Systematics Review Article
http://mjiri.iums.ac.ir
Medical Journal of the Islamic Republic of Iran (MJIRI)

Med J Islam Repub Iran. 2022 (7 Dec);36.149. https://doi.org/10.47176/mjiri.36.149

Willingness to Pay for Down Syndrome Screening: A systematics Review

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Received: 15 Sep 2021 Published: 7 Dec 2022

Abstract

Background: Financial ability to pay has a unique role in the accessibility of health care services, which indicates the necessity of raising enough funds by governments. However, how much households are willing to pay (WTP) for receiving a particular service? And what factors influence their WTP? The current systematic review aimed to, firstly, review studies on the WTP for Down syndrome (DS) screening, and, secondly, to identify factors that affect WTP for DS screening.

Methods: We systematically searched the Scopus, PubMed, Web of Sciences (ISI), and Embase databases to identify relevant studies from their inception to June 2020; the search strategy was updated on December 2021. Initially, 157 articles were identified, and 5 were found eligible for full-text review. In event of any disagreement, a third reviewer was used. Extracted WTPs were converted to US dollars in 2018 using exchange rate parity and the present value formula to make a comparison. The quality assessment of the selected studies was done using the "Lancsar and Louvier" and Smith checklist; also, vote counting was used to assess the influence of factors.

Results: Five eligible studies, published from 2005 to 2020, were fully reviewed. All final studies were scored as good quality. The extracted WTPs varied from $169 to $1118 in UK and Canada, respectively. Income and information/knowledge about screening tests were the most frequently investigated factors. Education level, detection rate, women's age, cost, and family history were significantly associated with higher levels of WTP for DS screening.

Conclusion: This study demonstrated a significant gap in WTP for DS screening in various countries. Women are WTP higher costs for tests with higher screenings. Also, a unique role was identified for income, occupation, information, and family history of DS in WTP for DS screening. In addition, a positive association was found for the variable of age.

Keywords: Willingness to Pay, Contingent Valuation, Down Syndrome Screening, Early Detection, Systematic Review

Conflicts of Interest: *None declared*
Funding: *This study was part of a PhD thesis that supported by Iran University of Medical Sciences (Grant No. IUMS/SHMIS_98-4-37-17365).*

Cite this article as: Nikjoo SH, Rezapour A, Moradi N, Nassiri S, Kabir A. Willingness to Pay for Down Syndrome Screening: A systematics Review. *Med J Islam Repub Iran.* 2022 (7 Dec);36:149. https://doi.org/10.47176/mjiri.36.149

Introduction

Birth defects are abnormalities developed during pregnancy, with potentially serious damage to the health of children during their lifetime (1). In many countries, one of the most prevalent birth defects is Down syndrome (DS), which is defined as trisomy of chromosome 21 in 95% of cases and translocation or mosaic in 5% of cases

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↑What is "already known" in this topic:
Down syndrome (DS) is the most common chromosomal condition. The most common cause of DS is when an extra copy of chromosome 21 randomly appears in either the egg or the sperm. Routine prenatal tests are provided to expectant mothers at various stages of the pregnancy and for various reasons, and these tests can assist determine the likelihood of DS.

→What this article adds:
This study identified factors affecting the WTP for DS screening and showed a significant gap in WTP for DS screening in different countries. A unique role was also identified for income, occupation, information, and family history of DS on WTP for DS screening.

[DOI: 10.47176/mjiri.36.149]

(2-4). According to the currently available evidence, DS is the main cause of intellectual disability, as nearly all cases experience impaired cognitive functions (3, 5). Apart from decreased quality of life, through impaired cognitive function, DS causes an increased risk of developing comorbidities and death (6-9), which in turn translates into considerable social consequences, mainly in the form of dependence (10). Moreover, about 33% of all moderate and severe mental handicaps in school-aged children are caused by DS. While no precise estimation is available about the prevalence of DS worldwide, there is a consensus that the prevalence of DS is on the rise, mainly because of increased life expectancy (11). Due to the lack of a national birth defects registry in Iran, the prevalence of birth defects, including DS, cannot be determined with any degree of accuracy. However, a systematic and meta-analysis study conducted by Zahed et al estimated that the prevalence of DS is 0.9 per 1000 in Iran (12).

