Parents' Willingness to Pay for Coeliac Disease Screening of Their Child

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ABSTRACT

Objective: The aim of this study is to determine Swedish parents' willingness to pay (WTP) for coeliac disease (CD) screening of their child.
Subjects and Methods: CD screening was undertaken involving 10,041 12-year-old children, with 7567 (75%) agreeing to participate. Blood samples from the children were analysed for CD serological markers. Parents received a questionnaire including a scenario describing the health-related risks of having CD and screening and diagnostic procedures. Parents were also asked whether they were willing to pay for CD screening, should this not be offered free of charge, and, if so, what their maximum WTP would be. Their WTP was compared with the average cost per child for the screening and case ascertainment procedures.
Results: The questionnaire was answered by 6524 parents, and of 6057 valid responses 63% stated that they were willing to pay something. The mean WTP was 79 EUR and the median 10 EUR. The average cost per child for the screening and case ascertainment procedures was 47 EUR, which 23% of the parents stated they were willing to pay. Parents' WTP increased with higher education and income, and with child symptoms that may indicate CD.
Conclusions: Swedish parents' WTP for school-based CD screening of their child was higher than the average cost per child; however, only a minority of the parents were willing to pay that amount.

Key Words: children, coeliac disease, cost–benefit analysis, screening, willingness to pay

(*JPGN* 2011;52: 452–459)

Coeliac disease (CD) is defined as a permanent intolerance to wheat gluten and related proteins in rye and barley. Gluten ingestion causes inflammation and atrophy of the small intestinal mucosa, resulting in malabsorption (1). With a gluten-free diet the intestinal mucosa usually recovers with signs and symptoms fading (2), but if untreated the negative health consequences are extensive (3). There are 5 to 10 undiagnosed cases for every diagnosed case in certain Western European countries (4). In Swedish screening studies, 4 of 5 adult cases (5) and 2 of 3 child cases were found to be undiagnosed (6). In the latter study, the prevalence was as high as 3%, compared with the often-cited prevalence of 1%. In the United States, patients with CD were reported to have symptoms indicative of CD for an average of 11 years before diagnosis (7).

The vast majority of CD cases is difficult to identify through clinical practice and will only be detected through mass screening efforts (8,9). Ten principles for early disease detection through screening were elaborated in 1968 by Wilson and Jungner and have since been widely used (10). Most of these principles are fulfilled for CD mass screening (9). However, insufficient knowledge of long-term consequences of living with undiagnosed CD makes such a screening ethically debatable (11,12). Also, current evidence from health economic evaluations is limited (13); thus, we do not yet know whether mass screening would be economically defendable.

The only previous comprehensive health economic evaluations of CD mass screenings are cost-effectiveness analyses conducted by the research group of Shamir in Israel (14,15). In their first study they concluded that, based on cost per life-years saved, CD mass screening could be considered over a wide range of ages, provided a relatively high CD population prevalence, and assuming a standardised mortality ratio of 1.5 or higher for untreated cases. In their second study they demonstrated an incremental cost-efficiency ratio of 48,960 USD per quality-adjusted life-year (QALY) for CD screening versus nonscreening in young adults, which suggests that mass screening would be a cost-effective strategy. However, as the authors also state, the models used were partly based on assumptions that need to be investigated further. Other health economic studies have evaluated the cost-effectiveness of different CD serological markers (16–18) and screening of risk groups such as irritable bowel syndrome (19,20) and Down syndrome (21). Many researchers have emphasised a need for additional health economic evaluations before the implementation of mass screening can be considered (9,22,23).

According to Drummond et al (24), a comprehensive economic evaluation of health services should consider 3 different consequences: changes in health, resources saved, and other values created. It is common to measure changes in health by using established methods such as QALYs, in which the resources saved are measured in monetary units. In contrast, other values created from an intervention are commonly ignored, but they are likely to be relevant in the case of screening. Important consequences of

Received May 13, 2010; accepted October 25, 2010.
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The study was performed in cooperation with the county councils of Västerbotten, Stockholm, Östergötland, Kronoberg, and Skåne and was undertaken within the Umeå Centre for Global Health Research at Umeå University. The study was funded by the Swedish Research Council; the Swedish Research Council for Environment, Agricultural Sciences and Spatial Planning; the Swedish Council for Working Life and Social Research; and the Vårdal Foundation. The study was part of the European Union–supported project PREVENTCD.
The authors report no conflicts of interest.

