approach offers a more equitable and empowering framework, allowing communities to actively shape their own health and well-being. This study was designed with the fundamental principle that participants are not only subjects but also equal partners and experts in their own lived experiences. The first author led the study's design and execution, with four data collectors from the academic institution who were trained in public health and research. These data collectors received specific training in participatory research and qualitative interviewing by the first author. The co-authors, who are part of SEWA Rural, maintained a supportive role throughout the study, refraining from any interference in data collection, analysis, or interpretation. The co-authors also critically reviewed the manuscript and provided valuable input to enhance the presentation of the research findings.

Sample

Description of sample for both qualitative and quantitative study are described below.

Sample for qualitative data: About 59 in-depth interviews were conducted. The study had diverse range of participants, ensured a comprehensive understanding of the SCD situation within the community. Direct beneficiaries (38) included individuals undergoing Sickle Cell Disease (SCD) screening, individuals who diagnosed positive for SCD, those who underwent prenatal screening, those who terminated their fetus due to SCD diagnosis, individuals with Sickle Cell Trait, and two participants had a child with SCD. They provided valuable insights into the lived experiences of individuals and families affected by SCD. Another group, community stakeholders (11), included family members, community leaders, teachers, and elected village heads – Sarpanch who offered crucial perspectives on community perceptions, beliefs, and social norms related to SCD. Service providers (10), such as Auxiliary Nurse Midwives, Accredited Social Health Activists, prenatal screening service providers, genetic counsellor, gynaecologist, members of the Sickle Cell project team, and Hematologist provided valuable insights into the challenges and successes of current SCD prevention and management programmes within the community. This multi-faceted approach to participant recruitment allowed for a nuanced understanding of the SCD situation, encompassing the perspectives of all key stakeholders. Details of the same are provided in Table 1.

Sample for quantitative data: Programme data of 968 women with sickle cell gene attending SEWA Rural OPD during 2022 and 2023 were considered for the study. Secondary data of all beneficiaries who visited pre-natal screening services was gathered from the project records.

Sampling technique

For qualitative data, theoretical sampling was employed. The sample was diversified based on Sickle Cell Disease status: couples for prospective screening, couples having sickle cell disease, women who underwent prenatal screening, women who underwent an abortion, women who did not undergo termination of pregnancy, geographic locations within Jhagadia block, participants from different income strata, etc.; and different stakeholders from family, community and service providers. Data collection was continued until data was saturated. For quantitative data, all pregnant women with sickle cell gene attending the Gynec OPD clinic at SEWA Rural were considered for the study.

Research tools

In-depth interviews were conducted with 59 participants using a semi-structured interview guide. The guide, developed in English based on relevant literature and research questions, was translated into Gujarati and back-translated to ensure accuracy. Pilot interviews with five participants (three beneficiaries – one with SCD, one with SCD trait, and

Table 1 Type of participants of the study and its characteristics

Type of participants and total number	Description
Direct beneficiaries (38)	Participants about to undergo SCD screening (9); participants who screened SCD positive (4); participants who underwent pre-natal screening (4); participants terminated fetus due to diagnosis of SCD (3); couple having SCD positive child (2); and participants with Sickle Cell trait (16)
Community stakeholders (11)	Family members (6), community leaders (2), Teacher (1), Sarpanch /village head (2)
Service providers (10)	Auxiliary Nurse Midwifery (ANM) (1), Accredited Social Health Activist (ASHA) (3), PNS service provider (2), genetic counsellor (1), gynaecologist (1), Sickle Cell project team (1), and an external subject matter expert (1)

one with SCD-affected child; and two project team members) were conducted to refine the guide. The final guide explored participants' knowledge and perceptions of SCD, experiences with prenatal screening, decision-making processes, and ethical considerations.

Data collection procedure

The first author, a trained qualitative researcher, conducted rigorous training for four research team members on participatory research approaches and in-depth interview techniques. To familiarize the research team with the local context and refine data collection strategies, the team carried out pilot study at the SEWA Rural office.

Interviews were conducted in Gujarati by the first author and four trained team members, with support from the SEWA Rural team. Interviews were primarily conducted at the OPD managed by SEWA Rural, with patients and stakeholders from the village. Three stakeholders, who were medical professionals and unable to provide time for in-person interviews. To ensure autonomy of responses, interviews with married couples were conducted with each spouse individually.

While in-person interviews were the primary method of data collection (n = 56), three participants were interviewed telephonically. This decision was made based on logistical and practical considerations. Telephonic interviews can be advantageous in reaching participants who may be geographically distant, have limited mobility, or face scheduling challenges (Bernard 2002). In this study, telephonic interviews were conducted to ensure the inclusion of key stakeholders who were located in remote areas and whose participation would have been significantly impacted by the logistical constraints of in-person interviews.

The quantitative data encompassed information on all beneficiaries who visited their pre-natal screening services, counselled for pre-natal screening services, participants who availed prenatal services and those who underwent termination of pregnancy.

Data analysis

Thematic analysis was used to analyze qualitative data, identifying recurrent themes and patterns within the interviews. The analysis focused on understanding the cultural, social, and psychological factors shaping participants' perspectives. Descriptive statistics were utilized for quantitative data analysis.

All interviews were audio-recorded, transcribed verbatim, and subsequently translated into English by bilingual researchers fluent in the local language. The thematic analysis process followed a six-phase approach as outlined by Braun and Clarke (2006), encompassing familiarization with the data, generating initial codes, searching for themes, reviewing and refining themes, defining and naming themes, and finally, producing the report. This involved immersing oneself in the data through repeated readings of transcripts, field notes, and other relevant documents, systematically identifying and labeling key concepts, grouping codes into potential themes, and refining these themes through an iterative process. Themes were then clearly defined and given concise and meaningful names. Throughout the analysis, researchers maintained a reflexive approach, acknowledging their own biases and perspectives to ensure that interpretations were grounded in the data.

Ethical considerations

This study received ethical approval from the SEWA Rural Institutional Ethics Committee on August 9, 2023. Prior to participation, all participants were informed about the study's objectives and procedures. They were also explicitly informed that their participation was entirely voluntary and that they could withdraw from the study at any time. Written informed consent was obtained from all participants.

Results

Socio-demographic profile of study participants

A total of 59 participants, including 38 direct beneficiaries, 11 community stakeholders, and 10 healthcare service providers, were interviewed for the study. Of the total direct beneficiaries (38) interviewed, 13 had Sickle Cell Disease, 16 had Sickle Cell Trait, and 9 had neither condition. About 17 participants were belonged to low socio-economic strata, and 13 did not receive formal education. The gender distribution showed 29 women and 9 men among direct beneficiaries. In the case of community stakeholders (11), seven were men, six were from low socio-economic strata, and three received no formal education. Table 2 presents the socio-demographic characteristics of study participants.