Although there is evidence indicating the contribution of environmental factors and genetics in the development of DS, no exact cause is reported for this condition (13). Hence, few options are available to prevent or treat DS, including dietary interventions (eg, folate or iodine) and preconception health care (eg, predictive testing) (14-16). For such conditions, predictive tests can provide valuable information for parents-to-be and health care professionals to make informed decisions, which has resulted in the fast growth of tools or methods that can provide such information, including screening (14). Antenatal screening for DS is available in several countries, including Iran, to provide information regarding the risk of having DS (7). In cases where the result of the screening test is positive, a decision should be made to perform further tests and terminate the pregnancy if the results are positive (17, 18). Therefore, as screening/testing is the optimal process, one of the first decisions is to undergo screening. For those who want to utilize these services, there are a number of tests for screening pregnant women that are accessible, each with a different level of accuracy and a varied amount of waiting time for test results. The current systematic review aimed to examine previous research on WTP for DS screening and to identify variables that influence WTP for DS screening. It is important to note that our search of the literature found few studies reporting the WTP for DS.

Methods

The present study was conducted based on the PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-Analyzers) guidelines, including identification, screening, eligibility assessment, and inclusion for systematic reviews and meta-analyses.

Identification

We systematically searched the Scopus, PubMed, Web of Sciences (ISI), and Embase databases to identify relevant studies from their inception to June 2020. The search strategy was updated on December 2021. The keywords were selected using the US National Library of Medicine's Medical Subject Headings (Mesh) as follows: willingness to pay, contingent valuation, contingent evaluation, discrete choice experiment, choice experiment, conjoint analysis, stated preference, dichotomous choice, iterative bidding, payment card, revealed preference, open-ended, choice modeling, pair comparison, contingent rating, contingent ranking, Down Syndrome, trisomy, and Mitotic

Table 1. Search Strategies Administered for Various Databases

Database	Search Type	Search Strategy	Number
PubMed	Advanced Search	Keywords: (("willingness to pay"[tiab] OR "willingness-to-pay"[tiab] OR WTP[tiab] OR "contingent valuation*"[tiab] OR "contingent evaluation"[tiab] OR "contingent-valuation"[tiab] OR CVM[tiab] OR "discrete choice experiment"[tiab] OR DCE[tiab] OR "choice experiment"[tiab] OR "conjoint analysis"[tiab] OR "stated preference*"[tiab] OR "dichotomous choice"[tiab] OR "iterative bidding"[tiab] OR "payment card"[tiab] OR "Revealed preference"[tiab] OR "Open-ended"[tiab] OR "Choice modeling"[tiab] OR "Pair comparison"[tiab] OR "Contingent rating"[tiab] OR "Contingent ranking"[tiab]) AND ("Down Syndrome"[Mesh] OR "down* syndrome"[tiab] OR (Trisomy 21[tiab] AND Mitotic Nondisjunction[tiab]))	32
Scopus	Advanced Search	(TITLE-ABS-KEY ("Down Syndrome" OR "down* syndrome" OR "Trisomy 21" OR "Mitotic Nondisjunction") AND TITLE-ABS-KEY ("willingness to pay" OR "willingness-to-pay" OR WTP OR "contingent valuation*" OR "contingent evaluation" OR "contingent-valuation" OR CVM OR "discrete choice experiment" OR DCE OR "choice experiment" OR "conjoint analysis" OR "stated preference*" OR "dichotomous choice" OR "iterative bidding" OR "payment card" OR "Revealed preference" OR "Open-ended" OR "Choice modeling" OR "Pair comparison" OR "Contingent rating" OR "Contingent ranking"))	61
Web of Science	Advanced Search	(TS= ("willingness to pay" OR "willingness-to-pay" OR WTP OR "contingent valuation*" OR "contingent evaluation" OR "contingent-valuation" OR CVM OR "discrete choice experiment" OR DCE OR "choice experiment" OR "conjoint analysis" OR "stated preference*" OR "dichotomous choice" OR "iterative bidding" OR "payment card" OR "Revealed preference" OR "Open-ended" OR "Choice modeling" OR "Pair comparison" OR "Contingent rating" OR "Contingent ranking")	60
EMBASE	Advanced Search	Keywords: 'willingness to pay' OR 'down syndrome' AND screening OR "Contingent rating" OR "dichotomous choice" OR "contingent valuation*" OR "conjoint analysis" OR "WTP" OR "discrete choice experiment"	4