DOI: 10.1097/MPG.0b013e3182037efd

screening are, for example, the experienced value of the information about whether a person has a certain disease, and reduced (or increased) anxiety. These kinds of consequences (other values) have, to our knowledge, not yet been explored in the context of CD mass screening.

The contingent valuation methodology is commonly used to derive the value that people put on goods, services, and amenities (25,26), and it has previously been used in other areas of health care. From a scenario that describes the goods or services under study and consequences expected from consumption, responders are asked to state their willingness to pay (WTP). The scenario is important because poor information can lead to a hypothetical bias of the WTP (27). The most common formats of the WTP question are the open-ended format, the payment scale approach, and the dichotomous choice format (26). These formats have different strengths and weaknesses; for example, it has been shown that the open-ended format gives a lower estimate of WTP compared with the closed-ended format (frequently used dichotomous choice format) and the payment scale approach (28).

The aim of this study was to determine Swedish parents' WTP for CD screening of their child, and explore any relation to socioeconomic factors, previously diagnosed CD in the family, and the child's health and well-being.

SUBJECTS AND METHODS

Study Design

This study emanates from a school-based cross-sectional CD screening entitled ''Exploring the Iceberg of Celiacs in Sweden'' (ETICS), involving 5 regions of the country (6). During the 2005–2006 school year, all of the children in the 6th grade (12-year-olds) were invited to participate. After receiving informed consent, childrens' blood samples were obtained and analysed for CD serological markers. Those suspected to be carriers of the disease were referred for a small intestinal biopsy, which is considered the criterion standard for diagnosis (1). Before the results from the blood sample were known, parents were asked to respond to a questionnaire with prepaid postage. The study was approved by the regional ethical review board at Umeå University (Dnr 04–156M).

Subjects

In total, 10,041 children were invited to the CD screening, of whom 7567 agreed to participate. There were 7207 children without previously diagnosed CD who had their blood sample analysed for CD serological markers. Through a small intestinal biopsy, 145 of them were diagnosed as having CD. In the study, 67 previously diagnosed cases were also identified (6). Parents of 6524 (86%) children responded to the questionnaire, with 6352 eligible for the WTP analyses. Thus, 172 questionnaires were excluded, with reasons being that the child had been previously diagnosed as having CD ($n = 59$) or that the parents had been informed about the result of the CD serological markers before responding ($n = 113$).

Questionnaire

The questionnaire included a section on WTP in which a scenario was given introducing the concept of disease screening and describing CD with respect to health risks, screening and diagnostic procedures, and the treatment option (Fig. 1). Thereafter, parents were asked to estimate in monetary terms the value of being informed about their child's CD status (question 34 in Fig. 1). They were asked whether they would be willing to pay for a screening, should this not be offered free of charge, and, if so, what their maximum WTP would be in SEK (10 SEK $\approx$ 1 EUR) (question 35 in Fig. 1). If parents were unwilling to pay anything, they were asked for their motives. The questionnaire also included sections on parental education and household income, previously identified CD in the family, the child's health with respect to well-being, and some diseases and symptoms.

WTP Analyses

The proportion of parents willing to pay anything to be informed about their child's CD status was calculated from valid responses to question 34, that is, parents clearly responding that they were willing or unwilling to pay ($n = 6057$). Thereafter, parents' maximum WTP was estimated by applying both a conventional and an inclusive approach, with the latter considered our main method (Table 1).