As presented in the Table 2, healthcare service providers were categorized by gender and occupation. Among the providers, 60% (6 out of 10) are women, while 40% (4 out of 10) are men. Occupationally, the group includes one ANM (10%), two ASHAs (20%), three Gynecologists (30%), two members of the Sickle Cell Project team (20%) and one each respectively, Hematologist (10%) and Counsellor (10%).

Uptake of pre-natal Screening, testing, and termination of pregnancies at SEWA rural hospital

Figures 2 and 3 present the uptake of pre-natal services at SEWA Rural Hospital. A total of 968 pregnant women with sickle cell disease or trait visited the service and

Table 2 Socio-demographic characteristics of study participants (N = 59)

Characteristics	*f*	%
Direct Beneficiaries (n = 38)		
Gender		
Women	29	76
Men	9	24
Socio-economic Status		
Low Socio-Economic Status	17	45
Moderate Socio-Economic Status	18	47
High Socio-Economic Status	03	08
Education		
Not received formal education	13	34
Completed five years of Schooling	08	21
Completed ten years of Schooling	07	18
Completed 12 years of schooling	06	16
Completed Graduation	04	11
Sickle Cell Disease Status		
Sickle Cell Disease	13	34
Sickle Cell Trait	16	42
None	09	24
Community stakeholders (n = 11)		
Gender		
Women	04	36
Men	07	64
Socio-economic Status		
Low Socio-Economic Status	06	55
Moderate Socio-Economic Status	04	36
High Socio-Economic Status	01	09
Education		
Not received formal education	03	27
Completed five years of Schooling	02	18
Completed ten years of Schooling	01	09
Completed 12 years of schooling	04	36
Completed Graduation	01	09
Type of stakeholder		
Spouse	03	27
Mother-in-law	02	18
Father-in-law	02	18
School teacher	02	18
Sarpanch/community leader	02	18
Healthcare Service providers (n = 10)		
Gender		
Women	06	60
Men	04	40
Occupation		
ANM	01	10
ASHA	02	20
Gynecologist	03	30
Haematologist	01	10
Counsellor	01	10
Sickle Cell Project team	02	20

nearly 24% (236/968) husbands were counselled for sickle cell screening. Of the total husbands counselled, 94% (221/236) were agreed for pre-natal screening. About 24% (54/221) of them were detected with sickle cell disease. It means 54 pregnant women and their husbands both were carrying sickle cell genes and were eligible for prenatal testing.

These eligible couples (54) who were detected with sickle cell disease were further counselled for amniocentesis of pregnant women and 43% (23/54) of them underwent an amniocentesis test. Among eligible couples, four couples had at least one member detected with sickle cell disease; and about 50 couples where both of them had sickle cell traits. Of those who underwent amniocentesis test (54), 20 had sickle cell trait and 9 had sickle cell disease.

Figure 3 provides details of patients counselled for amniocentesis test and its consequences. Of 54 couples who underwent PNS, 39% (9/23) couples had a foetus with Sickle cell disease. Of the 9 couples, two couples had at least one member suffering from sickle cell disease and seven had only sickle cell trait. Of them, 66% (6/9) couples agreed and terminated their pregnancies.

Reasons reported by patients for not terminating pregnancy were: 1). None of the children are alive at present; 2) previous experience of miscarriages; 3) stillbirth, and 4) husband/mother-in-law denied permission for termination of pregnancy. Reasons attributed by patients for terminating pregnancy were: 1) concerns about the potential of having future children also affected by SCD; 2) fear of painful crisis episodes in child, 3) challenges faced by other families raising child with sickle cell disease and 4) first pregnancy so that they can plan other in future. They mainly emphasized the suffering of the unborn child and their willingness to prevent sickle cell disease in society. The following excerpts aptly highlight these sentiments.

> "I would not want to abort, but what if the child suffers?" - A Pregnant woman.
> "Our religion forbids abortion, but I worry about the child's health." - A Husband without SCD.

This underscores the intricate interplay of cultural, religious, and practical considerations in shaping perceptions of SCD, pre-natal screening, and abortion. Verbatim accounts illustrated how individuals sought a delicate balance between honouring cultural and religious values and making practical decisions for the well-being of their families:

> "Our culture and faith are a big part of who we are, but we also need to think practically. How can we put our children in danger of suffering?" – A man with SCD

Both of the participants had formal education and were of moderate socio-economic status. Such examples were limited; the majority of participants did not accept medical

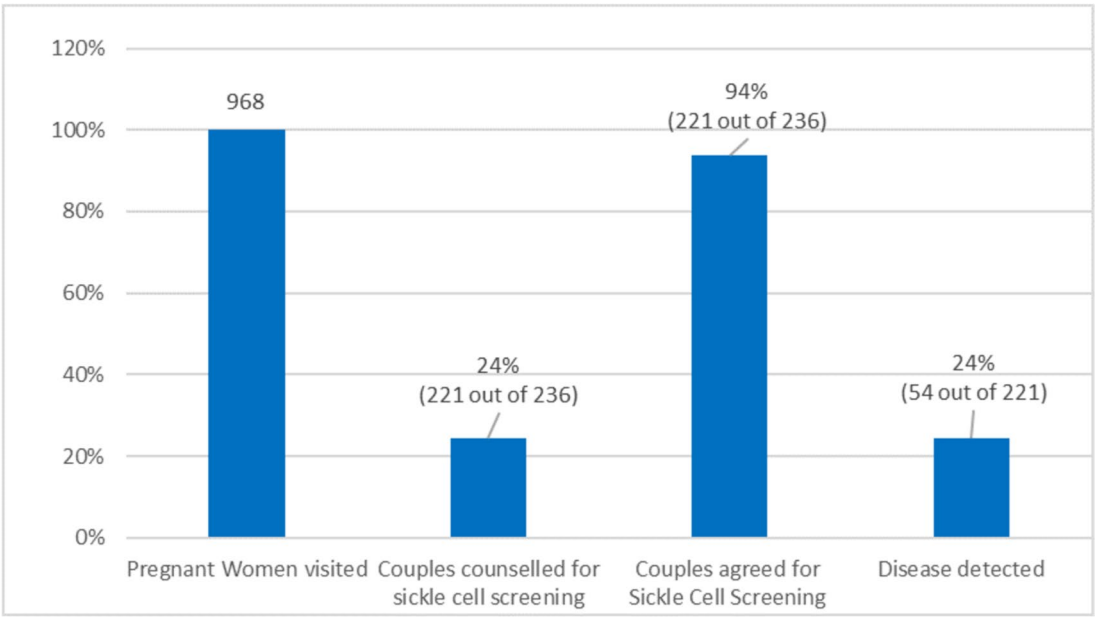
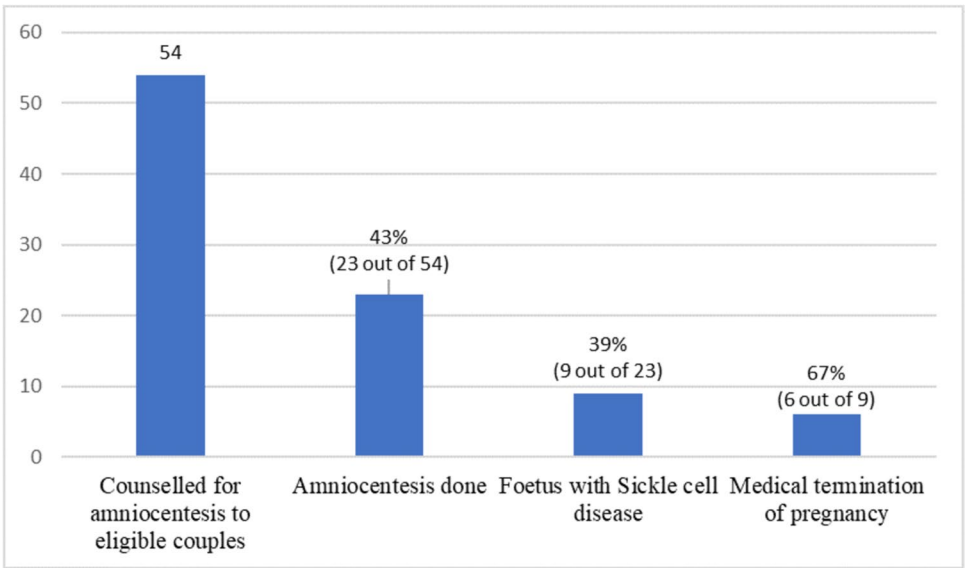