[DOI: 10.47176/mjiri.36.149]

Nondisjunction. Moreover, the World Health Organization database was also searched to prevent losses of related studies or important information. Our search strategy was developed using the descriptors of MeSH and Entree (Embase subject heading), adapting to the Embase database.

The search strategy used for various databases is provided in Table 1.

Inclusion Criteria

This systematic review included all studies that reported female and male's willingness to pay (WTP) for hypothetical or specific DS screening tests using various methodologies published until December 2021. The only defined restriction was including publications that are in English.

Exclusion Criteria

Studies published in languages other than English, those whose complete text was unavailable, and those with insufficient quality ratings were excluded.

Study Selection

After searching the aforementioned databases, all the identified studies were transferred to Endnote X8, and duplicates were removed (n = 7). Then, titles and abstracts were screened against the inclusion and exclusion criteria. Afterward, the full texts of all eligible articles were reviewed by 2 reviewers, and key specifications designed were extracted. Any disagreement between the researchers was investigated using Cohen's kappa and then by a third reviewer. In addition, to prevent bias, all disagreements were discussed by the research team. Articles that did not meet the inclusion criteria were excluded and the remaining entered the qualitative evaluation phase. The search strategy was repeated by a second independent reviewer to ensure the adequacy of the search process.

Adjusting extracted values from economic evaluation studies is of crucial importance to make them comparable. In this regard, using the net present value calculation and exchange rate parity, the extracted WTPs from final studies were converted to US dollar value in 2018. In cases where the year was not reported, the year of publication was used for currency conversion.

Quality Assessment

Quality assessment was performed using the Lancsar and Louviere checklist for discrete choice studies and the Smith checklist for contingent valuation studies (19, 20). The Lancsar and Louviere checklist is a tool to assess the validity of discrete choice studies. This tool has 13 criteria in 4 categories; choice task design, experimental design, conduct, and analysis. Items are identified by the colors green, red, and yellow. A recommendation for the application of contingent valuation studies is included in the Smith checklist for health professionals. This checklist has 34 items that are categorized into 4 groups: CV development and context; CV scenario development, CV reporting and results, and CV validity and reliability. A "Yes" sign, denoting a score of 1, would be used to indicate items that have been reported by a study, and a "No" sign, denoting a score of 0. Also, the effects of different factors on DS screening WTP were identified using vote counting (21). Using this tool, the factors extracted from the selected studies with signs and significance, including significant positive effect, significant negative effect, nonsignificant positive effect, and nonsignificant negative effect, are voted.

Data Extraction

Data were extracted using a researcher-developed checklist that included information on the authors' name, year of publication, title, place, type of study, year of realization, data collection method, study population, evaluation method, and effective variables.

Results

Initially, 157 articles were identified. Our manual review did not reveal any new studies. In addition, there was no disagreement in the search process. Seven duplicates were identified. Therefore, 150 articles were screened based on the inclusion and exclusion criteria, which led to the exclusion of 109 articles. Then, of the remaining 41 articles, 36 articles were excluded after full-text review due to irrelevance or unavailability of the full text. Finally, the quality of 5 articles was assessed (Fig. 1). The validity of 4 articles was checked using the Lancsar and Louviere checklist; 1 study had 100%, 2 studies had more than 90%, and 1 study had about 80% validity. The Smith checklist was used to rate one article, and it yielded a score of 71% (Tables 2 and 3).

Study Characteristics

Eligible articles were published from 2005 (22) to 2020 (23). Two studies were conducted in the UK (22, 24), 1 in China, 1 in Canada, and 1 in the Netherlands. Also, the highest and lowest numbers of participants were 147 (26) and 50 (22), respectively, in 3 studies on pregnant women (22, 23, 25, 26) and 1 study on both sexes (24). Various factors that may influence WTP were examined in all final studies, including age, education, employment status, information, knowledge, test cost, diagnosis rate, income, waiting for test results, and family history.