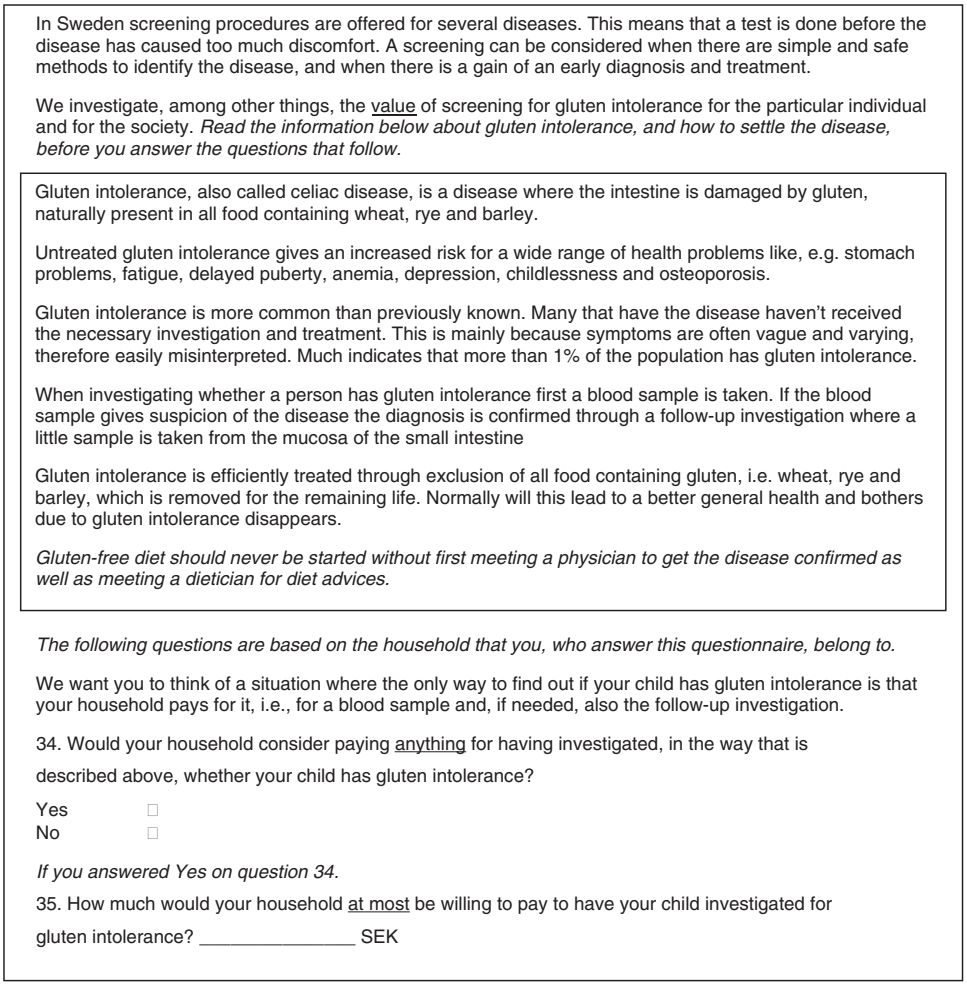
The conventional approach only included those who either explicitly stated a positive WTP (yes, question 34) and specified an amount (>0 EUR, question 35) or explicitly stated that they were not willing to pay anything (no, question 34) combined with either a stated WTP of 0 EUR or no stated amount (question 35).

In addition, the inclusive approach considered those who explicitly stated a positive WTP (yes, question 34), if they implicitly revealed their WTP, such as the fee for visiting a physician (25 EUR) or with an interval response (eg, 10–20 EUR = 15 EUR) (Table 2). It also considered all of the other responses from those who explicitly stated that they were not willing to pay (no, question 34) with a WTP of 0 EUR. Those who did not respond to question 34 (neither yes nor no) were included based on the same conditions as those who explicitly stated a positive WTP in this approach, whereas it was required for those who marked both alternatives to question 34 that they responded 0 EUR to question 35.

Estimates of Costs

The question posed to the respondents was how much they were willing to pay to have their child tested for gluten intolerance (Fig. 1). Immediately preceding the question, the following instruction was given in the questionnaire: ''We want you to think of a situation where the only way to find out if your child has gluten intolerance is that your household pays for it, i.e., for a blood sample and, if needed, also the follow-up investigation.'' The cost items for a CD mass screening that we consider associated with this description were blood sampling at school for all of the children (including costs for nurses' salary, vials), analyses of CD serological markers for all of the children, a gastroscopy with a small intestinal biopsy and pathological anatomic evaluation for each child with suspected CD, a visit to a physician for all of the children with suspected CD, and a visit to a dietician for all of the children with confirmed CD. The costs listed above, corresponding to the cost for the screening and diagnostic procedures, were estimated based on information from the health care divisions from Västerbotten and Östergötland counties councils in Sweden, and from the ETICS screening. The average cost per child for each activity was calculated based on the actual total cost for the children who were exposed to that specific activity within the ETICS screening study divided by the number of participating children in the whole study (Table 3).