Fig. 2 Uptake of prenatal screening, and testing

Fig. 3 Patients counselled for amniocentesis test and its consequences



termination of pregnancy as an appropriate option for community members.

Perception on pre-natal screening of SCD and termination of pregnancy as a preventive strategy

The study identified critical emergent themes from the data: knowledge about SCD and management, knowledge about pre-natal screening and barriers to pre-natal screening, and medical termination of pregnancy in the case where a fetus is detected with sickle cell disease. The study also explored ethical dilemmas related to pre-natal screening and termination of pregnancy.

1.1. Knowledge about Sickle Cell Disease and Management

All participants had heard the term "sickle cell disease." However, the depth of understanding varied significantly. Participants with SCD (13/38), family members (8/10), and community leaders (2/10) demonstrated a comprehensive understanding of SCD, including its genetic nature, potential health complications, and management strategies.

Participants with the sickle cell trait (16/38) and some community leaders (2/10) exhibited limited awareness. They often lacked a basic understanding of the disease's cause (genetics) and struggled to articulate its specifics. While some (6/16) recognized symptoms, they lacked knowledge about management, including preventive measures and appropriate medical interventions. All participants with sickle cell trait did not know how to manage SCD, such as how to treat complications and care (diet, preventive measures) to be taken by the patient to reduce the severity and when to consult a doctor. Most patients preferred medication during the crisis stage. The following verbatim of participants echo their knowledge about SCD and its management.

> "I have known about my condition [SCD] when I was in school. I have a girl child with SCD, and I know how important it is to take care of a person with SCD. It is important to take regular medication, check-up and take proper care" – A woman with Sickle Cell Disease and a child with SCD.
> "I have heard about it but do not know more details about it. I was diagnosed in my school and taking treatment" – A woman with Sickle Cell Trait.

While exploring further, we noticed varying sources of information about sickle cell disease. Information about SCD came from various sources, including family, friends, and healthcare providers. However, the accuracy of information obtained outside of healthcare settings was often questionable, leading to inconsistencies and confusion. Verbatim excerpts from one of the participants illustrate the scenario aptly.

> "I know about sickle cell [SCD] from my friend… She said…no major symptoms, and medicines are not required... I was getting sick often but never went to the doctor. It was during my pregnancy I was diagnosed with SCD." – A woman with SCD.

Gender disparity in knowledge about sickle cell disease surfaced. Women with lower socio-economic backgrounds and limited formal education (10/29) often had less access to accurate information compared to men, relying primarily on their partners' or family's understanding. Conversely, women with higher socio-economic backgrounds and formal education (19/29) generally possessed more accurate knowledge about SCD and prenatal screening. Participants' verbatim indicated gaps in understanding the implications and significance of SCD, emphasizing the need to improve health literacy about SCD through behaviour change education initiatives.

> "SCD is a blood disease in which red blood cells contort into a sickle shape and block the blood flow, causing pain. Upon diagnosis, treatment can be beneficial… sometimes, blood change [blood transfusion] is required. This can be done at *Mota Davakhane* (Tertiary Hospital) …" – A woman with formal education and upper socio-economic status.
> "I only know it is a blood disease that makes children weak and often sick. My distant relative has it." – A woman had no formal education.

One participant (mother-in-law—community stakeholder) said *"Sickle cell [SCD] is a curse, passed down from ancestors."*

2.2. Knowledge of pre-natal Screening

Most participants (direct beneficiaries- 33/38; community stakeholder – 8/10) were unaware of pre-natal screening, except five patients who recently underwent pre-natal screening at SEWA Rural Hospital. Two community stakeholders –a Sarpanch whose wife had sickle cell disease and the mother-in-law of a person with SCD – were aware of prenatal screening.

Most participants – direct beneficiaries and community stakeholders (41/48) heard the term "pre-natal screening" for the first time. The lack of clarity and accessibility of prenatal screening is reflected in the following excerpts of participants.

> "I heard about the test from the hospital, but where and how much? Will it harm the baby?" – A pregnant woman
> "It is not offered at PHC. Need to go to Jhagadia, which requires travel, and the private hospitals in Bharuch are too expensive for people to afford it." – A Sarpanch.

Participants having SCD favoured pre-natal screening services; however, family members and other stakeholders had limited information about pre-natal screening. When asked, participants (especially those with trait sickle cell disease, family members of trait sickle cell disease, sarpanch) reluctantly agreed favourably to pre-natal screening; however, after explaining the details of the pre-natal screening procedure and its purpose, the majority of them understood the importance of pre-natal screening and expressed willingness to uptake screening facilities if services available.

