

Decision support in Down's syndrome screening using multi-criteria decision analysis: a pilot study

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ABSTRACT

BACKGROUND: the aim of the study was to develop and pilot use of a decision support system (DSS) to help women choose the option that best respects their personal values among the different screening/diagnostic tests for Down's syndrome.

METHODS: value-bearing considerations were elicited through qualitative interviews. Ten women post-birth and ten health professionals working in the Obstetric Department at UCLH were interviewed. Performance data for the various possible screening strategies on these attributes were entered into a Multi-criteria Decision Analytic model using the Annalisa implementation. Participants piloted the DSS, entering necessary weights for the attributes and observing the resulting scores. Main outcome measures were DSS clarity, usefulness and feasibility in a clinical setting.

RESULTS: most participants found the DSS valuable because it stimulated women to seek information about testing and helped them focus on the main issues affecting their decisions. Annalisa proved a user-friendly DSS that helps women understand the issues around Down's screening and diagnosis. There was unanimity that its use should be complementary to health professionals' consultation. Most favoured offering it before consultation so that women could be better informed about options before attending the antenatal booking.

CONCLUSIONS: the overall positive comments confirm that a user-friendly decision analysis-based support system can be a valuable instrument at supporting health decisions in this area. Further research is needed to assess whether the intention to make an informed choice is always best addressed by a decision support system, or these remain useful tools only to women more inclined to seek information anyhow.

Key words: Down's syndrome; Screening/diagnosis; Support system; Multi-criteria decision analysis

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INTRODUCTION

Down's syndrome is one of the commonest chromosomal abnormalities, diagnosed

prenatally in about 1.57 per thousand women and affecting about 1.05 live births per thousand in the UK [1, 2]. A wide range of biochemical tests, in conjunction with measurements

taken during antenatal ultrasounds have been developed to assess the probability of Down's syndrome more precisely than can be achieved using maternal age alone [3, 4]. These are routinely used in clinical practice as non-invasive screening tests [5].

In the UK women are initially offered a screening test on the understanding that there is a possibility that they will be offered a further diagnostic test if the result shows that they are at high risk [6]. The most effective available options are Combined and Integrated tests, both integrating the risk related to maternal age alone with biochemical tests and ultrasound findings. Combined testing is performed between 11-13+6 weeks with ultrasound measurement of Crown-Rump-Length (CRL), nuchal translucency and blood for Pregnancy-associated plasma protein A (PAPP-A) and Beta Human Chorionic Gonadotropin (β -hCG) and a result within the 1st trimester. Integrated testing is the scan information and PAPP-A, bloods taken again at 15-20 weeks for Alpha Fetoprotein (AFP), unconjugated oestriol (uE3), inhibin-A and β -hCG and only then a result issued. A summary of performance

characteristics of the different screening tests is available in Table 1.

Women at high risk from screening, should be offered a diagnostic test, either amniocentesis or chorionic villous sampling (CVS), depending on the gestational age. The woman could decide to proceed or to opt out of further investigations, since these invasive procedures are related to a risk of miscarriage estimated as around 1.4% for amniocentesis and 1.9% for CVS [7], including spontaneous and procedure-related foetal loss. If a woman proceeds to diagnostic testing and confirmatory abnormalities were identified, she would be offered the option of a termination of pregnancy.

However, many women find it hard to interpret the results of screening tests and, even more important, to determine which testing strategy is best for them as an individual [8]. Decision aids have the potential to facilitate their making appropriate choices [9, 10].

It is essential to distinguish between the descriptive and prescriptive uses of decision models. Apart from qualitative studies [11] large number of decision models, including decision analytic ones, has been used to *describe*

TABLE 1

DOWN'S SYNDROME SCREENING TESTS PERFORMANCE				
TEST CHARACTERISTICS	GESTATIONAL AGE		TEST PERFORMANCE DR(%)=85%	
	11-14 WEEKS	15-20 WEEKS	FPR (%) (FALSE POSITIVE RATE)	N DS PREGNANCIES DETECTED FOR EACH PROCEDURE-RELATED UNAFFECTED FOETAL LOSS
Double	-	β -hCG, uE3	13.1	1.8
Triple	-	β -hCG, uE3, AFP	9.3	2.6
Quadruple	-	β -hCG, uE3, AFP, inhibin A	6.2	3.8
Combined	NT, β -hCG, PAPP-A	-	6.1	3.9
Integrated	NT, PAPP-A	β -hCG, uE3, AFP, inhibin A	1.2	19.2
Serum Integrated	PAPP-A, β -hCG	uE3, AFP, inhibin A	2.7	9.1

