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ORIGINAL STUDY

Knowledge, Attitude, and Perception of the General Public Towards the Impact of Hemoglobinopathies on Pregnancy Outcomes: A Questionnaire-based Study

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Abstract

Background: Knowledge about hemoglobinopathies in the general public is unsatisfactory. Many couples are unaware of their carrier status and a child can be born with unexpected, serious hemoglobinopathies. Willingness to participate in screening programs can substantially help in bringing down the birth of children affected with hemoglobinopathies. To understand the knowledge, attitude, and perception of the general public on various hemoglobinopathies and their pregnancy outcomes we performed a questionnaire-based descriptive survey.

Methods: After obtaining approval from the institutional ethics committee and validation by an expert panel we circulated a Google form consisting of 21 questions via WhatsApp which was made available for 3 months. Efforts were made to collect as many responses as possible. The responses were extracted onto a Microsoft Excel sheet and analyzed.

Results: It was observed that though the majority of the population from various sectors of India agreed on the importance of early detection, only about 75.5% would consider signing up for screening themselves. Around 33.6% percent of the participants had not even heard about hemoglobinopathies and about 92.7% of the participants felt the need for the availability of more easily accessible information on the topic.

Conclusion: The study concludes that there is a need for more awareness programs among the general public as well as better access to information on hemoglobinopathies so that the implications of being affected are clearly understood, and strategies for prevention by timely screening can be strengthened.

Keywords: Knowledge, Attitude, Perception, Hemoglobinopathies, Pregnancy outcomes, Awareness

1. Introduction

The World Health Organization (WHO) estimates that overall, at least 5.2% of the World's population carries a significant variant of a hemoglobin disorder; among pregnant women, the prevalence is even higher at over 7% [1]. India has a huge burden of hemoglobin disorders with an estimated 1,00,000 patients with β -thalassemia syndrome and around 1,50,000 patients with sickle cell disease, but few among them are optimally managed [2]. β -Thalassemia is prevalent across the

country, with an average frequency of carriers being 3–4%. HbS is highly prevalent in the tribal populations of Southern, Central, and Western states reaching as high as 48% in some communities. HbE also has a carrier frequency as high as 50% in some areas [3].

Yet, knowledge about this condition in the general public is unsatisfactory. Despite current screening recommendations, many couples are not aware of their carrier status and hence it is common for a child to be born with unexpected, serious hemoglobinopathies [4]. As per a study, the awareness

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amongst pregnant women was found to be very limited ranging from 0.2% to 20.7% depending on the region [2]. In a similar study, patients who were antenatally registered and had a regular follow-up, along with their spouses, were found more likely to have undergone screening for hemoglobinopathies. It was demonstrated that appropriate counseling improves acceptance of screening methods among at-risk patients and their relatives [5].

Through consistent prenatal screenings and preventive tests, the life span of the mother/fetus/newborn may be improved considerably. Experience suggests that adherence to careful management principles will increase the likelihood that significant crises and infections can be reduced, thus, resulting in a healthier pregnancy outcome [6]. Basic knowledge amongst the general public about the risks of hemoglobinopathies is a prerequisite to increasing the acceptance of prenatal screening tests. Willingness to participate in screening programs can substantially help in bringing down the birth of children affected with hemoglobinopathies over time. Hence, it is of utmost importance to evaluate the awareness of the general public on this subject. Therefore, this questionnaire-based study was conducted to assess the knowledge, attitude, and perception of the general public on various hemoglobinopathies and their pregnancy outcomes.

2. Material and methods

We performed a descriptive survey using a convenient sampling technique on the general public as the study population after obtaining approval from the institutional ethics committee (vide number JSS/IEC/13042022/22NCT/2021-22 dated 25.04.22). A questionnaire set consisting of 21 questions was prepared and widely circulated on social media and efforts were made to collect as many responses as possible.

