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Subjective Valuation of Screening for Spinal Muscular Atrophy and Analysis of its Influencing Factors: Evidence from Iran

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Abstract

Background: Spinal muscular atrophy is an inherited neurodegenerative disorder that typically leads to severe physical disability. The present study aimed to determine the subjective evaluation of this disorder screening and analyze its influencing factors in Iran.

Methods: A cross-sectional study was performed using data from the second survey of women either pregnant or planning to become pregnant in Tehran, the capital of Iran, in 2022. The dependent variable was the willingness to pay for this disease screening test. The independent variables included sociodemographic, economic, and health characteristics, the history of this disease or other diseases of the person and family, and knowledge about this disease in the included population. Logistic regression was utilized to identify independent variables associated with the dependent variable, and the results were reported as unadjusted and adjusted odds ratios and P values with 95% CIs. A questionnaire was used as a research tool, and STATA 17 software was used for data analysis. The monetary value of spinal muscular atrophy (SMA) screening was calculated by estimating willingness to pay using the congenital valuation method.

Results: In total, 578 women were included. About 64.85% of respondents had a willingness to pay for SMA screening as the dependent variable, with a mean of $526. University education ($P = 0.009$) and pregnancy experience ($P = 0.021$) were associated with the dependent variable.

Conclusion: Iranian women expressed their willingness to undergo screening tests, but due to financial constraints, they expected the government and nongovernmental organizations to bear most of the cost.

Keywords: Spinal Muscular Atrophy, Willingness to Pay, Carrier Screening, Contingent Valuation Method, Logistic Regression Model

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Introduction

Among the motor neuron diseases, spinal muscular atrophy (SMA) is one of the most common ones (1), caused by the mutation or deletion of the motor neuron survival gene (2). SMA is classified into 4 types according to the age of onset and the severity of the clinical course. Type 1, known as Werding-Hoffman disease, is the most severe and common. Approximately 60% to 70% of affected patients die in the first 2 years of life due to respiratory failure. Type 2, or Dubowitz disease, is moderate and starts before 18 months of age. Patients of this type can sit but cannot stand or walk without emergency help. Type 3, or Kugelberg–Welander disease, begins in adolescence,

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↑What is "already known" in this topic:
Spinal muscular atrophy (SMA) is a hereditary neurological disease that usually leads to severe physical disability and has dire consequences on the health of patients and their families.

→What this article adds:
This study is the first to examine the subjective value of willingness to pay (WTP) for the SMA screening carrier test and its related factors in Iran. More than half of the respondents stated that they would be willing to pay out-of-pocket for an SMA screening test.

and patients have a normal life expectancy and moderate disability compared to other types (3, 4). Type 4 or adult-onset SMA is the mildest type of this disease, which the patient and his family may not notice until adulthood (5, 6).

This disease is a prevalent issue in various regions of the world. In 2017, in a study on the prevalence of SMA, the global average was about 8 children with this disease per 100,000 births, with a variation between 5 and 24 children (7). In European countries, Australia and the United States, the estimated prevalence of this disease is less than 9000 patients (8). A study in Italy revealed that 6.56 of every 100,000 patients younger than 20 years had SMA (9).

Type 1 is the most common type of SMA in most countries, including Iran, where precise epidemiological information is not available because infants with this type of SMA usually do not survive the first 2 years of life. In addition, accurate disease diagnosis and recording of SMA cases and deaths are complex (10, 11). Family marriages are common in Iran. In some areas, they can make up half of all marriages. Unfortunately, this practice has led to a high incidence of genetic disorders in the country (12). Studies have shown that the frequency of SMA carriers in Iran is 1 in every 20 people, which is relatively high among Iranian couples (13, 14). In 2022, the prevalence of SMA in Iran was 23.1% (95% CI, 21.2-25.1). This rate was equal to 23.8% in men, 22.5% in women, and 26% among teenagers and young adults (15).