All tests include maternal age and free β -hCG is used rather than total hCG. The first trimester markers PAPP-A and free β -hCG are based on the median in DS pregnancies at 10 completed weeks and the NT SD in unaffected pregnancies is applicable to 10 completed weeks. Number of DS detected is obtained dividing number of procedure related unaffected foetal losses in 100 000 women screened (80% uptake rate for Amniocentesis or CVS and 0.9% foetal loss rate attributable to the procedure) by the number of DS pregnancies detected (assuming 90% uptake rate for Amniocentesis and CVS because women with affected pregnancies tend to have higher risk and so are more likely to accept diagnostic testing).

This table has been partially reproduced from SURUSS, Wald et al, 2003 [39].

women's prenatal choices. The most prevalent have been subjective expected utility models [12] and social cognitive models [13-15].

There is clear evidence that decisions about prenatal testing are determined not only by Down's syndrome risk status, but also by psychosocial factors [16]. Furthermore we know that the prospect of different situations or outcomes vary widely between women e.g. anxiety due to not knowing baby's status or losing a normal baby through miscarriage [17]. It is also well accepted that decisions involving risk are affected by emotions, emotions playing a critical role in the decision-making process [14, 18, 19]. Furthermore recent findings in neuropsychology show that decision-making involves emotions [20], especially moral decisions involving possible negative consequences for another person, engage emotions [21].

To include psychosocial variables and the role of subjective norms in influencing screening and diagnostic tests seeking, Social Cognitive Models have been enrolled to explain Down's prenatal decisions [13-15].

The purpose of the modelling underlying Decision Support Systems or Aids modelling is not to describe or explain the way decisions are made, or the choices that women actually make in our Down's syndrome case. By definition the aim is to move the eventual decision closer to some normative standard of a 'better decision', which itself requires to be defined and may involve both process and outcome criteria. A *prescriptive* standard represents some compromise between the *normative* ideal and the *descriptive* reality. Any actual decision aid will necessarily occupy some intermediate position between these poles.

We have chosen to implement an aid based

on the well-established technique of multi-criteria decision analysis (MCDA). Within the MCDA framework we have taken particular positions on four key issues (i) the criteria which appear in the model are regarded as fully compensatory (if there are non-compensatory attributes for an individual women, such as termination, these are assumed to be addressed out with the aid); (ii) the model involves the clear separation of beliefs (probabilities) and preferences ('utilities'); (iii) the magnitudes of both are based on scales with ratio properties; and (iv) the two types of input are integrated into an overall score for each option by the principle of expectation.

The aim of the present study was to elicit women's main concerns (considerations, attributes, outcomes) in making decisions about Down's syndrome screening/diagnosis and to incorporate them into a Multi-Criteria Decision Analysis model acting as a computer-based decision support system via the Annalisa implementation of MCDA. The system was piloted in a clinical setting to generate feedback before testing on a larger scale.

METHODS

To identify the main attributes to be incorporated into the decision analysis model, 20 semi-structured interviews were carried out with pregnant women. To include a wide variety of opinions women were selected by parity and maternal age. The hypothesis was that the decision-making process could have been affected by previous antenatal experiences since attitudes might change after experiencing pregnancy and childbirth [22]. For ethical reasons women diagnosed with a Down's syndrome

TABLE 2

SEMI-STRUCTURED QUESTIONNAIRE USED TO ELICIT VALUES TO BE INCLUDED INTO THE MCDA MODEL

- 1) What experiences have you had in relation to screening or diagnosis of Down's syndrome in pregnancy?
- 2) What options were available to you?
- 3) What information did you want before reaching a decision about screening/diagnosis?
- 4) What information were you were given, before reaching a decision about on screening/diagnosis?
- 5) Probe for what was helpful or clear, and what was unhelpful, confusing, alarming
- 6) How easy or difficult was it to reach a decision on screening/diagnosis options?
- 7) What factors influenced you most in reaching a decision?
- 8) Probe for helpful, unhelpful factors etc.
- 9) How do you think this aspect of antenatal care should be handled in future?

baby in previous pregnancies were excluded to avoid triggering distress at recalling relevant events. Women accessing UCLH antenatal care service for a 20-weeks scan were recruited accordingly and interviewed using a semi-structured questionnaire available in Table 2.