Four papers were based on discrete choice analysis (DCE) (22-25) and 1 paper was based on probabilistic valuation (26). The first is a survey-based approach to extracting preferences, which in turn allows the evaluation of the value of each of the proposed options to an individual/user in cases where their provision is free or not yet introduced (27). The second method is one of the important methods of probability assessment, which is designed to show the WTP of users of a particular service or product. This method is based on asking respondents to select a value that represents their maximum WTP. Then, their WTP can be considered as values higher than the value shown (28). Respondents answered questions using face-to-face interventions (22) or self-administered questionnaires (23), reporting response rates ranging from 43% to 98% (22, 23). Through the use of a literature study,

[DOI: 10.47176/mjiri.36.149]

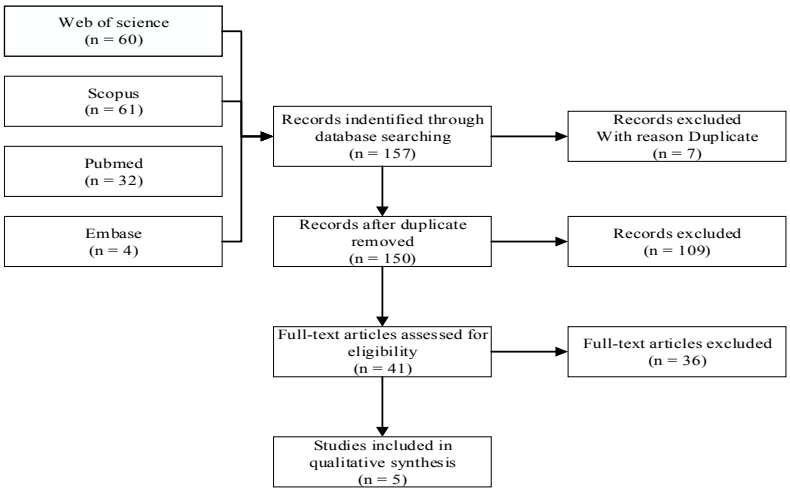

Fig. 1. Study selection process.

Table 2. Validity assessment of included studies

	Criteria	Study: Carroll et al 2013	Regier et al 2009	Ryan et al 2005	Wu et al 2020
Choice Task Design	Attributes and levels grounded in qualitative work with target population				
	No conceptual overlap between attributes				
	Uni-dimensional attributes				
	Opt-out/status quo option or justification of forced choice				
Experimental Design	Experimental design optimal or statistically efficient				
Conduct	Piloting conducted amongst target population				
	Target population(s) appropriate for research objective				
	Sampling frame representative of target population				
	Response rate sufficient to minimize response bias				
Analysis	Any pooled analysis from different subgroups appropriate				
	Econometric model appropriate for choice task design				
	Econometric model accounts for serial correlation of choices				
	Relative attribute effects compared using a common metric				

Red = criteria not met (no evidence or not enough evidence to justify the criteria in the text); Green = criteria met (the text sufficiently confirmed the criteria).
Yellow=unclear
N/A=not applicable

interviews, and a panel of expert directors, discrete studies were able to pinpoint selection traits and levels. The range of levels in the DCE trials was between 9 and 17, and the range of characteristics was between 3 and 5. All studies employed the parameters of cost and outcome waiting time (21-24). The detection rate was the next most common feature. Other characteristics used include pregnancy, number of children whose genetic status was determined by the test, level of information, method of testing, and abortion.

WTP for DS Screening

In the present study, final articles have reported a different range of values for WTP, which are not comparable because of differences in the year of calculation. Hence, all values were converted using a specific discount. Because the latest study was performed in 2020, the WTPs were converted to US dollars in 2020 and discounted at the discount rate of 3% using the net present value formula. WTP for DS screening is provided in the last column of Table 4. As reported in the Table, the highest and lowest values are reported for Canada (ie, $1118) (24) and Netherland (ie, $169) (22), respectively.