A consequence of being diagnosed as having CD is a lifelong sequence of altered costs and benefits for the individuals, the households, the health care sector, and other sectors of the society. The costs after the diagnostic stage and the recommended first visit to a dietician, including both revisits to physicians and dieticians, are not included in cost estimates, nor are the households' cost for



In Sweden screening procedures are offered for several diseases. This means that a test is done before the disease has caused too much discomfort. A screening can be considered when there are simple and safe methods to identify the disease, and when there is a gain of an early diagnosis and treatment.

We investigate, among other things, the value of screening for gluten intolerance for the particular individual and for the society. *Read the information below about gluten intolerance, and how to settle the disease, before you answer the questions that follow.*

Gluten intolerance, also called celiac disease, is a disease where the intestine is damaged by gluten, naturally present in all food containing wheat, rye and barley.

Untreated gluten intolerance gives an increased risk for a wide range of health problems like, e.g. stomach problems, fatigue, delayed puberty, anemia, depression, childlessness and osteoporosis.

Gluten intolerance is more common than previously known. Many that have the disease haven't received the necessary investigation and treatment. This is mainly because symptoms are often vague and varying, therefore easily misinterpreted. Much indicates that more than 1% of the population has gluten intolerance.

When investigating whether a person has gluten intolerance first a blood sample is taken. If the blood sample gives suspicion of the disease the diagnosis is confirmed through a follow-up investigation where a little sample is taken from the mucosa of the small intestine

Gluten intolerance is efficiently treated through exclusion of all food containing gluten, i.e. wheat, rye and barley, which is removed for the remaining life. Normally will this lead to a better general health and bothers due to gluten intolerance disappears.

Gluten-free diet should never be started without first meeting a physician to get the disease confirmed as well as meeting a dietician for diet advices.

The following questions are based on the household that you, who answer this questionnaire, belong to.

We want you to think of a situation where the only way to find out if your child has gluten intolerance is that your household pays for it, i.e., for a blood sample and, if needed, also the follow-up investigation.

34. Would your household consider paying anything for having investigated, in the way that is described above, whether your child has gluten intolerance?

Yes ☐
No ☐

If you answered Yes on question 34.

35. How much would your household at most be willing to pay to have your child investigated for gluten intolerance? _____ SEK

FIGURE 1. Scenario and questions to parents of Swedish 12-year-olds within the Exploring the Iceberg of Celiacs in Sweden (ETICS) study about their willingness to pay for coeliac disease screening of their child.

TABLE 1. Parents' WTP for CD screening of their 12-year-old child and criteria for the inclusive (I) and conventional (C) approaches

	WTP								
	Yes		No*		Both yes and no		Missing		
Maximum WTP (EUR)	n	Approach	n	Approach	n	Approach	n	Approach	N
0	4	I	15	I and C	2	I	3	I	24
>0	2273	I and C	15	I	9	—	10	I	2307
Interpretable response†	388	I	0	I	2	—	1	I	391
Noninterpretable response	779	—	22	I	14	—	29	—	844
No response	365	—	2196	I and C	0	—	225	—	2786
Total	3809		2248		27		268		6352

CD = coeliac disease; WTP = willingness to pay.
* When unwilling to pay for a CD screening, the maximum WTP was set to 0 EUR, irrespective of amount reported.
† Responses that could be translated into precise amounts, for example, the fee for visiting a physician (25 EUR) or an interval response (10–20 EUR = 15 EUR).

TABLE 2. Decision rules for the inclusive WTP approach when responses were given as a text or imprecise value

Responses	Decision	Value used	n
Interval answers	Included	Midpoint of interval	86
Fee, eg, for visiting a physician	Included	Corresponding value*	129
Imprecise value	Included	Interpreted one by one	152
Yes, if suspicion of disease	Included/excluded	Included if response could be translated into a precise amount for a symptom-free child	22/75
>5000 EUR	Excluded	—	6
Not capable of answering	Excluded	—	594
Cost does not matter	Excluded	—	39
Dislike question	Excluded	—	15
Not able to make an estimate of the value	Excluded	—	28
Noninterpretable answers	Excluded	—	51
Total			1197

WTP = willingness to pay.
* Costs were based on information from the health care divisions within the Västerbotten and Östergötland counties' councils in Sweden.

the gluten-free diet and other economical benefits or losses, for example, the possibility of fewer sick days.