Information was recorded and then analysed according to the principles of grounded theory because of the flexibility of this approach. Data analysis was carried out using N-vivo software. The interviewing and analysing process ended when we felt we had reached saturation.

The attributes arrived at for the model, were as follows: having a baby with Down's syndrome; having an early miscarriage as a consequence of testing (first trimester); having a late miscarriage as a consequence of testing (second trimester); having an early termination of pregnancy (first trimester); having a late termination of pregnancy (second trimester); test "bother" (defined as the physical and psychological discomfort related to different screening/diagnostic strategies); and uncertainty/anxiety associated with the testing

(for which the duration of any uncertainty was used as a proxy).

The MCDA model requires ratings for each of the testing strategies on each of these attributes. We established the best available mean estimates from the literature (including age-specific rates where relevant). A snapshot of the Annalisa screen for a 35-year-old woman appears in Figure 1. It should be noted that while the best available evidence at the time of testing was entered, the focus of this pilot was exclusively on the interface, rather than the substantive content and implied recommendations.

Ten women who had already given birth to a healthy baby and ten health professionals working in UCLH Foetal Medicine Unit were then recruited between May and August 2008. After they were given instructions (Table 3), women & professionals piloted the DSS for clarity, usefulness and feasibility in a clinical setting. Through a brief questionnaire (Table 4) the researcher collected discussion notes to evaluate patients' & professionals' opinions.

FIGURE 1

ANNALISA SCREEN AS PRESENTED TO SUBJECTS, REFERRING TO A 35 YEAR OLD WOMAN



Women would extend the weighting bars as much as they feel the specific factor being relevant in the decision, based on their personal preferences. At the end of the weighting process Annalisa would show (on the top of the screen) a percentage score corresponding to the different options of being screened/diagnosed for Down's syndrome, based on the expressed preferences. The best option would be highlighted using a different colour (bright orange). In this case, based on the preferences of a 35 years old woman, the best option would be undertaking a combined test, if positive a CVS, if positive a termination of pregnancy. Note that weightings are displayed in non-normalised form, which is one of the optional settings in Annalisa. The scores are always based on normalised weights however, i.e. ones that add to 100%.

RESULTS

General Impressions

Both groups of women were asked to give their overall impressions after their experience of the aid and, in particular, whether they found it interesting and whether it stimulated their curiosity.

All ten women found it provided support to their decisions. "It's interesting and I would like to know more about it" (WW 7). Three said what they liked most about it was its simplicity and straightforwardness. Two women focused on the possible practical and effective support that the DSS could give to women in taking such a difficult decision: "It's interesting because people

TABLE 3

INSTRUCTIONS ON HOW TO USE THE DECISION SUPPORT SYSTEM GIVEN TO WOMEN AND HEALTH PROFESSIONALS FOR PILOTING THE DEVELOPED MCDA SUPPORT SYSTEM

This decision support system has been created to help pregnant women to make an informed choice about their Down's syndrome antenatal care. It is based on the best available epidemiological evidence and on information collected through interviewing pregnant women about their concerns in taking the decision. We are now trying to assess whether this tool could be a feasible, comprehensive and valid instrument to help women in taking the decision, and to refine the model on the basis of your very much welcomed critiques.

The idea behind this support system is that each woman, on the basis of her personal value, would weight each concern (attribute into the model) in a different way. Her decision should then be taken on her own personal values and what is most important to her.

On the screen in front of you, you will see two different parts: Upper and Lower. First concentrate on the Lower part of the screen (Weighting) and start your "weighting process".

You will find different concerns that women have identified and told us as the most important in taking this decision. Below each concern you will find a blue bar. On the basis of what is important to you, you are asked to enlarge the bars; for example if it's very important to you not having a baby with Down's you would enlarge the bar corresponding to it.

Once you have enlarged all different bars related to concerns on the basis of your own preferences and you are satisfied with your weighting process, look at the Upper part of the screen (Scores). In this part, you will see different screening/diagnostic strategies listed and ranked. The best strategy, on the basis of the expressed preferences, will be represented by a bright orange bar. After you are finished, you'll go through your results with a midwife, who will help you out to understanding your results.