Data collection procedures and instruments used – The content validity index (CVI) and content validity ratio (CVR) were calculated for each question to evaluate its relevancy and clarity. The content validation of the questions was conducted by arranging an expert panel meeting consisting of 10 members via an online platform. In the meeting, the panelists were outlined about the survey, and the content validation form was sent to them which contained the guidelines to review the questionnaire. Each question in the questionnaire was given a score between 1 and 4 based on the relevance and later the scores were recoded as 0 for 1 & 2 and 1 for 3 & 4 respectively. CVI was calculated using the formula: $CVI = \text{questions in agreement with the}$

panelist/total number of the panelist. Further, we investigated CVI using item level CVI (I-CVI) and content validity index for scales (S-CVI) indexes. $I-CVI = N_p/N$ where N_p = number of panelists voting 3 or 4 and N = total number of panelists. I-CVI ranges from 0 to 1, $I-CVI > 0.79$, the item is relevant, between 0.70 and 0.79, the item needs revisions, and if the value is below 0.70 the item is eliminated. S-CVI (Average) was calculated by the number of items considered relevant (score 3 or 4 or the number of items with CVI equal to 1) divided by the total number of items. $S-CVI/Ave \geq 0.9$ has excellent content validity. CVR was calculated using the formula: $CVR = [(E - (N/2))/(N/2)]$, where E is the total number of expert panelists who rated the question as relevant and N is the total number of panelists. The questionnaire was then circulated online via Survey Monkey-based platforms and was made available for 3 months. The questionnaire was administered in English. The participants enrolled in the study belonged to any professional/non-professional background and were above 18 years of age. Uneducated individuals and those below 18 years of age were excluded from the study.

The questionnaire began with an introduction stating details of the study and consent for participation was obtained. Participants voluntarily answered the questions. The participants' email and information related to age, sex, state, and district of residence as well as qualification and profession was collected. The second part of the questionnaire gathered information related to their basic knowledge about hemoglobinopathies, the prevalence in their region, familiarity with the disease and its complications, the need for implementing screening programs, methods for primary prevention, impact on pregnancy outcomes, the burden of having a child with hemoglobinopathy, further course of action, and the need to expand knowledge.

Data analysis –The responses obtained were extracted onto a Microsoft Excel sheet and analyzed.

3. Results

The questions used for the survey were considered suitable with scores for CVI, I-CVI, S-CVI, and CVR of 0.89, 0.98, 0.98, and 0.97 respectively. The study observed the participation of ~61% females and ~39% males out of a total of 330 participants. Responses were received from people belonging to a total of 19 states (Fig. 1). Maximum participation was observed from Haryana (53%), followed by Karnataka (18%) and New Delhi (12%). A total of 53% of the participants belonged to various non-medical fields including, but not limited to daily wage workers,

Count of State/U.T.

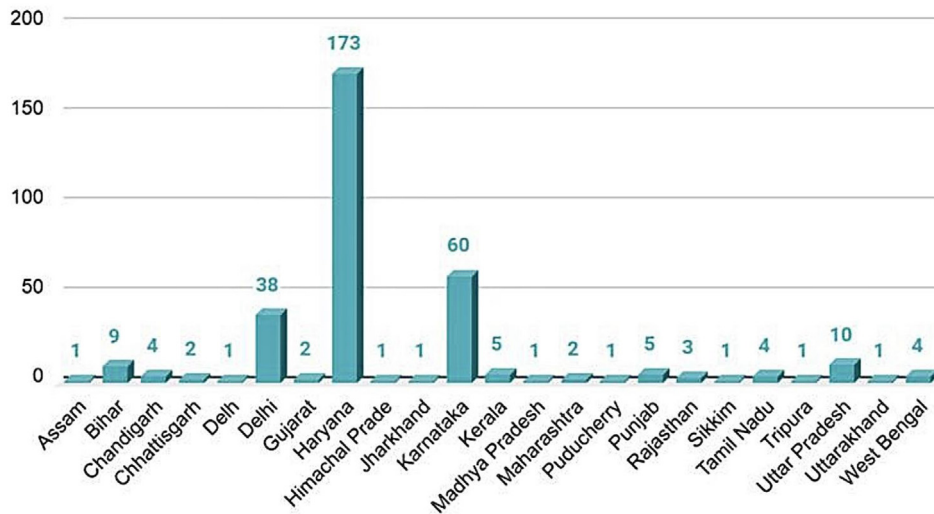


Fig. 1. State-wise distribution of participating population.

farmers, teachers, business persons, office workers, accountants, and homemakers. Doctors constituted 20% of the total participants. Out of the 27% of students, around 55% were currently pursuing MBBS. The majority of the subjects had completed or were pursuing their post-graduation ($N = 175$), followed by undergraduates ($N = 67$), graduates ($N = 58$), and finally, 30 people who had completed their education up to 12th grade.