Definition of Explanatory Variables

Parental education was dichotomised into low and high, in which the latter implied that at least 1 parent had >12 years' schooling. For household income a truncation was done at 16,000 EUR per month. CD in the family was defined as mother, father, and/or a sibling having CD. Child well-being was defined as high when it was reported to be excellent or very good, and low when it was good, fairly good, or poor. The child was defined as having other disease if his or her parent reported at least 1 of the following diseases: lactose intolerance, cow's-milk protein allergy, any other food intolerance, diabetes, anaemia, rheumatic disease, thyroid disease, inflammatory bowel disease, vitiligo, alopecia areata, or dermatitis herpetiformis. Symptoms indicative of CD was defined as the parents' responding that their child often or always had at least 1 of the following symptoms: fatigue, abdominal pain, abdominal discomfort, flatulence, hard stools, or soft stools.

Statistical Analysis

Parents' maximum WTP, in terms of the mean and median value, was compared with the average cost per child for a CD screening. Logistic regression was used to identify explanatory factors for parents' willing to pay anything for a CD screening with results presented as odds ratios with a 95% confidence interval.

TABLE 3. Costs for a CD screening of 12-year-olds in Sweden

Cost item	Cost,* EUR	n	Average cost per child, EUR
Blood sampling at school			7.2
Nurses' salaries	5.4	7500	
Material	1.8	7500	
Analyses of CD serological markers			18
atTG†-IgA and total serum IgA	18	7207	
EMA‡-IgA	11	222	
atTG-IgG	20	170	
EMA-IgG	29	5	
Gastroscopy with a small intestinal biopsy and pathological anatomic evaluation			12
Gastroscopy	380	184	
Pathological anatomic evaluation	100	184	
A visit to physician	210	192	5.4
A visit to dietician§	210	145	4.1
Total	348803		47

IgA = immunoglobulin A; CD = coeliac disease; EMA = anti-endomysial antibodies.
* Costs for different activities were based on information from the health care divisions within the Västerbotten and Östergötland counties' councils in Sweden and from the ETICS screening study. Average cost per child was calculated from the total actual cost for each activity divided by the 7500 children involved, also including those (n = 293) for whom the blood sampling failed.
† Anti-human tissue transglutaminase.
‡ Endomysial antibodies.
§ The fee for a physician's visit was used.

TABLE 4. Frequency of responses to WTP for a CD screening of 12-year-olds in Sweden question, dividing responses into intervals for the inclusive and conventional approaches

	Conventional approach		Inclusive approach	
Maximum WTP, EUR	n	%	n	%
0–2.5	2215	49	2262	46
2.6–7.5	28	0.6	32	0.6
7.6–12.5	336	7	348	7
12.6–25	575	13	727	15
26–35	359	8	380	8
36–45	29	0.6	44	0.9
46–60	471	11	482	10
61–80	12	0.3	29	0.6
81–150	330	7	341	7
>150	129	3	284	6

CD = coeliac disease; WTP = willingness to pay.

Interval regression, using data from the inclusive approach, was used to identify explanatory factors for the WTP (29). The reason for the latter choice of method was that even if our question was open-ended the responses were clustered, as if there would have been response alternatives with intervals. Based on the peak values the following intervals in EUR were chosen for the analyses: 0 to 2.5, 2.6 to 7.5, 7.6 to 15, 16 to 25, 26 to 35, 36 to 45, 46 to 60, 61 to 80, 81 to 150, and >150. The frequencies of responses for each of the intervals for the stated WTP are given in Table 4 for both the inclusive and conventional approaches. Factors included in the logistic and interval regressions were parental education, household income, CD in the family, the child's health with respect to well-being, and some diseases and symptoms. In interval regression the coefficient for dichotomous variables can be interpreted as in an ordinary linear regression model, that is, as the added value for a positive outcome of a variable. Statistical significance was defined as $P < 0.05$. Microsoft Access was used for data handling and Stata 10 (StataCorp LP, College Station, TX) for statistical analysis.