WHAT DO THE ATTRIBUTES/CONCERNS MEAN?

Having a baby with Down's: how much is relevant to you not to have a baby with Down's syndrome

Miscarriage: how much is relevant to you not to lose the baby

Early Miscarriage: Miscarriage happening in the first trimester of pregnancy

Late Miscarriage: Miscarriage happening in the second trimester of pregnancy

Termination: how much is important to you not to terminate a pregnancy

Early Termination of pregnancy: Termination of pregnancy in the first trimester of pregnancy

Late Termination of pregnancy: Termination of pregnancy in the second trimester of pregnancy

Test "Bother": how much is relevant to you "bothering" related to different tests, either in terms of commuting, physical and emotional discomfort

Uncertainty/Anxiety: how much is relevant to you the level of uncertainty and anxiety related to not being 100% sure about status of the baby

TABLE 4

BRIEF QUESTIONNAIRE FOR PILOTING THE DEVELOPED MCDA SUPPORT SYSTEM IN A CLINICAL SETTING

DOWN'S SYNDROME PILOTING TOPICS GUIDE

Preliminary

1. Which kind of decision support would you prefer?
 - Talking to Health Professional
 - Computer
 - Both
 - In which order
2. Are you familiar with computers?
3. Do you have information about screening strategies available to you?
4. Which test did you undertake in your last pregnancy?

AFTER HAVING USED DSS

5. How did you find the tool?
 - Interesting/curiosity
 - useful
 - colourful and attractive
6. What didn't you like about it?
7. Did you find anything difficult to understand or that could have been explained better? Did you find anything unclear or confusing?
8. Did it make you think about the options and your knowledge of the different strategies available to you?
9. Do you think you have better considered consequences related to the different options available? Do you think it have helped you to focus your main concerns in taking these decision?
10. How do you think it could be used? Do you think it should be used before or after consultation?

would find difficult to think about concerns and take decisions" (WW 2); "If you play around it gives you a sensible decision" (WW 4). One woman added that she was particularly interested in the "test bother" attribute and expressed her surprise: "I'm interested in test bothering, because I wouldn't have thought that a woman did care about it. It's interesting that someone mentioned it" (WW 9).

Health professionals had similar general impressions. In fact four of the ten highlighted the simplicity and straightforwardness of the aid. Most of the comments were positive, such as "it's a very good idea", "fascinating", "very easy to use", "very simple". Three of the health professionals stressed the good coverage: "It covers everything that would be a concern in taking decisions" (HP 7); "Because there are all the outcomes involved in pregnancy" (HP 10); "I think it's a good way to make them think about it and make informed decisions rather than go for what they are told" (HP 2). Only one interviewee voiced a concern: "It's also an emotional decision and it doesn't take account of it. Because of anxiety it would be difficult to go through the weighting process" (HP 5).

Usefulness in decision-making

Subsequently the women and health professionals were asked about the usefulness of this tool in supporting the decision making process of women regarding their Down's syndrome antenatal care. All confirmed the helpfulness and effectiveness of the aid, giving reasons such as: it would make the decision easier, it would help to get a general idea complemented by antenatal discussion, it would help to make decision and have time to decide about having the baby and not discovering later. Health professionals thought that it could help to clarify women's thinking, especially because most women are not aware of the available options. "It could be very helpful in terms of evaluating pros and cons and see where you sit in a way... It could be useful because they would have options and concerns in front of them" (HP 7). Another suggested that the partner's opinion might be included in the model or, as an alternative; it would be desirable to use the aid with the couple instead of the woman alone.

Visual Aspects

Every one of the 20 interviewees found the aid visually simple, practical to use and easy to manipulate. "It's quite clear, easy to the eyes" (WW 9), "I like it graphically, it's nicely done" (HP 5) "I don't think there is anything would put off people to using it" (WW 4). One woman and two health professionals particularly appreciated the choice of using increasing the length of bars to weight values, viewing it as a straightforward way to give your opinion corresponding to what you feel. "I like the design...percentage and bars all on the screen in scores" (HP 7). One woman suggested that it would be better to expand vertically rather than horizontally and that the weighting part should be in the upper part of the screen; while another proposed a pie chart instead of bars.