Approximately 66% of the participants had heard about the disorder hemoglobinopathy while approximately 34% ($N = 111$) had not heard about this condition. Their details are shown in Table 1. Around 22.6% of the participants were acquainted with an individual who suffered from hemoglobinopathy. The affected person was found to mostly be

Table 1. Profession-wise distribution of participants unfamiliar with hemoglobinopathies ($N = 111$).

Profession	Numbers
Students	22
Teachers	21
Engineers	20
Homemakers	15
Office workers	14
Army officers	3
Lawyers	6
Lab technicians	3
Farmers	1
Business persons	6
Education	Numbers
Postgraduates	50
Graduates	34
Undergraduates	27

a family member. Friends, co-workers, and relatives of some of the participants were also affected by the same. The majority (93.5%) of the participants chose the correct definition for hemoglobinopathies among the four options given. Conflicting opinions were observed when asked about grading the seriousness of hemoglobinopathies. Around 70.9% of individuals believed that the severity of said disease ranged from moderate to severe while 24.2% were uncertain regarding the severity of the same. A preponderance of agreement (93.2%) for the importance of screening was noted (Table 2).

The popular opinion (93.8%) recognized that it is important to identify carriers in early pregnancy. Approximately 94.1% of the participants agreed that primary prevention should be a priority whereas 0.9% disagreed with the same and 5% remained neutral. A majority i.e. 87% of the subjects agreed that prenatal screening during pregnancy can reduce risks while 10.2% stayed neutral. Similarly, 86.3% of the subjects opted to agree with secondary prevention being carried out via newborn screening, while 10% remained neutral and 3.8% disagreed. Most of the subjects (87.2%) opined that complications may be fatal, however, 3.4% disagreed. Around 82.9% of the participants think that raising an affected child may be mentally exhausting for parents. A still large percentage of the subjects (64.2%) believed that the treatment may not be affordable while 26.8% were neutral. A significant majority (92.8%) were found to agree that genetic counseling would have its benefits.

The popular opinion (93.5%) went in favor of increasing awareness to possibly reduce the burden

Table 2. Percentage of respondents for each item in the questionnaire.

Questionnaire	Agree (%)	Neutral (%)	Disagree (%)
It is important to screen for hemoglobinopathies during pregnancy	93.2	5.6	1.2
It is important to identify carriers in early pregnancy	93.8	5.3	0.9
Primary prevention (like screening, diagnostics, etc) should be the priority for at-risk couples	94.1	5	0.9
Prenatal screening during pregnancy can reduce the chance of the birth of a child with hemoglobinopathy	87	10.2	2.8
Secondary prevention can be done by screening the disorder in newborns	86.3	10	3.8
Complications due to hemoglobinopathies may be fatal	87.2	9.4	3.4
Raising a child with hemoglobinopathy can take a mental toll on the parents	82.9	13.4	3.7
The treatment for an affected newborn will not be affordable by the family	64.2	26.8	9
Genetic counseling would be helpful for at-risk couples	92.8	6.2	1
Increasing awareness can reduce the burden of hemoglobinopathies in the country	93.5	4.6	1.9
Is enough information readily accessible regarding hemoglobinopathies	51.4	25.2	23.4
Information on this subject should be made more readily available to the general public	92.7	4	3.3
Better access to information on the subject would reduce the risk of affected newborns	91.6	6.5	1.9
There is a need for public information campaigns on hemoglobinopathies	93.4	6.2	0.4
Would you advise screening to an acquainted, expecting couple?	90.3	7.8	1.9
Would you ever consider signing up for a screening program?	75.5	21.3	3.2

of diseases. Interestingly enough, a large portion of the participants (51.4%) were of the opinion that there is not enough information available about hemoglobinopathies and an overwhelming majority (92.7%) felt the need to make more information available to the general public. Furthermore, about 91.6% of the subjects believed that better access to information would reduce the risk of affected newborns. Also, it was noticed that the whopping majority (93.4%) was in favor of having more public information campaigns on the same and 90.3% of the participants showed interest in advising screening to an expecting couple. When it came to the subjects, however only 75.5% would consider signing up for screening themselves while 21.3% remained neutral.