> "Prenatal screening can help determine whether a child will have sickle cell disease. We do not mind what the doctor says as long as services are available." – A father-in-law

3.3. Barriers to uptake of prenatal screening and termination of pregnancy

The study explored several barriers hindering the uptake of prenatal screening and termination of pregnancy in cases where the fetus tested positive for sickle cell disease. Apart from a lack of lived experience with SCD and limited knowledge about the screening services, barriers included familial pressure, cultural beliefs, religious beliefs, stigma, and emotional reactions. All these contributed to the perception that such screenings were unnecessary.

Familial pressure: Husbands and in-laws, particularly mothers-in-law, often held strong opinions against termination of pregnancy, creating conflict and emotional stress. Pressure to bear children, regardless of the risk of passing on sickle cell, conflicted with concerns about the child's future well-being. Father-in-law said, *"God has chosen this path; we must accept it."* A pregnant woman reported her opinion as follows:

> "I am fine with abortion, but my husband and mother-in-law are not in favour. We had two daughters, and the family looks forward to a boy child. Hence, I need to keep this child." – A pregnant woman.

Traditionally, women had little say in their reproductive choices. Decisions were often made by mother-in-law or husbands. One woman said, "We [married women] just have to listen [to mother-in-law and husband]." However, there were reflections of empowered voices. Some tribal women navigate these challenges with a sense of agency. For example, some women advocated for themselves and their families, seeking healthcare professionals' guidance on reproductive health options and strongly favour termination of pregnancy.

Cultural beliefs: A pervasive theme that emerged was the profound impact of cultural beliefs and traditions on decision-making regarding pre-natal screening and abortion. Tribal communities often hold deep-rooted beliefs about the sanctity of life and natural processes. The notion that interfering with the course of nature is taboo influences individuals to resist pre-natal screening and termination of pregnancy. Termination of pregnancy was not considered culturally appropriate. The most common belief reported was *"Abortion is a sin."* Participants stated,

> "…it [womb with sickle cell disease] is God's decision. Abortion is a sin. Whatever is given by the Almighty, we must accept it...must not disobey God's gift." – A community member.
>
> "If people find out, they will point fingers and talk behind our backs. It is not just a medical decision but a social one." – A husband with SCD.

Religious beliefs: Religious beliefs profoundly influenced the perceptions of individuals with SCD within tribal populations. Verbatim accounts underscored the intertwining of faith and the experience of living with SCD:

> "I believe this is a test from God. He knows I can handle it, and I find strength in my faith." – A husband with SCD.

This sub-theme revealed a perspective that framed SCD not solely as a medical condition but as part of a divine plan. The verbatim responses highlighted how religious beliefs provided individuals with a sense of purpose, resilience, and coping mechanisms in the face of the challenges posed by SCD. Conversely, some participants expressed a struggle with religious interpretations that attributed SCD to divine punishment or a consequence of past actions:

> "People say it is because of sins in past lives. It is hard not to feel guilty and question why God chose this for me." – A husband with SCD

This dual nature of religious beliefs, serving as both a source of strength and a potential source of guilt, adds complexity to the emotional and psychological impact of SCD within tribal communities.

Stigma: Openly discussing sickle cell was uncommon, leading to isolation and lack of understanding within communities. Prevalent stigma regarding the termination of pregnancy was also expressed by study participants.

> "Abortion is not accepted in society; hence, family members do not allow it. Abortion is considered disobedience of the Nature." – A woman with sickle cell trait.
>
> "Our community frowns upon abortion. It brings shame, and people will talk." – A husband.

One participant – a healthcare professional – said, *"… It is [termination of pregnancy] not just a medical decision; it is a social one."* Moreover, attributing the disease to supernatural causes or past karmic deeds perpetuated stigma and limited access to medical care.

Emotional reactions: Participants faced emotional reactions. Women participants grappled with guilt, grief, and the potential social implications of aborting a desired pregnancy. Emotional burdens and societal expectations led to a complex decision-making landscape.

> "The emotional burden is heavy. How do you decide to end a life, even if it is to prevent suffering?" – A woman with SCD

Acceptance of PNS and TOP

Findings revealed high acceptance but mixed preference for prenatal screening, primarily due to complex decision-making processes. Persons with SCD have acceptance and high preference for pre-natal screening; however, persons living with SCD trait, their family members, and community

members had assigned the lowest priority and preference to under-go pre-natal screening and termination of pregnancy if required. Most participants did not favour the decision regarding the termination of pregnancy. It is clear from the data that limited awareness and knowledge, barriers to service uptake, and societal and ethical concerns influenced the decision-making process.

Indeed, faith, morality, and practical considerations shape individuals' perspectives and influence their decisions regarding pre-natal screening and termination of pregnancy. For example, one spouse of a pregnant woman who underwent pre-natal screening and abortion said, *"Our faith says every life is a gift, but does that mean we let our child suffer? I cannot see my wife and child in pain."*

Ethical dilemma in decision-making by beneficiaries and service providers

In situations where the severity of SCD varies greatly or where access to optimal healthcare is limited, navigating the ethical considerations becomes even more complex. Results revealed beneficiaries' as well as provider's ethical dilemmas.

Beneficiaries' perspectives

Participants grappled with the ethical implications of interfering with the natural course of life and the potential guilt associated with decisions. Participants expressed concerns about interfering with natural processes and the moral implications of making decisions about the unborn. A father-in-law said, "Our elders say we should not try to play with God. What if it is against His will?" The verbatim accounts underscored the deeply rooted beliefs that framed pre-natal screening as a potential transgression against divine will.

Cultural, religious, and personal beliefs are crucial in shaping individual choices. The right to choose whether or not to undergo prenatal screening and make informed decisions about pregnancy continuation. This requires access to comprehensive information about SCD, its severity, management options, and potential implications of a positive test. The study revealed that despite counselling couples on the above aspects, very few agreed to the termination of pregnancy. A positive SCD diagnosis during pregnancy triggers anxiety, grief, and uncertainty about the future.

> "The diagnosis did not affect me until the doctor informed me about the disease. It creates fear and sadness visualizing unborn child in pain." – A woman with SCD.

Couples find difficulties deciding on pregnancy continuation, abortion, or postnatal care. A pregnant woman said, "I am ready to do what the doctor suggests as they know better than us. However, abortion is a difficult decision to make." Societal views on SCD and pre-natal screening were unfavourable. Most participants (direct beneficiaries and critical stakeholders) were unaware of the genetic conditions of SCD and its implications, which can influence parental decisions. Furthermore, when the termination of pregnancy for SCD is grappled with stigma and disapproval from family and society, the emotional burden is multiplied, making it difficult for them to decide.