[DOI: 10.47176/mjiri.36.149]

Table 3. Smith Checklist

Checklist of what should be reported in published CV studies	Verweij et al. (2013) (26)
CV development and context	
Country where the CV survey has been conducted and health care financing details	Yes
Focus—methodological or policy	Yes
Specificity of questionnaire (part of wider survey)	Yes
Details of other measures of QoL incorporated	No
Scenario development	Yes
Welfare measure (WTP or WTA)	Yes
CV scenario description	
Intervention(s)	No
Partiality (single good or close substitutes)	No
Outcomes (health status, probability and time)	Yes
Non-outcomes (information, care, other)	No
Payment vehicle	Yes
Presentation of uncertainty/risk	Yes
Survey period	Yes
Time period for WTP	Yes
Question/elicitation format	Yes
CV reporting and results	
Method of data collection	Yes
Type of respondent	Yes
Sample size	Yes
Response rate	Yes
Type of outcomes incorporated (use, option, or externality value)	Yes
Duration of interview/length of questionnaire	Yes
WTP values (results of the studies)	Yes
Transformation of values from one context/time to another	No
Price year	Yes
Currency	Yes
Cost of intervention	No
Cost–benefit ratio	No
Time period used in analysis	Yes
CV validity and reliability	
Tests for bias—order effect, starting point, range, interviewer, strategic	Yes
Statistical analysis performed	Yes
Assessment of zero/high bids	No
Distributional issues consider	Yes
Validity tests	No
Reliability tests	No
Rate of responses	71%

Factors Affecting WTP for DS Screening

Different factors affect the WTP and understanding them is important for raising funds for screening programs. Final studies also have investigated factors affecting the WTP. After counting the votes, income (25) and cost (23, 25) were the most frequently investigated factors. Other significant factors included detection rate (25), women's age (22), information/knowledge about screening tests (23), and family history (23).

Some studies investigated a wide range of influencing factors (23, 24), while the rest examined a limited number of factors (22, 25, 26), which indicates the lack of a unique producer to identify and report factors that affect the WTP (Table 5).

Discussion

Given the increasing number of those who suffer from DS, health policymakers should pay special attention to the utilization of early detection methods to identify high-risk pregnancies. To the best of the authors' knowledge, this is the first systematic review of its kind, therefore it aimed to examine all research that looked into WTP for DS screening. Unsurprisingly, the results showed that the WTP increased with income level, especially after the threshold of the yearly income of $30,000. In addition, it was found that women are WTP higher costs for more accurate screening tests, while they did not care much about waiting time for test results (25). Working women also tended to choose more expensive screenings and had higher WTP. In addition to having a different mindset than housewives, this is due to their financial independence, which allows them to select a more expensive option. Verweij et al in 2013 (22) reported higher WTP for older women, which is also confirmed by Lo et al (27).

Moreover, studies that investigated the impact of access to extra information on WTP reported a positive effect (23, 24, 26-28). Hence, it can be argued that obtaining information about possible side effects of DS screening does not reduce WTP, which is similar to the findings reported for other diseases (29).

[DOI: 10.47176/mjiri.36.149]