There are 2 screening methods for SMA—carrier and newborn. Carrier screening is one of the fastest ways to diagnose SMA before or during pregnancy (16). Undergoing prenatal screening has the significant advantage of reducing the risk of giving birth to an infected child, leading to a safer pregnancy and delivery experience (17, 18). The American College of Medical Genetics recommends carrier testing for all couples (5, 19). According to research, specific studies have not been conducted on the economic evaluation and costs caused by SMA carrier screening, and most studies are on the economic evaluation of newborn screening. Cost-effectiveness analysis of SMA newborn screening in the United States concluded that newborn screening with presymptomatic treatment for positive SMA type 2 tests incurred a total cost of $3,150,087 and produced 269,997 quality-adjusted life years (QALYs). For 10,000 newborns without SMA screening plus symptomatic treatment for SMA type 1, the QALYs generated were 269,988 at a total treatment cost of $2,628,116 over a lifetime horizon. The incremental cost per QALY gained was $57,969/QALY compared with no SMA screening (20).

Considering the high cost of SMA treatment drugs and the resulting physical and mental difficulties on the patient and their families, as well as the absence of universal screening for this disease in Iran due to its risks and aggravating factors such as the prevalence of family marriages in some regions of the country and the lack of simultaneous genetic tests and counseling in this case before marriage, it seems that it is necessary to conduct research in the field of valuing the willingness to pay for the screening of this disease. Therefore, this study is the first in Iran on the subjective evaluation of SMA screening by estimating willingness to pay (WTP—the maximum price the consumer is willing to pay for a particular product or service—and analyzing its influencing factors (21). In general, WTP estimates can be extracted with different questionnaire formats used in conditional valuation research, such as bidding games, dichotomous choice, open-ended, and payment scale formats (22). The dichotomous selection approach is divided into 2 types—single-bounded dichotomous choice and double-bounded dichotomous choice (23).

Methods

Study Design and Sampling Method

This study was conducted as a cross-sectional population-based survey from September 23, 2022, to January 20, 2023, in specialized clinics and obstetrics and gynecology hospitals in Tehran, the capital of Iran. The study population included women who were pregnant or trying to conceive (N = 578). The questionnaire was randomly delivered to anyone who could answer anonymously. The sample size was defined according to Pourhoseingholi et al and Mitchell and Carson (24, 25).

This study used a convenient sampling method that helps establish a potential hypothesis or study objective. This strategy is common in demographic research. One advantage of this type of sampling is that it is simple, affordable, and time-efficient (26).

The monetary value of SMA screening was calculated by estimating WTP using the congenital valuation method (CVM). This method has been confirmed in various scientific sources (27, 28). Focusing on rare congenital genetic diseases, especially this disease, and designing search keywords in this regard, we found 10 articles from different scientific sources and studies to create a scenario and identify factors influencing WTP for the SMA screening test. Two articles were about SMA screening and treatment (29, 30), while the others focused on rare genetic diseases (31-38). After analyzing these articles, we extracted various factors as independent research variables. An expert panel of gynecologists, geneticists, epidemiologists, and health economists reviewed the validity of the extracted factors and recommended their suggestions.

After reviewing the questionnaires of different studies—Donaldson et al (25, 26), Lin et al (23), Norström et al (30), and Anunsittichai et al (31)—and their validity and reliability, we designed the questionnaire for this study.

At the start of the questionnaire, an introduction was given about the purpose of the research, the necessity of its implementation, and the obligation to keep the data confidential. Then, 40 questions were asked about the factors as independent variables in different dimensions as follows:

Sociodemographic and Economic Characteristics

This included age, ethnicity, kinship relationship with the spouse, university education status, university education related to health care, occupation status, occupation in the health care system, monthly family income, and the

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status of basic and supplementary insurance coverage.

• Health Characteristics and History of SMA or Other Genetic and Nongenetic Diseases

This included the health status of children and pregnancy experience, history of SMA, and other genetic and nongenetic disorders such as diabetes and blood pressure in the individual and the family, as well as the history of SMA screening and other diseases, which were carried out by the respondent.

• Knowledge of SMA

The questionnaire included a question about the respondent's familiarity with SMA.