Dislikes

When women were asked what they dislike about the tool, five out of 10 responded that there was nothing in particular they didn't like about it. The remaining five women:

- found negative attributes difficult to understand
- experienced weightings as 'sort of arbitrary'
- thought a scale between 0 and 10 instead of 0 and 100% would have been more familiar
- that it would need to access more information through links to other websites, especially about other abnormalities which might be uncovered by tests
- thought it would be quite difficult for people not familiar with computers
- found it difficult to understand percentage scores.

Among health professionals, concerns were also varied. Four health professionals were worried about weighting concerns using normalised values (i.e. having all other weights adjust when one was changed). Two highlighted the complexity of handling percentage scores and one of interpreting negative attributes. One emphasised the need to link attributes and screening/diagnostic options to appropriate websites to get information and explanations. It was also suggested that

the "test bother" attribute should be refined. One of the interviewees suggested that the relative rating of test bother should be higher for the option "combined test plus CVS" rather than "Integrated plus amniocentesis". Another professional recommended separating emotional and physical bother, since it would be difficult to incorporate them in a single bar that gave appropriate weight to both. Finally, two suggested hiding the scores until the weighting process is completed otherwise it could influence women while they are balancing their concerns.

Clarification

Women and health professionals were asked to identify anything, which was unclear or confusing. Four out of ten women couldn't recognize any specific element of confusion and a couple of these women appreciated the explanation sheet given to them. The remaining patients commented mainly on the incompleteness of the legend offered to women as an explanation to use the aid autonomously and the amount of information about strategies and values received at the piloting. "You should be able to get information about the different concerns" (WW 1); "It doesn't tell you how to expand the bars" (WW 8); "It would be better if on weighting for each concern there was a link to explanation instead of using an information sheet" (WW 9); "An explanation of the options is missing" (WW 10).

None of the health professionals involved in the piloting identified anything that was confusing or unclear. One suggested that it could be better to start the weighting process having all the bars at the same length to avoid influencing women's decision-making process.

Stimulation

Our interviewees were asked to express an opinion about the tool's capability to stimulate thoughts about options or to increase demands for knowledge.

Eight out of ten women thought that it was a good first approach to stimulate thinking and searching for knowledge about options available. "I think it would get people more aware" (WW 2); "I suppose they could be

stimulated to find out more" (WW 4). Three of these women suggested that the aid could help on this matter only if either women had a background knowledge about options or used the tool in the appropriate setting with a health professional taking them through the strategies available to them. One woman underlined the necessity of getting more information at the GP practice and the aid would be useful in reaching this objective while another pointed out that the aid could help to make women generate questions and consequently enhance the search for knowledge.

All health professionals thought the tool as a possible instrument to stimulate women's search for knowledge and consequently a better understanding of screening and diagnostic strategies. To make it easier for women, an option would be to link strategies to informative websites, two professionals suggested, echoing a recurrent theme in the feedback. "Because people like different ways of getting information and this is a kind of interactive tool...this would help" (HP 6); "I think it would help women thinking about it" (HP 7); "They would be stimulated in finding out more" (HP 10). Among our interviewed health professionals, one pointed out that the aid would stimulate the search for the personal acceptable level of risk for a specific woman. Another affirmed that the tool would encourage the search for knowledge through generating questions about screening/diagnostic strategies. On the other hand, one health professional expressed concern that, through encouraging to think about options and main issues involved in the decision, the aid could generate anxiety.

Focusing

Women were asked whether the DSS would lead users to consider the consequences of different strategies and focus their main concerns. 80% of them answered positively. "It would encourage people to find out how they feel about different outcomes" (WW 2); "It could be useful for women to realize what is behind decisions" (WW 8); "It relates each test to its consequences, to what could potentially happen" (WW 9); "...women would think about different factors affecting the decisions rather than going for what they are offered" (WW 10). Another woman highlighted the

possibility that instead of considering the possible consequences, women might trust the aid's suggestion too much, and accept the option for granted rather than make their own analysis of pros and cons.

Eight out of ten health professionals also answered positively to this enquiry on the focusing benefits of the aid. "It will force you to weight the concerns, the elements involved in the decision. It's a practical way of focusing your concerns" (HP 4); "Yes and it helps because these attributes...because sometime it's difficult to talk about feelings related to termination of pregnancy for example..." (HP 6). One health professional suggested that it would be difficult to predict women's behaviour after using the aid. Another interviewee echoed the concern at the end of the preceding paragraph that, just as some women now were reluctant to take responsibility for an informed decision and preferred a paternalistic approach from health professionals, they could use the aid in the same way - instead of being a stimulus to make an informed choice the tool would be another way of being guided to do what was offered. Finally one health professional thought that, if the aid was available as an option it would probably be used, or used most, by a category of women already interested and looking for knowledge. The most challenging group as far as the health professional was concerned may not take advantage of it.