Table 4. Description of Study Characteristics

Author/year	Country	Aim	Respondents	Response Rate	WTP method	Significant factors	Number of scenarios	WTP value ($)
Carrol et al. (2013) (25)	United Kingdom	Identifying the most important attributes for Down's syndrome screening	103 (63 pregnant women, 40 male partners)	NA	Discrete choice experiment	Test cost Detection rate	8	86.87 $ (70.15-103.59) for increase detection rate from 75% to 90% in preference class 1 628.57 $ (545.97-711.18) for increase detection rate from 75% to 90% in preference class 1 -9.71 $ (-140.25-120.82) for immediately preparing the test results rather than 2-4 weeks in preference class 1 193.34$ (-8346-8733.09) for immediately preparing the test results rather than 2-4 weeks in preference class 2
Verweij et al. (2013) (22)	Netherlands	To investigate the attribute among pregnant women regarding non-invasive prenatal testing for Down's Syndrome	147 women	43%	Contingent valuation	Age Income	1	Median 169$, ranging from -1000 to 150$
Ryan et al. (2005)(23)	United Kingdom	The value pregnant women place on various alternative prenatal diagnostic tests	50 pregnant women	98%	Discrete choice experiment	Level of information Number of days to wait for the results Cost	1	WTP for reducing waiting time for one day was 24.7 $
Liangzhi et al. (2020) (26)	China	Eliciting women's preference for prenatal testing in China: a discrete choice experiment	92 women	NA	Discrete choice experiment	Test procedure; detection rate; miscarriage rate; time to wait for results; and test cost.	NA	Participants were willing to pay 4610 US Dollars for non-invasive tests and up to 537 US $ to increase the detection rate by one percent
Regier et al. (2009) (24)	Canada	Valuing the benefit of diagnostic testing for genetic causes of idiopathic developmental disability: willingness to pay from families of affected children	105 families	NA	Discrete choice experiment	Time to wait for results; and a higher detection rate.	NA	The families were willing to pay up to 1118 US $ (498-1788) for the screening test.

As mentioned before, although the exact etiology of DS is not identified yet, it is believed that genetic factors, such as family history of DS, are associated with an increased risk of developing DS (30, 31). As indicated by the findings, the WTP of pregnant women tends to increase when they have a family history of DS. A similar effect was found for the variable of age. To put it another way, an illness that a woman has experienced inspires her to take precautions against future risks.

Reviewed studies that investigated the effect of education level on WTP for DS screening reported a positive association between these 2 factors (23, 24, 28). In this line, it can be argued that education facilitates a better understanding of the importance of screening for the health of the fetus, mainly because of the higher level of study.

When extrapolating the findings to other locations and circumstances, care should be taken because, with the exception of 2 studies conducted in China, almost all of the final studies were conducted in developed nations.

[DOI: 10.47176/mjiri.36.149]

Table 5. Vote Counting

Author	Carrol et al. (2013) [22]	Verweij et al. (2013) [24]	Ryan et al. (2005) [20]	Wu et al. (2020) [23]	Regier et al. (2009) [23]	Vote
Test cost	↑↑		↑	↑↑	↑↑	(3 to 0)
Detection rate	↑			↑	↑↑	(1 to 0)
Time to receive test result		↑↑				(1 to 0)
Age		↑↑				(1 to 0)
Income		↑↑	↑↑	↑↑		(3 to 0)
Education			↑↑			(1 to 0)
knowledge about DS screening						(1 to 0)
Fertility history				↑↑		(1 to 0)
Test procedure				↑↑		(1 to 0)
Miscarriage				↑↑		(1 to 0)

↑↑Positive and significant effect. ↓↓ Negative and significant effect. ↑ Positive and non-significant effect. ↓ Negative and non-significant effect

Additionally, no studies were found for low- or middle-income countries, indicating the need for similar studies in these areas that take into account their unique characteristics. Furthermore, one of the important identified research gaps was the lack of studies on the impact of insurance coverage on the WTP for DS screening; hence, extra research is needed to decide about their effect. In addition, further studies are needed to extend our knowledge regarding the impact of demographic variables other than age on WTP for various health services, particularly DS screening. Also, this review suffers from heterogeneity concerning the number of scenarios and investigated factors.

The quality assessment of all qualified studies was one of the study's key benefits. Although the final studies were of moderate to high quality, our quality assessment found flaws in how scenarios and time periods were defined when planning the investigations, which was not surprising (31).

Conclusion

Even though our research only found a few pertinent and qualified studies, we may nevertheless draw reliable conclusions about some elements. In this regard, this study discovered a significant discrepancy in WTP for DS screening across various countries. We also found that women are WTP higher costs for tests with higher screenings. Also, a unique role was identified for income, occupation, information, and family history of DS in WTP for DS screening. In addition, a positive association was found for the variable of age.

Ethical Approval

The current study is a part of a thesis proposal (code: IR.IUMS.REC.1398.1234) approved by Iran University of Medical Sciences, Tehran, Iran.

Conflict of Interests

The authors declare that they have no competing interests.

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[DOI: 10.47176/mjiri.36.149]