All factors, except for age and monthly income, were inquired through closed-ended questions with 2 options of "yes" or "no" Respondents were given 5 options for the monthly income factor: <$90, $97 to $194, $194 to $258, $258 to $316, and >$316, and the respondents were required to select an option that best described their family's monthly income.

SMA Screening Test Scenario and Subjective Valuation of WTP

Subjective valuation of the SMA screening test by WTP was determined using the CVM. Using a survey design, we presented a hypothetical scenario consisting of general and specific information about SMA, and the carrier screening test for this disease was given to the study population, and the participants assuming out-of-pocket payment were asked for their maximum WTP for the screening test. The designed SMA screening scenario focused on the following points:

• General and specialized information about SMA and its different types.

• Guidance on implementing a carrier screening program to enable early diagnosis of SMA and an overview of its strengths and weaknesses.

• Information about drug therapy options for the treatment of SMA, including their limited availability and high cost, as well as their potential to reduce the risk of death and improve the condition of patients.

After reading the scenario, female respondents were asked a closed-ended question regarding their WTP for the SMA carrier screening test. They had to choose one of the options: "agree," "disagree," or "do not know." If they selected "agree," they had to select an exact price out of 14 suggested values, ranging from <$15.8 to >$4424 (Table 1).

The study used the minimum wage law of Iran's Ministry of Cooperation, Labor, and Social Welfare as the income distribution index. If respondents were unwilling to pay, the reasons for their decision were presented as closed questions with 5 options to choose from: "I do not believe in screening for diseases to detect them faster," "I do not believe in SMA screening," "Can diagnose the disease faster," "I cannot afford it," and "Other reasons." They had to select 1 or more reasons for their unwillingness to pay.

Table 1. Amounts WTP for SMA screening test

Less than 15.8 $*
15.8 $
31.6 $
63.2 $
158 $
316 $
632 $
1264 $
1896 $
2528 $
3160 $
3792 $
4424 $
More than 4424 $*

* Mentioning the exact amount

In the first survey, 74 eligible women were asked an open-ended question about how much they would be willing to pay for an SMA carrier screening test (39). The second survey included the actual prices for the SMA screening test through the suggested monetary amounts from the first survey as well as after calculating the mean, standard deviation, median, interquartile range (IQR), quartile 1 (Q1), and quartile 3 (Q3) extracted. Q1 equals the value below which 25% of the distribution falls. Q3 equals the value below which 75% of the distribution falls. The difference between these 2 values is equal to the IQR (40).

An expert panel also validated the questionnaire. Finally, a questionnaire was prepared for the respondents in the second survey to collect the actual WTP prices.

In this study, we converted the monetary amounts of WTP according to the International Monetary Fund data for purchasing power parity values in USD in the study year. Accordingly, each US dollar is equivalent to 31,645 Iranian rials, the information of which is extracted from the "CCEMG–EPPI Centre Cost Converter" (41).

Data Collection

During the COVID-19 pandemic, the questionnaires in the first survey were designed as an internet panel. They were distributed to respondents through email and social networks like Telegram and WhatsApp. However, with the subsidence of the epidemic, in the second stage of the survey, questionnaires were distributed and gathered in person among the study population.

Data Analysis

Descriptive analysis was performed on demographic, economic, health, and other variables to illustrate the characteristics of the study population. The median, mean, standard deviation, first and third quartiles, IQR, and 95% CIs were calculated. The logistic regression model evaluated the relationship between the WTP for the SMA carrier screening test and all independent variables. WTP was categorized as 1 and 0 for participants who agreed and disagreed with WTP. All variables were included in the principal logistic regression model, and variables with $P < 0.05$ were entered in the final logistic model and reported. Adjusted and unadjusted odds ratio (AOR, UOR), log-likelihood, and the chi2 test were reported. In this study,

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STATA 17 software was used for data analysis. This section used the maximum likelihood method to estimate the logistic model.

If the equation consists of only 1 independent variable, the UOR considers the influence of that 1 independent variable on the dependent variable. However, when more variables are included in the analysis, the AOR takes into account the effect of a predictor variable on the dependent variable, taking into account all additional variables included in the analysis (42).