Providers' perspective

Providers may face challenges in balancing the potential for a happy, fulfilling life with SCD against the possibility of significant pain and suffering. Healthcare providers faced ethical challenges in navigating the delicate balance between respecting cultural beliefs and preventing the spread of SCD. A Healthcare professional said,

> "We face challenges as not all sickle cell disease is severe. In situations where the severity of SCD varies greatly, suggesting pre-natal screening becomes complex and suggesting abortion becomes even more complex." – A Healthcare Professional (Haematologist)

The manifestation of SCD is highly variable from very severe disease to very mild disease. It is not possible to predict the severity of the disease of the unborn child. Therefore, the providers face ethical challenges to find the right ethical balance between being assertive and giving space to couples to make decisions about PNS and termination. Providers must strike a delicate balance between assertively informing couples about screening options and respecting their autonomy in decision-making. This complexity is further highlighted by a case encountered by a counsellor. A pregnant woman stopped attending antenatal care services after repeated encouragement to terminate the pregnancy based on a high-risk screening result. This case underscores the emotional and social burdens associated with such decisions, potentially leading to service dropout. Recognizing this challenge, the counsellor proactively followed up with the woman and facilitated both antenatal & postnatal care for her. This dedication not only addressed her immediate healthcare needs but also instilled faith in the services offered by the SEWA Rural Hospital.

Low literacy rates make it difficult for beneficiaries to understand complex medical information, which hinders informed decision-making. Furtherance to this context, one participant raised two ethical questions:

> "I have two questions: (1) To what extent is it appropriate to promote pre-natal screening and selective abortion as reproductive choices? (2) Is presenting an opportunity to opt for abortion appropriate? I believe

our ability to answer both questions keeping in mind better health outcomes of patients can help resolve our ethical dilemma." –A Healthcare Professional (Gynaecologist)

Discussion

The study findings highlight a varied level of knowledge about pre-natal screening and its consequences within tribal populations, with comprehensive understanding among individuals with full SCD and limited awareness among those with SCD traits. Study participants – direct beneficiaries and community stakeholders—reported limited awareness about prenatal screening. Previous Indian studies reported limited knowledge about SCD, often fuelled by societal stigma (Aderotoye-Oni et al 2018; Gajbhiye, & Agarwal 2019; Gupta et al. 2014; Patil et al 2017). The recent study by Arumugam and colleagues (2024) reported inadequate awareness and information dissemination regarding pre-natal screening, underscores the need for culturally sensitive communication strategies to improve knowledge and empower women to access prenatal services.

Cultural and social factors significantly influence perceptions of pre-natal screening, with stigma and societal expectations adding complexity to decision-making (Gajbhiye, & Agarwal 2019; Williams et al 2018). Based on the explanation of pre-natal screening for early detection and prevention of SCD in unborn children, the study participants expressed mixed responses, particularly unfavourable responses for termination of pregnancy. The emotional and psychological impact of a positive pre-natal screening result shapes decisions regarding the termination of pregnancy, revealing a complex interplay of fear, societal pressure, and emotional burden. Similar findings were echoed in a previous study conducted on susceptibility loci for neurodevelopmental disorders by van de Steen and colleagues in 2016 (van der Steen et al 2016).

Several studies have explored the ethical complexities surrounding prenatal screening and abortion, highlighting the diverse perspectives and potential dilemmas faced by different stakeholders (Chima, & Mamdoo 2015; Fadare 2009; Gupta 2010; Hewson 2001). These studies emphasize the importance of respecting women's right to choose regarding testing and pregnancy continuation. Limiting the autonomy of women in regard to their reproductive choices also infringes on their individuality. Pregnant women facing decisions about PNS grapple with significant ethical and moral dilemmas. Rapp's (1988) concept of "moral pioneers" aptly captures the conundrum participants experience. Their anxieties extend beyond secular morality, entering a "magico-religious space" where anxieties about "going against God's will" or incurring a "curse" become prominent. This entanglement of morals and religion reflects the complex ways individuals navigate decisions surrounding pregnancy and potential disabilities.

The most emotionally charged element of the SCD-related ethics debate is the possibility of TOP. While some view it as a compassionate option, allowing couples to avoid passing on a potentially debilitating condition, others raise serious ethical concerns, questioning the right to terminate a fetus's life based on potential health limitations (Stapleton 2017). Cultural and religious beliefs further complicate the issue, as Kalita (2013) emphasizes, with social pressures often influencing individual choices and access to safe and medical termination of pregnancy. This complex interplay extends beyond individual anxieties. Recognizing these factors is crucial because, as Short and Mollborn (2015) suggest, our embodied experiences of health, illness, and well-being are shaped by a complex interplay of biological, social, and cultural factors. Knowledge about the body is not universal, but rather socially constructed within specific contexts. Recognizing these "local biologies" is essential for culturally sensitive healthcare practices, particularly when dealing with sensitive decisions like PNS and medical termination of pregnancy.

Presenting options without judgment and respecting individual values and cultural contexts should be prioritized and focused. Respecting religious and cultural beliefs that influence decisions regarding prenatal screening and medical termination of pregnancy while presenting reproductive options without judgment is of paramount importance. As, during tough times, religion and spirituality offer people comfort, guidance, and a sense of purpose (Puchalski 2001). Therefore, culturally sensitive counselling and support are essential and should be integral to Sickle Cell Disease services, including prenatal diagnosis and treatment services. Few studies highlight the need for counselling to incorporate an empathetic approach, respect for diverse beliefs, and avoid imposing their ethical frameworks on patients (Clarke, & Wallgren-Pettersson 2019; Colah et al 2005; Mohanty & Das, 2011; Munung et al 2024; Muthuswamy 2011).

Addressing ethical complexities requires a multifaceted approach that fosters informed decision-making, respects individual autonomy, and acknowledges the interplay of medical, social, and cultural factors. Healthcare professionals have a fundamental duty to respect patient autonomy and cultural beliefs, while also upholding their responsibility to prevent the spread of potentially debilitating diseases. Balancing these seemingly opposing goals creates significant tension. Forcing or pressuring patients to terminate a pregnancy based solely on the potential for SCD might violate their right to bodily autonomy and informed decision-making. However, not actively encouraging prenatal screening and termination options could potentially lead to

the continued birth of children with SCD, impacting both the individuals and healthcare systems in the long run. We propose strategies to address ethical dilemmas.