Where and When

Finally our interviewees were asked to give their views about how the aid could be used and whether it would be better to use it before or after the antenatal consultation. Every one of our interviewees thought that it should be complementary to the antenatal consultation with health professionals and not replace it. On timing the entire spectrum of opinions existed. Four of ten women suggested that the best use would be before consultation: "They could use it on a website and then come in for a consultation" (WW 8). On the other hand two women suggested using it during consultation: "I think people should be assisted. May be it should be used during a consultation with a midwife or a doctor. I think it would be more useful for health professionals. It would help them understand which kind of person they

have in front of them and how to approach a specific woman" (WW 5). One suggested using it both before and during consultation: "Probably both on a website and then at consultation. It would be easier for the user to ask questions. It could be even more confusing though to use this aid if it doesn't explain by links to websites the different options" (WW 10). Two women suggested using it after consultation: once the necessary information is collected through the dialogue with a health professional, women would be able to use it either at home or in the waiting room.

Among health professionals, half suggested using it before consultation - at a GP practice, by accessing it on a website at home, or in the waiting room before the consultation at the antenatal care unit. The reasoning for this timing was mainly to give women time to generate and prepare their questions before the antenatal visit. One professional suggested that the aid should be used during consultation: "First women should be explained about options, then use the model then discuss with a midwife. They should use it with a midwife all the time" (HP 2). One health professional indicated as the best moment, after consultation: "It's a useful tool to think about what you want after you receive information" (HP 5). Finally two professionals recommended using the aid before and after consultation: "Use it on a website before going to a consultation, with link to publications. To use it twice, before and after consultation: first time told by the GP, getting information from websites and clarify women's thinking; then use the links to collect information and re-use it again after discussion" (HP 3).

DISCUSSION

The general reactions of both women and health professionals to the support system were very positive. Specifically they appreciated simplicity, straightforwardness and the coverage of all the different aspects involved in women's antenatal care choice.

Its usefulness in taking the decision was identified because it would clarify women's thinking and enhance awareness of available options. Our findings support the general opinion that decision aids improve knowledge of the options available and generate accurate

perceptions about the benefits and harms of different options [23].

Furthermore interviewees considered the aid a valuable instrument to stimulate women thinking and searching for knowledge about options. This general opinion confirmed that the use of decision aids in a clinical setting could improve patients' knowledge among other benefits [24, 25] and this could be highly relevant in this context since women's understanding and awareness are in some cases extremely low [10, 26].

Both health professionals and women highlighted how the aid could encourage people to consider consequences of different strategies and focus their own concerns related to a single choice. Our findings reaffirmed multi-criteria decision analysis as a valid basis for a decision support tool in this context where multiple options are available, information is not widely diffused and it is uncertain which would be the best course of action for a single user [27].

Most interviewees showed an overall appreciation for the visual presentation of the aid. Few suggestions of improvement were proposed, but some comments were in contrast with previous feedback at the design stage. In fact using vertical bars instead of horizontal ones, or a pie chart, to enter weightings had been rejected by the developers on the basis of previous feedback. The suggestion given to link different strategies to informative websites to satisfy the demand of knowledge and stimulate awareness has already been implemented into Annalisa version 2. Finally, two health professionals suggested hiding the scores until the weighting process is completed otherwise it could influence women while they are balancing their concerns. This is also possible in the new version and has claims to be regarded as best practice.

Concerns about the possibility that the aid, through stimulating to think about options and issues involved in the decision, could increase anxiety were expressed. It would be a misconception to consider the aid as a cause of emotions like anxiety. In fact the evaluation of options and main issues involved in a risky decision has been demonstrated to intrinsically determine such events. The importance of emotions in the decision-making process has been recognized especially in decisions involving a certain level of risk [14, 18, 19]. Moreover, the most recent Cochrane review

on this topic demonstrated no significant difference in anxiety levels between groups using decision aids and those experiencing usual care [23].