Results

The studied population consisted of 578 women. After the study population completed the questionnaires, some needed to complete information about their WTP for SMA screening, and in others, conflicting information was entered. Finally, 569 questionnaires were included in the study and analyzed.

Descriptive Characteristics of the Respondents

Table 2 shows the descriptive characteristics of the respondents. The mean age of the individuals who completed the survey was 32.43 years (SD, 7.968), ranging from 15 to 44 years. Also, 26.99% of the respondents had a familiar relationship with their spouse. Of the respondents who participated, 33.22% had completed a university education, with 31.77% having a university education related to health care. Also, 31.66% of the respondents were employed, with 34.97% working in the healthcare system.

Additionally, 34.97% of the working women were part of the health care system. More than 10.9% of women were familiar with SMA, and over 76% had experienced

Table 2. The descriptive characteristics of the respondents

Variable		Frequency (%)
Participants		578 (100)
Mean age		32.43 ± 7.968 (100)
Kinship relationship	Yes	156 (26.99)
	No	421 (73.01)
	Did not answer	1 (0.00)
University education	Yes	192 (33.22)
	No	386 (66.78)
University education related to health care	Yes	57 (31.77)
	No	135 (68.23)
Occupation status	Yes	183 (31.66)
	No	392 (68.33)
	Did not answer	3 (0.01)
Occupation in the health care system	Yes	64 (34.97)
	No	116 (65.01)
	Did not answer	3 (0.02)
Monthly income ($)	<97	65 (11.25)
	97-194	135 (23.36)
	194-258	168 (29.07)
	258-316	98 (16.96)
	316 <	107 (18.51)
	Did not answer	5 (0.85)
Familiarity with SMA	Yes	63 (10.90)
	No	513 (88.75)
	Did not answer	2 (0.35)
Basic insurance coverage	Yes	413 (71.45)
	No	160 (27.68)
	Did not answer	5 (0.87)
Supplementary insurance coverage	Yes	144 (24.91)
	No	429 (74.22)
	Did not answer	5 (0.87)
Children's health status	Yes	423 (97.08)
	No	22 (0.05)
	Did not answer	3 (0.00)
Pregnancy experience	Yes	443 (76.64)
	No	131 (22.66)
	Did not answer	4 (0.70)
History of SMA among family and relatives	Yes	12 (2.07)
	No	563 (97.40)
	Did not answer	3 (0.53)
The personal history of SMA screening test	Yes	10 (1.73)
	No	560 (96.89)
	Did not answer	8 (1.38)
History of other genetic diseases among family and relatives	Yes	73 (12.62)
	No	502 (86.85)
	Did not answer	3 (0.53)
The personal history of screening tests for other diseases	Yes	143 (24.74)
	No	432 (74.74)
	Did not answer	3 (0.01)

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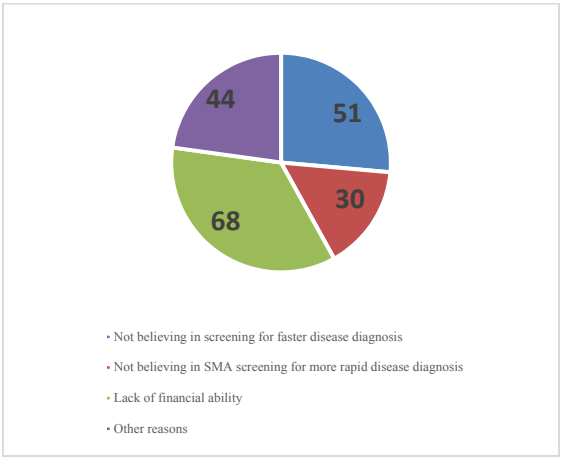

Figure 1. Reasons for unwillingness to pay for SMA screening and the frequency of respondents choosing each reason

pregnancy. Only 2.07% of women had a history of SMA in their family or relatives, and <2% had undergone a personal SMA screening test.