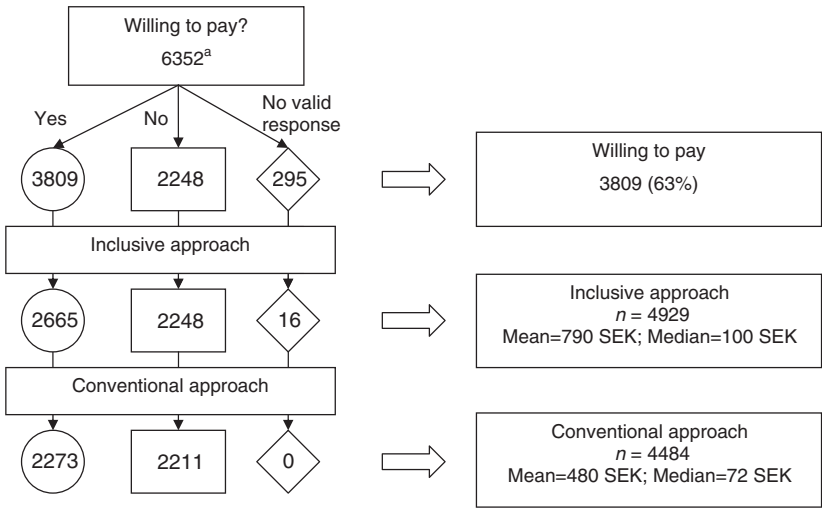
RESULTS

Parents' WTP

When parents were asked whether they were willing to pay for a CD screening of their child, after having been provided with information about the concept of screening and the disease itself, 3809 (63%) of 6057 valid responses were positive (Fig. 2). Responses from 295 parents were excluded due to either no response at all or responding both yes and no.

Motives Not to Pay Anything

Among the 2248 (37%) parents not willing to pay anything for a CD screening of their child, the most common motive was that they did not believe that their child had CD (66%) (Table 5). However, many parents indicated a positive attitude towards getting information about the child possibly having CD, despite being



[a] Parental questionnaires eligible for the WTP analyses.

FIGURE 2. Summary of parents' willingness to pay (WTP) for a CD screening of their 12-year-olds within the Exploring the Iceberg of Celiacs in Sweden (ETICS) study based on the scenario and questions in Fig. 1.

TABLE 5. Motives for parents' unwillingness to pay for CD screening of their 12-year-old child

Motives given*	n†	%
I do not believe our child has CD	1490	66
It is wrong that we should pay	478	21
We will visit the physician if our child has health problems	1226	55
We lack enough information	59	3
Our household cannot afford to pay	139	6
Other reasons	245	11

CD = coeliac disease.
* Parents were asked to choose ≥1 of the listed alternatives.
† In total, 2248 parents were not willing to pay anything (and all but 53 gave a reason), with 1101 giving 2 or more reasons.

unwilling to pay out of pocket. More than half stated that they would visit the physician if CD was suspected (55%), and as many as 21% stated that it is wrong that they should have to pay this by themselves.

WTP

The mean maximum WTP with the inclusive approach was 79 EUR and for the conventional approach 48 EUR, whereas the median WTP for the inclusive approach was 10 EUR and for the conventional approach 7.2 EUR (Fig. 2). There were 4929 responses considered in the inclusive approach and 4484 responses in the conventional approach (Table 1). A larger proportion of responses were excluded among parents willing to pay (conventional approach 1536 and inclusive approach 1144) compared with parents unwilling to pay (conventional approach 37 and inclusive approach 0).

Costs for the Screening Versus WTP

The average cost per child for our CD screening was 47 EUR. The estimated cost for each considered item (eg, blood sampling, analyses of CD markers) and number of children fulfilling criteria for each of these are given in Table 3. Parents' mean WTP for a CD screening of their child was higher than the cost, irrespective of using the inclusive (79 EUR) or conventional (48 EUR) approaches. However, still only 23% of the parents were willing to pay at least the average cost per child according to the inclusive approach and 21% according to the conventional approach.