Ethical framework for prenatal screening and testing for SCD

First, ensuring access to accurate and comprehensive information about PNS and highlighting both the challenges and the possibilities are crucial. This information should be disseminated through culturally sensitive channels, debunking myths and promoting realistic narratives. Second, respecting individual autonomy. The ultimate decision regarding termination of pregnancy should rest with the couple after careful consideration of their beliefs, values, and personal circumstances. Third, non-directive genetic counselling that respects individual values and beliefs without advocating for or against specific choices becomes paramount. Counselling spaces should provide a safe environment for couples to explore their anxieties and concerns without judgment, ensuring informed and autonomous decision-making (Muthuswamy 2011; Clarke, & Wallgren-Pettersson 2019). Fourth, addressing social stigma and cultural misconceptions surrounding SCD, PNS and TOP through behaviour change communication (BCC) strategies. Open dialogues that acknowledge diverse perspectives and facilitate understanding can create a more supportive environment for families facing these difficult choices (Sirohi & Mishra 2015). Fifth, advocating for accessible and affordable prenatal screening and facilities for safe medical termination of pregnancy, particularly in underserved communities, becomes essential in ensuring equitable access to reproductive healthcare.

Implications for practice, research and policy

The study's findings offer valuable insights for improving healthcare practices related to prenatal screening for SCD, particularly within tribal communities. Few studies have highlighted the effectiveness of BCC strategies in promoting reproductive health awareness and uptake of services in tribal communities (Banerjee et al 2012, 2013; Jones et al. 2017). It can contribute to healthier pregnancies, informed family planning, and improved maternal and child health outcomes in these underserved communities. BCC strategies should focus on enhancing health literacy, addressing cultural norms and beliefs surrounding PNS, and providing access to genetic counselling. Effective BCC can empower individuals and families to make informed decisions about prenatal screening while navigating the complex ethical considerations involved. Culturally sensitive messaging, delivered through community influencers, can dispel myths, address fears, and emphasize the health benefits of informed decision-making. Preferred communication approaches can be: 1) information booklet on PNS in [local] language, 2) involve community leaders in communication campaign, 3) document and disseminate success stories, 4) use of pamphlets, hoardings, wall paintings in public places and 5) use of video interviews of beneficiaries and healthcare professionals.

Future longitudinal studies would provide valuable insights into the long-term impact of prenatal screening for SCD on individuals, families, and communities. Generating evidence on culturally tailored psychosocial support intervention would be crucial. Further research could explore strategies for more active community engagement in shaping healthcare policies and interventions related to SCD. This participatory approach can ensure that interventions are culturally relevant, address community needs effectively, and promote sustainable improvements in healthcare practices.

This study underscores the critical need for policy changes that prioritize improved access to PNS for SCD, particularly within tribal communities. Making PNS more readily available at district hospitals would significantly reduce geographical and logistical barriers that currently impede access to this crucial healthcare service. Additionally, healthcare providers should be equipped with the knowledge and resources to provide culturally competent care and support couples (and families) throughout the decision-making process.

Moving beyond the sole goal of eliminating SCD births, a crucial shift is needed: promoting universal access to prenatal screening (PNS) and termination of pregnancy (TOP) services, while simultaneously respecting reproductive autonomy and informed decision-making. This aligns with fundamental principles of reproductive rights.

Empowering couples requires comprehensive information, counselling, and support mechanisms. This ensures they can make choices based on their specific circumstances, values, and cultural contexts, rather than following external pressures. Family-centred counselling strengthens this approach further. By involving both partners and family members, healthcare providers can create a supportive environment that acknowledges the broader social and cultural influences on these complex choices. To achieve this, developing a guideline on Family-centered Genetic Counselling for Sickle Cell Disease is essential. Integrating this guideline into existing National Sickle Cell Disease Control Programmes would ensure a holistic approach. Ultimately, creating a healthcare system that provides access to PNS and TOP services, respects individual autonomy, and promotes informed decision-making through counselling is imperative for improving reproductive autonomy and the well-being of tribal communities.

Strength and limitations

This study is one of the first in India to our best knowledge to explore the perceptions and ethical considerations surrounding PNS for SCD. The study includes perspectives from pregnant women, persons with SCD, their families, community members, and healthcare professionals. This comprehensive approach captures the diverse experiences and viewpoints surrounding the issue, leading to a more nuanced understanding. The mixed-method approach provided a comprehensive understanding of the complex factors influencing the decision-making process around PNS for SCD. Furthermore, the study employed a participatory approach. Engaging the community throughout the research process fosters trust and collaboration, ensuring that the findings are grounded in the specific cultural context and resonate with the lived realities of the participants. This participatory approach can also contribute to the sustainability of any interventions or recommendations arising from the study. Despite numerous strengths, the authors acknowledge the focus on a specific tribal community in Jhaghadia, Gujarat. While the findings offer valuable insights into this particular context, they might not be directly generalizable to other populations or regions. Further research is needed to explore the broader applicability of these findings.

Conclusions

The study provides a comprehensive overview and highlights the complex interplay of knowledge, barriers, sociocultural norms, and ethical dilemmas surrounding sickle cell disease, prenatal screening and testing, and termination of pregnancy in Jhagadia block. Although couples with sickle cell disease favour pre-natal screening and, to some extent, termination of pregnancy, the larger community expressed low acceptance of pre-natal screening and has reservations towards the termination of pregnancy. Root causes highlight the need for culturally sensitive interventions that address deep-seated beliefs, alleviate fears of stigma, build trust in the healthcare system, and consider the complex interplay of gender dynamics and economic constraints. Efforts to bridge the gap between cultural beliefs and modern healthcare practices must be tailored to the local context to foster informed decision-making and improve maternal and child health outcomes.

Acknowledgements The authors gratefully acknowledge the active participation of all participants from the study's design phase onwards. We would also like to thank Dr Damini Joshi, Dr. Shubhangi Patel and Shweta Mulchandani from Parul University Vadodara, and Bakul Limbachiya from SEWA Rural for their invaluable support in facilitating data collection.

Authors contribution A.P., S.D. and G.D. conceptualized the study. A.P. designed the study, collected data, analyzed the data, and prepared the first draft manuscript; K.D. supported data collection, and critically reviewed the manuscript. All authors commented on previous versions of the manuscript. All authors read and approved the final manuscript.

Funding Apurvakumar Pandya has received research grants from SEWA Rural (Agreement-01–20/06/2023). Shrey Desai and Gayatri Desai are trustees of SEWA Rural, Jhagadia. Kapil Dave is an employee of SEWA Rural, Jhagadia. SEWA Rural is a charitable organization that operates a hospital in Jhagadia, Bharuch District, Gujarat state, India. This study's data collection, data analysis, and interpretation of results were solely performed by Apurvakumar Pandya. Kapil Dave, Shrey Desai, and Gayatri Desai, who are affiliated with SEWA Rural, have not influenced the research process, data analysis, and interpretation of results.

Data availability Data is provided within the manuscript.