4. Discussion

As indicated by the results of the study, a diverse population was engaged in the survey though the distribution was a bit skewed towards the native state and profession of the principal investigator which is understandable. All the participants had received basic education with a majority having education levels up to post-graduation.

Only ~66% of the participants had heard about hemoglobinopathy. Still, most of them were having a degree in the medical field or were related to someone affected by any hemoglobinopathy. The

majority of this group was found to have a fundamental understanding of the disease hemoglobinopathy and the complications associated with it. Most of them agreed upon the importance of screening, especially at the prenatal stage, in reducing the risk of the disease. Genetic counseling was also considered significant as bringing up a child with hemoglobinopathy may be mentally and financially exhaustive for most families. There was a general agreement in the study population about increasing awareness about the disease in the general population through public information campaigns and other measures.

Approximately 34% of the participants were not aware of hemoglobinopathies. It is quite concerning as this group included well-educated professionals mainly (Table 1). The data retrieved from the present study provides an insight into the gap present between the information possessed by the general public regarding hemoglobinopathies and their pregnancy outcomes currently and the amount of information necessary to bring down such cases. When the well-educated population is not in complete comprehension of genetic disorders like hemoglobinopathies, the situation of the illiterate can easily be imagined. It is warranted that appropriate measures may be taken to improve the existing knowledge gap which heralds the capacity to decrease the instances of such diseases to the lowest possible level.

The importance of screening for the detection of hemoglobinopathies is well perceived by the population as the majority of the participants claimed to advise screening to an expecting couple. Although when it came to the subjects themselves, not many agreed to undergo screening themselves. On the other hand, 3.2% disagreed to sign up for the screening program. Screening refers to the medical examination of an asymptomatic population for the early detection or exclusion of a clinical condition. Early detection of the disease is always favorable and may be life-saving in some conditions [7]. Therefore, in 1968, the World Health Organization (WHO) published “Principles and Practice of Screening for Disease” as a guideline to evaluate whether screening would be beneficial for a variety of conditions, this is still followed after contemporary amendments [8]. A type of screening known as genetic carrier screening is used to identify couples at high risk of carrying a child with a recessive disorder. When both partners are carriers of the same autosomal recessive disorder, there is 1 in 4 chance (25%) of having an affected child in each pregnancy and when the female is a carrier of an X-linked disorder, there is a 1 in 2 (50%) chance for sons to be afflicted with the disease and a 1 in 2 chance (50%) for daughters to be carriers of disease. It has been reported that there are more than 1800 recessively inherited rare diseases, whose symptoms may range from very mild to severe extent [9,10]. Various options available to the couples include; not having children, using a gamete donor, preimplantation genetic diagnosis or embryo selection, prenatal diagnosis with the option of termination if the fetus is affected by the condition, and avoidance of marriage to another carrier. However, the variety of options and their acceptance vary greatly with culture and ethnicity [11,12].

Under National Health Mission, the Government of India is presently involved in the implementation of a prevention and control program for 2 major types of hemoglobinopathies i.e. thalassemia major and sickle cell disease. For this, guidelines for the prevention and timely management of these diseases have been formulated and measures are being taken to increase awareness among the general population about these to avert a public health crisis [13].

A paradigm shift is being observed from communicable to non-communicable diseases in the Indian population, with congenital defects being among the 20 top commonest diseases. The genetic diversity of the Indian population, with large numbers of endogamous ethnic, geographical, religious, and social groupings adds to the complexity of the prevalence and burden of genetic disorders [14,15].

Therefore, policymakers must devise cost-effective preventive strategies to reduce the burden of genetic diseases. In India, screening for the detection of carriers in school-going adolescents and/or of all pregnant women is the most feasible prevention strategy to reduce the risk of hemoglobinopathies besides improving genetic literacy in the population [16].

5. Conclusion

The study concludes that there is a need for more awareness programs among the general public as well as better access to information on hemoglobinopathies so that the implications of being affected are clearly understood, and strategies for prevention by timely screening can be strengthened.

Statement by authors

The manuscript has been read and approved by all the authors.

The requirements for authorship have been met for all authors.

Each author believes that the manuscript represents honest work.

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Conflicts of interest

None.

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