Explanatory Factors

There was a statistically significant relation between whether parents were willing to pay anything for a CD screening of their child and a higher education and income, a previous CD case in the

TABLE 6. Explanatory factors for parents' WTP for CD screening of their 12-year-old child

	Responders		Willing to pay		LR*, n = 3948		IR†, n = 3324	
Characteristics	n‡	%	n	%	OR	95% CI	Coeff	*P*
Parental education§								
Low	2530	45	1448	60	1		4.46	0.01
High	3075	55	1958	66	1.19	1.03–1.39		
Household income‖	4536				1.003	1.0002–1.0066	0.224	<0.001
CD in the family¶								
No	6203	98	3708	63	1		4.08	0.46
Yes	149	2	101	74	2.01	1.16–3.48		
Child well-being#								
Low	1081	17	668	66	1		−3.95	0.09
High	5194	83	3092	62	0.82	0.66–1.01		
Child with other disease**								
No	5233	82	3055	61	1		3.02	0.16
Yes	1119	18	754	71	1.36	1.11–1.67		
Child's symptoms indicative of CD††								
No	4810	76	2846	62	1		4.14	0.04
Yes	1542	24	963	67	1.26	1.05–1.52		
Constant					21.5			

CD = coeliac disease; CI = confidence interval; IR = interval regression; LR = logistic regression; OR = odds ratio; WTP = willingness to pay.
* LR to identify explanatory factors for whether parents were willing to pay or not, with analysis limited to those responding to all questions. Results are given with ORs and 95% CI.
† IR, using an inclusive WTP approach, to determine factors that affect the stated WTP in EUR, with analysis limited to those responding to all questions.
‡ Number of responses among all questionnaires eligible for WTP analysis.
§ Parental education; cutoff between low and high was set to at least 1 parent having >12 years' schooling.
‖ Household income in thousands, truncated at an income of 16,000 EUR/month.
¶ CD in family: mother, father, and/or a sibling with CD.
# Child well-being: high implies excellent or very good, and low implies good, fairly good, or poor.
** Child with other disease: lactose intolerance, cow's-milk protein allergy, any other food intolerance, diabetes, anaemia, rheumatic disease, thyroid disease, inflammatory bowel disease, vitiligo, alopecia areata, and/or dermatitis herpetiformis.
†† Child's symptoms indicative of CD: often or always fatigued, abdominal pain, abdominal discomfort, flatulence, hard stools, and/or soft stools.

family, and whether the child had other diseases or symptoms that may indicate CD (Table 6); however, reported well-being of the child was not a significant explanatory factor. Also, the parents' maximum WTP was significantly increased with higher education and income and with child symptoms that may indicate CD. The other potential explanatory factors mentioned above did not, however, influence the maximum WTP, not even having a previously diagnosed CD case in the family (Table 6).

DISCUSSION

In our study, 63% of the parents were willing to pay something for a CD screening of their child. The mean WTP was 79 EUR, which is higher than the average cost of 47 EUR per child. Only 23% of the parents, however, were willing to pay at least this cost, illustrating that the mean WTP is strongly influenced by some parents responding with high amounts. The main reason for not being willing to pay for a CD screening was that they had no suspicion of their child having CD. Parents' stated WTP was higher for those with more education and/or a higher income, a relation shown in other WTP studies (30). Also, parents' WTP was higher if their child had symptoms that in the scenario had been described as possibly suggesting CD. A surprising result was that previously diagnosed CD in the family only meant that a larger proportion of parents were willing to pay something for CD screening, but not that their stated WTP was significantly higher. One reason may be statistical uncertainty because only 2% reported previously diagnosed CD in the family.

An important issue for interpreting our results is the costs and benefits the respondents included as a basis for their stated WTP. In the questionnaire, the respondents were told ''to think of a situation where the only way to find out if your child has gluten intolerance is that your household pays for it, i.e., for a blood sample and, if needed, also the follow-up investigation.'' With this background we have chosen to compare stated WTP with these explicitly mentioned costs; however, in the scenario, long-term potential health consequences were also mentioned (Fig. 1). For the lifelong perspective, the value of improved health-related quality of life, an increase in productivity, and the savings in health care costs when the sequels are avoided appear on the benefit side. A drawback of CD mass screening is the increased food expenditure from following a gluten-free diet. Although specifying the screening components for the respondents, one cannot rule out the possibility that some also considered the long-term effects. In such a case, our WTP estimate may be overstated. Notably, our CD screening was performed in a setting with unexpectedly high prevalence of CD (3%), and with two-thirds of the CD previously undiagnosed (6), which increased the cost.