Different decision analysis models have been applied to explain women's prenatal choices, specifically subjective expected utility theory (SEU) [12] and social cognitive models [13-15]. Despite the well-known positive characteristics of the mentioned decision models, they do not include the emotional aspects of decisions. In contrast, Multi-criteria Decision Analysis methods clearly separate weightings that represent the subjective importance of the criteria from the ratings that represent evidence about consequences [20]. These characteristics of MCDA ensure there is a clear route for the representation of emotions in the model. The criticism that the aid does not or cannot take account of the emotional component of women's choice is therefore a misperception.

Some of the suggested improvements related to ineffective delivery of the aid, are being addressed before testing on a larger scale. Despite our a priori beliefs that an information sheet could have been practically easier to use, one patient suggested links as a better option. Rollover pop-ups in version 2.0 provide this functionality. Other simple improvements regarding practical matters such as how to expand the bar on the screen and how to start the process with bars all at the same length, if they wish to, could easily be implemented while testing on a larger scale.

Another category of suggestions was related to possible cognitive limitations of respondents and whether or not these should be addressed in the aid or outside it. Difficulties in understanding negative attributes, normalized values, percentage scores and a scale ranging from 0 to 10 were noted. Concerns about possible technical complications if the aid was used by 'computer illiterates' were expressed.

Furthermore it was proposed to divide "test bother" in two different values: physical and psychological discomfort. It would be difficult to assign an appropriate score to a single bar, since the perception of the two discomforts is dichotomous. This assertion is in contrast with previous qualitative work, analysing psychological and physical aspects related to prenatal diagnostic tests. In fact in previous work interviewed women were

able to distinguish physical and psychological consequences of prenatal diagnostic testing and to a certain extent to quantify the level of concerns related to different aspects of the process they went through [28].

Moreover "test bother" value was considered inappropriate and health professionals suggested refining it. "Test bother" rating related to "combined test plus CVS" should be higher. In fact, in accordance with previous literature, physical discomfort was perceived by our health professionals as greater for CVS, rather than amniocentesis [29, 30].

Assuming the involvement of a partner in the decision, one woman suggested that partner's opinion should be included into the model. Otherwise the model should be used with couples rather than a single individual. Using the DSS, either women or couples would be able to give subjective weight to single concerns on the base of personal preferences [20]. Women would find the elicited attributes on a computer screen and could visualize and focus their beliefs about possible outcomes and contemplate values associated with them. This procedure would stimulate women's or couples' awareness of well-founded values [31] affected at different levels by norms, experiences and emotions.

There was unanimity that the aid should be complementary to antenatal discussion with health professionals and a variety of alternatives about timing and settings for aid use were favoured. The majority of people planned to use it before consultation to increase awareness and prepare themselves for consultation. Despite this general opinion, previous findings suggested that interventions before medical consultations produce only limited benefits to patients, but the relevant interventions were mainly question checklists and patient coaching [32, 33].

The majority of women expressed the desire to use the decision aid autonomously, either on a website or in the waiting room, but the entire spectrum of options was covered. These findings confirm the theory that patients more and more express the desire and expect to be involved in their health decisions [34]. The present conclusions support the recent trend in health policy encouraging health professionals to move away from a paternalistic approach and to empower women to make more autonomous health choices [12, 35]. A couple

of interviewees expressed concerns about the ability of women to use the aid appropriately and merely reproduce one paternalistic approach with another. But a recent review showed a gap between women's intentions to make an informed choice about prenatal screening and their actual skills in achieving it and thereby encouraged the implementation of useful interventions aiming to aid women taking the appropriate decision [10].

CONCLUSIONS

The overall positive comments of participants confirmed previous findings that decision aids are a valuable instrument to support health decisions, particularly in obstetrics [36-38].

One of the strengths of our study was to involve both patients and health professionals in this pilot. In fact further research is needed to evaluate acceptability of decision aids in this context, in particular by health professionals [38]. The most relevant limitation of our study was the small sample size to test the developed support system, but the intention was to conduct a pilot to refine the DSS based on users' preferences, and then to test it further on a larger scale.

One of the interviewees suggested that people accessing the support system could be more inclined to seek information anyhow, resulting in a selection bias in our sample. Further research is needed to assess whether the common intention to make an informed choice is always best addressed by a decision support system, or whether these are useful tools only to women who seek to achieve an informed choice anyway.

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