WTP for SMA Screening and Associated Factors

Of the female respondents, 64.85% had chosen "yes"; in other words, they expressed their WTP screening fees directly out of pocket. Figure 1 shows reasons for unwillingness to pay for SMA screening and the frequency of respondents choosing each cause. The respondents stated that the lack of financial ability to pay for SMA screening was the most crucial reason for their unwillingness to pay. Table 3 shows the descriptive statistics of the WTP for the SMA carrier screening test chosen by the respondents. The WTP for the SMA screening test was $525.68.

In the 95% CI, the 2 variables of university education (AOR, 0.526; P = 0.009) and pregnancy experience (AOR, 1.845; P = 0.021) are associated with the WTP of screening SMA. Also, considering the 90% CI, age (AOR, 0.975; P = 0.071) and occupation (AOR, 0.64; P = 0.07) were added to the above 2 variables. The overall significance of the regression model was confirmed (P < 0.05). Other variables—including kinship relationship, university education related to health care, occupation in the health care system, income, familiarity with SMA, basic and supplementary insurance coverage, children's health status, history of SMA and other genetic diseases among family and relatives, and the personal history of SMA and other diseases screening test—were excluded from the final logistic regression method.

Table 4 shows the logistic regression results of significant independent variables. The marginal effects after logistic regression also confirmed these results.

In women's university education variable, AOR was higher than UOR, indicating that the chance of influencing these variables and other independent variables on WTP for screening for this disease was higher than its chance alone on the dependent variable. The variable of pregnancy experience is the same, and AOR was higher than UOR.

The analysis showed that the WTP for noncollege-educated women is higher than that of college-educated women. Also, women with pregnancy experience had more WTP than women without this experience.

Table 3. The descriptive statistics of the WTP for the SMA carrier screening test

WTP for SMA screening	Amount
Median	$64.51
Mean	$525.68
Standard deviation	$1204.88
1st quartile	$31.60
3rd quartile	$316.00
Interquartile range (IQR)	$284.4
CI 95%	396.74-654.78

Table 4. The results of logistic regression by Odds Ratio and coefficients output

variable	AOR (95% CI)	UOR	P-value
Age	0.975 (0.95-1.00)	0.991	0.071
University education	0.526 (0.33-0.85)	0.415	0.009
Occupation	0.640 (0.39-1.04)	0.432	0.070
Pregnancy experience	1.845 (1.10-3.11)	1.651	0.021
Costant	0.897 (0.39-2.06)	0.460	0.797

LR chi2(4): 30.24, Prob > chi2: 0.000, Pseudo R2: 0.043, Log likelihood: -339.21

Discussion

This study is the first to examine the subjective valuation of WTP for the SMA screening test and its related factors in Iran. The study analyzed the WTP benefits of SMA carrier screening and found that about 35% of the women who participated were not willing to pay for the SMA screening test. Logistic regression results showed that university education and pregnancy experience were the main factors associated with WTP for screening tests, while women's age and occupation had a less significant effect. In a similar study conducted by Lin et al (29), WTP for the SMA screening test was discussed, but their study focused on newborn screening, whereas this study focused on SMA carrier screening.

Considering that more than half of the studied population expressed satisfaction with paying for SMA screening tests, it can be concluded that Iranian women are generally willing to perform them. Their opinion is based on the accepted theory that prevention is preferable to treating any kind of disease and abnormality, including SMA as a congenital genetic abnormality. Of course, we should remember the influential role of the government, private, and nongovernmental organizations in financing and supporting patients with SMA and their families.

According to our findings, there are very few studies on WTP for screening and the treatment of this disease. However, there are several studies on the WTP for various intervention screening and treatment in other congenital and rare genetic disorders, such as cystic fibrosis and celiac disease.

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In the study by Lin et al (29), most respondents (78%-79%) supported screening their infants for SMA. Also, in focused research on WTP for celiac disease screening, Norstrom et al concluded that 63% of participants had WTP for a celiac disease screening test (36). In a study on the WTP for the breast cancer sensitivity test as a test to detect breast cancer as a nonrare, noncongenital disease, Blouin-Bougie et al concluded that 57% of the participants were willing to have this test had to pay (43). While in this study, more than 64% stated they would be WTP for SMA screening.