There is an inherent problem in providing medical information to laypeople. All of the important information needs to be summarised and translated to everyday language. The space available is extremely limited in a questionnaire study and cannot be overloaded with information. Furthermore, the medical profession has not always arrived at complete consensus. One controversy in this particular case is clinically silent CD and the possible benefits of treating it. Among others, Kumar argues against screening for such reasons (12). On the contrary, there are indications that those with clinically silent CD will have symptoms later if not put on a gluten-free diet (31). The scenario did not explicitly state that CD can be clinically silent; however, it was indicated that this disease typically has vague or even no symptoms, which also is commonly referred to in the country's well-known mammography screening. Regarding health benefits, we used the wording that individuals affected by CD ''normally'' improve their health if excluding food containing gluten. We think this wording indicates that not everybody benefits from the gluten-free diet. Despite our attempt to provide balanced information, some may have overestimated the potential benefits, which could possibly lead to bias in our estimate.

Of 10,041 invited children (and their parents), as many as 2474 did not participate. Considering that they did not enter the ETICS study, which was free of charge, it is likely that a high proportion of these children would not participate in a scaled-up screening. If our assumption about a low participation rate is correct, this group will have only a small influence on the cost per child for CD mass screening, and the effect on the stated WTP would most likely also be small. Also, in our main approach we extended our inclusion criteria to allow for WTP responses expressed only in text and with imprecise amounts, in an effort to reduce bias in the WTP estimates. We are aware that this is not common practice; however, we have not found any information in the literature on how to handle such responses for the open-ended format. Despite this, we considered this approach to be a fairer way of handling responses intended to give a positive value than to designate them as missing values.

It is recommended that WTP be considered in the context of alternative use of money (32,33), which was not explicitly the case in our study. When no alternatives to the intervention under study are given (eg, diabetes screening or even something outside the health sector such as a sports centre), it tends to inflate the WTP of the intervention in focus (34). There is also evidence from many studies that there is a difference between people's responses and their actual WTP, implying that people state an exaggerated WTP (27). These facts suggest that parents likely overestimated their WTP in the present study. There are also several reasons to believe that our result is an underestimation of their WTP. First, in Sweden, health care for children is free of charge, and citizens pay higher taxes than in many other countries. Responses clearly indicated that parents were not willing to pay ''a second time'' for a CD screening, which will contribute to an underestimation of both the proportion willing to pay anything and the mean WTP. Second, the open-ended format used in the present study has been shown to give lower WTP estimates compared with other formats (28,35). Finally, our WTP analysis was restricted to 4929 responses, implying that as many as 1423 were excluded. Among the excluded, there were many more willing to pay than unwilling to pay for CD mass screening.

There is an ongoing debate about potentially introducing CD mass screening (in high-prevalence societies), as a complement to present active case findings strategies (9,22,23). Results from the comprehensive health economical evaluations done so far suggest that this may be cost-efficient under certain circumstances (14,15). However, the modelling in these studies is dependent on several assumptions that are not yet fully validated, which the authors also state. Health-related quality of life in relation to CD is one of these issues (31,36,37), but still the knowledge is insufficient for screening-detected CD cases before and after initiated gluten-free diet. An alternative to mass screening could be to introduce ''selective'' screening offered to those willing to pay at least the cost involved, which would be supported by our finding of a skewed distribution of WTP. This would require a different screening strategy, for example, inviting families to have their child tested at the health care centre, likely resulting in higher costs per child than in the present study. It is also doubtful whether such a strategy would be in accordance with the equity goals formulated in Swedish law and policy. Screening of high-risk groups for CD, for example, those with diabetes and Down syndrome, is no longer a controversial issue (38).

The present study contributes to the ongoing debate about potential future CD mass screening in high-prevalence societies. In a field with only 2 solid health economic evaluations to date, the present study is important because it is the first to estimate parents'

experienced value of the information whether their child has CD or not. We have demonstrated that Swedish parents' WTP for a school-based CD screening on the whole is higher than the cost; however, only a minority of the parents were willing to pay an amount covering the cost for screening their child. Parents' WTP increased with higher education and income and with child symptoms that may indicate CD.

Acknowledgments: We thank all of the parents who responded to the questionnaire.

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