Declarations

Ethical approval This study received ethical approval from the Institutional Ethics Committee of SEWA Rural (SEWARural IEC/01 dated 09/08/2023). All procedures were conducted in accordance with ethical standards, both at the institutional and national levels, and aligned with the Helsinki Declaration of 1975, as revised in 2000. Prior to participation, all potential participants were fully informed about the study's objectives, procedures, and their right to withdraw at any time. Of the total individuals contacted, two individuals did not participate. Written informed consent was obtained from all participants. The privacy and confidentiality of all participant data were strictly maintained throughout the study.

Competing Interests Apurvakumar Pandya declare that he has no conflicts of interest. Apurvakumar Pandya has received research grants from SEWA Rural (Agreement-01–20/06/2023). Shrey Desai, and Gayatri Desai are trustee of SEWA Rural, Jhagadia. Kapil Dave is an employee of SEWA Rural, Jhagadia. SEWA Rural is a charitable organization who operates a hospital in Jhagadia, Bharuch District, Gujarat state, India. This study's data collection, data analysis, and interpretation of results was solely performed by Apurvakumar Pandya. Kapil Dave, Shrey Desai, and Gayatri Desai, who are affiliated with SEWA Rural, have not influenced the research process, data analysis, and interpretation of results.

References

Aderotoye-Oni S, Diaku-Akinwumi IN, Adeniran A, Falase B (2018). Unprepared and misinformed parents of children with sickle cell disease: Time to rethink awareness campaigns. Cureus 31;10(12):e3806. https://doi.org/10.7759/cureus.3806

Arumugam S, Kalluri SS, Sharmila V, Mocherla A, Subbiah NK, Kulkarni JP, Ghoshal JA (2024) Understanding the awareness of prenatal genetic screening tests among pregnant women in India: A cross-sectional study. Cureus 16(3):e56932. https://doi.org/10.7759/cureus.56932

Ashorobi D, Naha K, Bhatt R (2023) Hematopoietic stem cell transplantation in sickle cell disease. In: StatPearls [Internet]. Treasure Island (FL): StatPearls Publishing. Available from: https://www.ncbi.nlm.nih.gov/books/NBK538515/

Banerjee SK, Andersen KL, Buchanan RM et al (2012) Woman-centered research on access to safe abortion services and implications for behavioral change communication interventions:

a cross-sectional study of women in Bihar and Jharkhand. India BMC Public Health 12:175. https://doi.org/10.1186/1471-2458-12-175

Banerjee SK, Andersen KL, Warvadekar J, Pearson E (2013) Effectiveness of a behavior change communication intervention to improve knowledge and perceptions about abortion in Bihar and Jharkhand, India. Intl Pers Sexl Reproduct Health 142–151. https://www.jstor.org/stable/2359612

Baum F, MacDougall C, Smith D (2006) Participatory action research. J Epidemiol Community Health 60(10):854–857. https://doi.org/10.1136/jech.2004.028662

Beck CE, Trottier ED, Kirby-Allen M, Pastore Y (2022) Acute complications in children with sickle cell disease: Prevention and management. Paediatrics Child Health 27(1):50–62. https://doi.org/10.1093/pch/pxab096

Bernard, H. R. (2002). Research methods in anthropology: Qualitative and quantitative approaches (3rd ed.). Walnut Creek, CA: Altamira Press

Bourdieu, P. (1986). *Handbook of theory and research for the sociology of education*. Greenwood Press

Braun V, Clarke V (2006) Using thematic analysis in psychology. Qual Res Psychol 3(2):77–101

Brousse V, Rees DC (2021) Sickle cell disease: More than a century of progress. Where do we stand now? Indian J of Med Res 154(1):4–7. https://doi.org/10.4103/ijmr.ijmr_1435_21

Census of India (2011) Bharuch District Population, Caste, Religion Data (Gujarat) - Census 2011. Retrieved from https://www.censusindia.co.in/district/bharuch-district-gujarat-488. Accessed 31 May 2023

Charmaz K (2006) Constructing grounded theory: a practical guide through qualitative analysis. Sage Publications, London, Thousand Oaks, Calif. https://archive.org/details/constructinggrou0000char

Chavda J, Goswami P, Goswami A (2015) Hematological profile of sickle cell disorder in tertiary care hospital. IOSR J Dent Med Sci 14:51–54. https://doi.org/10.18203/2320-6012.ijrms20195902

Chima SC, Mamdoo F (2015) Ethical and legal dilemmas around termination of pregnancy for severe fetal anomalies: A review of two African neonates presenting with ventriculomegaly and holoprosencephaly. Nigerian J Clin Pract 18(Suppl):S31–S39. https://doi.org/10.4103/1119-3077.170820

Clarke AJ, Wallgren-Pettersson C (2019) Ethics in genetic counselling. J Community Genet 10(1):3–33. https://doi.org/10.1007/s12687-018-0371-7

Colah R, Surve R, Nadkarni A, Gorakshakar A, Phanasgaonkar S, Satoskar P, Mohanty D (2005) Prenatal diagnosis of sickle syndromes in India: dilemmas in counselling. Prenat Diagn 25(5):345–349

Colah RB, Mukherjee MB, Martin S, Ghosh K (2015) Sickle cell disease in tribal populations in India. Indian J Med Res 141(5):509–515. https://doi.org/10.4103/0971-5916.159492

Creswell JW (2013) Qualitative inquiry & research design: choosing among five approaches, 3rd edn. SAGE, Thousand Oaks, CA

Desai G, Dave K, Banerjee S, Babaria P, Gupta R (2016) Initial outcomes of a comprehensive care model for sickle cell disease among a tribal population in rural Western India. Int J Community Med Public Health 3:1282–1287. https://doi.org/10.18203/2394-6040.ijcmph20161399

Fadare JO (2009) Some ethical issues in the prenatal diagnosis of sickle cell anaemia. Ann Ib Postgrad Med 7(2):26–28. https://doi.org/10.4314/aipm.v7i2.64084

Gajbhiye V, Agarwal S (2019) Stigma associated with sickle cell disease in India: Perception of patients and healthcare providers. Intl J of Comm Med 10(8):553–557

Gupta JA (2010) Exploring Indian women's reproductive decision-making regarding prenatal testing. Cult, Heal Sexu 12(2):191–204. https://doi.org/10.1080/13691050903429316

Gupta D, Mishra R, Saxena R (2014) Knowledge and awareness of sickle cell disease among antenatal women in North India. Intl J of Reprod Contrace Fam Plann 3(4):14–19

Hardouin G, Magrin E, Corsia A, Cavazzana M, Miccio A, Semeraro M (2023) Sickle cell disease: from genetics to curative approaches. Annu Rev Genomics Hum Genet 24(1):255–275. https://doi.org/10.1146/annurev-genom-120122-081037