The mean and median WTP of respondents for SMA screening test in this study were $65 and $526, respectively. In a study by Lin et al on parents' WTP for newborn screening tests for early detection of SMA, the mean and median WTP for SMA screening tests without available treatment were $142 and $253, respectively (29). The median was higher than the median of the present study, and the mean WTP was lower than the mean of the present study.

According to research by Donaldson et al, the main motivation behind the desire to pay people for cystic fibrosis carrier gene testing was the need for certainty about the test results and to avoid the risk of having a child with cystic fibrosis (32). However, in the present study, the unwillingness of the respondents to pay was primarily due to financial and economic issues. They believed the government or other responsible organizations should intervene and financially support the patients and their families.

This study investigated and analyzed factors related to WTP for the SMA screening test. University education and pregnancy experience were significantly associated with WTP for the SMA screening test. In general, a college-educated person makes more rational decisions about various issues, including paying out-of-pocket for SMA screening tests, than a noncollege-educated person. Therefore, it can be concluded that those who have attended college spend less on SMA screening than those who have not because the former group does not hold government, private, or nongovernmental organizations and institutions accountable for this expense.

Their ability to cover the expenses is well established. Like this study, Norström et al found that education played a significant role in WTP for the screening test for celiac disease in a study on the topic that was carried out in 5 regions of Sweden. The study also found that income was a significant factor in WTP for the screening test (36).

It was expected that some independent variables, such as income or insurance, would significantly affect the WPT. Still, the nonsignificance of such variables on the desire to pay for spinal muscular atrophy screening can be attributed to the influence of noneconomic factors, such as social and cultural factors attributed to the WTP.

Also, this study concluded that women with a history of pregnancy are more willing to pay compared to women without a history of pregnancy. This issue can be justified by the fact that pregnant women suffer from physical, financial, and psychological problems during pregnancy. During this period, they understand the psychological pressure related to the health status of the fetus and the uncertainty of the birth of a healthy baby. Therefore, they are willing to reduce this psychological pressure by spending more money to receive preventive interventions and screening tests to ensure the increase of fetal health and the birth of a healthy baby.

Limitations

This study had some limitations. Since some participants refused to answer some of the questions in the questionnaire, the questionnaire was prepared considering possible bias. Another limitation of this study was the social conditions caused by the COVID-19 pandemic and the lack of access to experts and the study population in the pretest phase, resolved by following health guidelines and developing electronic tools. Also, there is a risk of initial bias in different questionnaire designs using the CVM method, meaning that the base price offered affects consent to pay. Therefore, several prices were presented to the respondents.

Conclusion

Considering the willingness of most people in this study to pay for SMA screening tests, it can be concluded that Iranian women are willing to perform SMA screening tests. On the other hand, these people stated that the most important reason for their unwillingness to pay is their lack of financial ability or their belief that they are not responsible for the screening cost for this abnormality. Thus, the community members expect the decision-makers, policymakers, and health system trustees, along with the financial and nonfinancial support of the patients and their families, to be aware of the preferences of the people and the monetary valuation of the benefits resulting from prevention interventions, and make decisions based on scientific evidence regarding its targeted performance at diverse levels of the health care system.

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Authors' Contributions

Conceptualization: Khosravi M, Moradi N. Data management: Khosravi M, Azadi N, Nassiri S. Formal analysis: Khosravi M, Azadi N. Funding acquisition: Rezapour A. Methodology: Moradi N. Project management: Rezapour A. Visualization: Moradi N, Nassiri S. Writing the original draft: Rezapour A, Moradi N. Writing, reviewing, and editing: Khosravi M, Azadi N, Nassiri S.

List of Abbreviations

SMA: Spinal Muscular Atrophy; QALYs: quality-adjusted life years; CVM: congenital valuation method;

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IQR: interquartile range; CI: confidence interval; AOR: Adjusted odds ratio; UOR: Unadjusted odds ratio; MLE: maximum likelihood method; BCST: breast cancer sensitivity test.

Conflict of Interests

The authors declare that they have no competing interests.

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