Hewson B (2001) Reproductive autonomy and the ethics of abortion. J Med Eth, 27 Suppl 2(Suppl 2), ii10–ii14. https://doi.org/10.1136/jme.27.suppl_2.ii10

Hockham C, Bhatt S, Colah R et al (2018) The spatial epidemiology of sickle-cell anaemia in India. Sci Rep 8:17685. https://doi.org/10.1038/s41598-018-36077-w

Jones E, Lattof SR, Coast E (2017) Interventions to provide culturally-appropriate maternity care services: factors affecting implementation. BMC Pregnancy Childbirth 17(1):267. https://doi.org/10.1186/s12884-017-1449-7

Kalita T (2013) Interpretation of the problem of abortion: Issue of foetus killing in the light of traditional and contemporary ethics. Accessed on 12.12.2018, Retrieved from: gyan.iitg.ernet.in/bitstream/handle/ 123456789/400/TH-1177_ 07614101.pdf?...1

Kishore RR, Gupta M, Gupta K (2023) A new era dawns on sickle cell disease in India. Indian J Med Res 157(6):491–493. https://doi.org/10.4103/ijmr.ijmr_1045_23

Linton EA, Williams EC, Early ML, Prince E, Stewart RW, Lanzkron S, Pecker LH (2023) Family planning needs of young adults with sickle cell disease. Ejhaem 4(3):587–594. https://doi.org/10.1002/jha2.711

Mohanty D, Das K (2011) Genetic counselling in tribals in India. Indian J Med Res 4:561–571

Munung NS, Kamga KK, Treadwell MJ, Dennis-Antwi J, Anie KA, Bukini D, Makani J, Wonkam A (2024) Perceptions and preferences for genetic testing for sickle cell disease or trait: a qualitative study in Cameroon, Ghana and Tanzania. Eur J Hum Genet. 2024. https://doi.org/10.1038/s41431-024-01553-7.

Muthuswamy V (2011) Ethical issues in genetic counselling with special reference to haemoglobinopathies. Indian J Med Res 134(4):547–551

National Heart, Lung, and Blood Institute (n.d.) *What is sickle cell disease?* Retrieved from https://migrc.org/resource/nhlbi-sickle-cell-disease-management-guidelines/. Accessed 31 May 2023

National Sickle Cell Elimination Mission (2023) Ministry of Health and Family Welfare, Government of India. Retrieved from https://sickle.nhm.gov.in/home/guidelines. Accessed 31 May 2023

Panepinto JA (2012) Health-related quality of life in patients with hemoglobinopathies. Hematology Am Soc Hematol Educ Program 2012:284–289

Patel J, Patel B, Gamit N, Serjeant GR (2013) Screening for the sickle cell gene in Gujarat, India: A village-based model. J Community Genet 4:43–47. https://doi.org/10.1007/s12687-012-0116-y

Patil SS, Thikare AA, Wadhva SK, Narlawar UW, Shukla S (2017) Knowledge, attitude and practice regarding sickle cell disease in adult sufferers and carriers in a rural area. Int J Community Med Public Health 4(4):1075–1080. https://doi.org/10.18203/2394-6040.ijcmph20171327

Piel FB, Hay SI, Gupta S, Weatherall DJ, Williams TN (2013) Global burden of sickle cell anaemia in children under five, 2010–2050: modelling based on demographics, excess mortality, and interventions. PLoS Med 10(7):e1001484. https://doi.org/10.1371/journal.pmed.1001484

Platt O, Dornelas G, Shackleton S (2017) Sickle cell disease. New Eng J Med 376(23):2396–2406

Puchalski CM (2001) The role of spirituality in health care. Proc Bayl Univ Med Cent 14(4):352–357. https://doi.org/10.1080/08998280.2001.11927788

Rapp R (1988) Moral pioneers: Women, men and fetuses on a frontier of reproductive technology. Women Health 13(1–2):101–117. https://doi.org/10.1300/J013v13n01_09

Saxena D, Yasobant S, Golechha M (2017) Situational analysis of Sickle Cell Disease in Gujarat. India Indian J Community Med 42(4):218–221. https://doi.org/10.4103/ijcm.IJCM_284_16

Serjeant GR, Ghosh K, Patel J (2016) Sickle cell disease in India: A perspective. Indian J Med Res 143(1):21–24. https://doi.org/10.4103/0971-5916.178582

Shiladaria DP, Patel DS, Oza HV (2013) Prevalence of anemia, thalassemia, and sickle cell disorder in young adults of Gujarat. Natl J Integr Res Med 4:61–3. Retrieved from https://nicpd.ac.in/ojs-/index.php/njirm/article/view/2207

Short SE, Mollborn S (2015) Social Determinants and Health Behaviors: Conceptual Frames and Empirical Advances. Curr Opin Psychol 5:78–84. https://doi.org/10.1016/j.copsyc.2015.05.002

Sickle Cell Anemia Control Program (2013) A new initiative of Government of Gujarat: Best health practices. Gujarat Health and Family Welfare Department, Gujarat. [Last accessed on 2024 April 11]. Retrieved from: http://www.sickle.cell.gujarat.gov.in/Docs/News_GR_NC/131120_120132.pdf\

Sickle Cell Foundation (2023) About sickle cell. Retrieved from https://www.sicklecelldisease.org/. Accessed 31 May 2023

Sirohi N, Mishra A (2015) Ethical challenges in managing sickle cell disease in India. J Clin Res Bioeth 1(1):34–43

Stapleton G (2017) Qualifying choice: ethical reflection on the scope of prenatal screening. Med, Health Care Philos 20(2):195–205. https://doi.org/10.1007/s11019-016-9725-2

Tewari S, Rees D (2013) Morbidity pattern of sickle cell disease in India: a single centre perspective. Indian J Med Res 138(3):288–290

van der Steen SL, Riedijk SR, Verhagen-Visser J, Govaerts LC, Srebniak MI, Van Opstal D, Joosten M, Knapen MF, Tibben A, Diderich KE, Galjaard RJ (2016) The psychological impact of prenatal diagnosis and disclosure of susceptibility loci: first impressions of parents' experiences. J Genet Couns 25(6):1227–1234. https://doi.org/10.1007/s10897-016-9960-y

Vaughn LM, Jacquez F (2020) Participatory Research Methods – Choice points in the research process. J Part Res Meth, 1(1). https://doi.org/10.35844/001c.13244

Wallerstein I (1979) The capitalist world-system. Academic Press

Williams H, Silva RNS, Cline D, Freiermuth C, Tanabe P (2018) Social and Behavioral Factors in Sickle Cell Disease: Employment Predicts Decreased Health Care Utilization. J Health Care Poor Underserved 29(2):814–829. https://doi.org/10.1353/hpu.2018.0060