The following full text is a publisher's version.

For additional information about this publication click this link.
http://hdl.handle.net/2066/69097

Please be advised that this information was generated on 2017-07-07 and may be subject to change.
A review of decision support technologies for amniocentesis

M.A. Durand¹, J. Boivin² and G. Elwyn¹,³

¹Department of Primary Care and Public Health, School of Medicine, Cardiff University, Neuadd Meirionnydd, Health Park CF14 4YS, UK; ²School of Psychology, Cardiff University, Psychology building, Park Place, Cardiff CF10 3AT, UK
³Correspondence address. Tel: +44-2920-687195; Fax: +44-2920-687219; E-mail: elwyng@cardiff.ac.uk

BACKGROUND: There is an increasing interest in designing decision tools [decision support technologies (DSTs)] that support patients when they have to decide about health matters. The purpose of this review was to describe and evaluate existing DSTs for amniocentesis testing. METHODS: Ten medical and psychological databases were searched up to January 2008 and key authors and organizations contacted to identify DSTs for amniocentesis (published or otherwise). DSTs that described amniocentesis testing were included. RESULTS: Six DSTs met the inclusion criteria and were evaluated using the International Patient Decision Aids Standards Instrument. The evaluation suggested that most DSTs provided a satisfactory level of information on the index decision and, on the specific features of a diagnostic test; provided structured guidance in making a decision; were based on scientific evidence and disclosed the funding sources and authors’ credentials. However, most DSTs failed to communicate probabilistic information, to clarify patient values and use plain language. The majority of DSTs did not use a systematic development process. Furthermore, the DSTs’ evaluation often lacked scientific rigour. In most cases, neither the quality nor the effectiveness of the DST could be inferred from the evaluations. CONCLUSIONS: The review highlights variations in the development, evaluation and quality of existing DSTs for amniocentesis. We do not know what impact DSTs may have when implemented in clinical settings. Decisions in this context have high stakes and strong emotional impacts. It is important to ensure that DSTs achieve high standards.

Keywords: decision support techniques; decision aids; amniocentesis; prenatal testing; prenatal diagnosis

Introduction

Women need to be informed about the benefits and potential harms of diagnostic procedures such as amniocentesis. Amniocentesis is a procedure to remove a small sample of amniotic fluid from the amniotic sac surrounding the fetus in the womb. It is the most common method of obtaining fetal cells for karyotyping and is usually conducted after 15 weeks of pregnancy. Amniocentesis carries a risk of miscarriage; the most often quoted rate is one miscarriage per 100 procedures (Papantoniou et al., 2001) and may lead to a decision to terminate the pregnancy (Asch, 1999). The decision to undertake amniocentesis is generally preceded by the decision to undertake prenatal screening tests.

Prenatal screening tests, i.e. ultrasound scan (nuchal transluency scan) or blood tests (maternal serum screening tests), determine the woman’s chance of having a baby affected by Down’s syndrome. The British National Health Service offers maternal serum screening tests (triple tests, quadruple tests, integrated tests or combined tests) for Down’s syndrome to all women between 15 and 18 weeks of pregnancy (Weisz and Rodeck, 2006). About 5–10% of women who undertake second-trimester maternal serum screening test receive a high-risk (of abnormality) result and are offered amniocentesis (Benn et al., 2006; Gidiri et al., 2007). Any pregnant woman undertaking prenatal screening for Down’s syndrome may have to decide about amniocentesis and face the consequences this might have on her pregnancy, life and family. The decisions to have prenatal screening and amniocentesis testing are closely related. Expectant parents should be informed about the benefits, potential harms and implications of invasive diagnostic procedures such as amniocentesis before embarking on prenatal screening. Research shows that pregnant women commonly undertake screening tests for Down’s syndrome without realizing they could be offered an amniocentesis, receive a diagnostic of chromosomal abnormality and/or be offered a termination of pregnancy (Jaques et al., 2004; Dormandy et al., 2006).

The decision to undertake prenatal screening tests along with the anxiety subsequent to an indication of high risk have been extensively documented in the literature (Santalahti et al., 1998; Michie et al., 1999; Wildschut et al., 2006). However, the amniocentesis decision has been commonly confounded with the decision to undertake prenatal screening tests. As amniocentesis is only offered to a minority of women who have prenatal screening tests, it has rarely been at the centre of the investigation. Yet, the decision to undergo amniocentesis has risks and may therefore be distressing, emotionally charged (Sarkar et al., 2006) and needs to be undertaken in a short timeframe.
Deciding about amniocentesis should be determined by the expectant parents’ awareness of the purposes of the test, their attitudes to the risks involved and how possible harms and benefits are valued and evaluated. Difficult decisions such as these should involve patients in choosing the option that is consistent with their knowledge, values and long-term goals. In addition, the decision to undertake amniocentesis is especially relevant in infertile populations. Pregnant women who have had in vitro fertilization are twice as likely to be offered amniocentesis after prenatal screening tests (Wald et al., 1999; Benn et al., 2006). This decision therefore affects a significant proportion of the couples who undergo infertility treatments.

To achieve these goals, technologies that support individuals when they face difficult decisions for themselves or others in their families are being developed. Decision support technologies (DSTs), also known as decision aids (O’Connor et al., 2003) provide information about available options and their associated harms, benefits and outcomes in a variety of clinical contexts. The number of published DSTs has tripled since 1999 (O’Connor et al., 2007). DSTs are intended to help individuals learn about the features, issues and implications of their treatment or screening options while improving communication and open discussion with their health professionals.

Most of these interventions are developed in a practical manner and use a wide range of media. DSTs have been noted to achieve different levels of clinical effectiveness and to impact on several decision outcomes (Entwistle et al., 1998). According to the Cochrane systematic review for people facing health treatment or screening decisions (O’Connor et al., 2003), DSTs that have been evaluated in trials improve knowledge as well as the agreement between personal values and the choice made, increase active participation in decision making and decrease decisional conflict.

To date, DSTs for amniocentesis have not been formally reviewed nor evaluated. In the literature, attention has been primarily focused on the decision to undertake screening tests rather than on the decision to undergo invasive diagnostic tests such as amniocentesis (Van den Berg et al., 2005; Weisz and Rodeck, 2006; Garcia et al., 2008). Furthermore, reviews had shown that there was limited research on how best to provide information on amniocentesis testing (Green et al., 2004; Hunt et al., 2005; Van den Berg et al., 2006). The aim of this study was to identify, describe and assess the quality and effectiveness of DSTs for amniocentesis and examine to what extent they were used in clinical practice.

To meet our aims, the review was organized around three questions:

(i) How many DSTs for amniocentesis exist and what are their aims?
(ii) Do DSTs for amniocentesis meet published quality standards?
(iii) What is the effectiveness of DSTs for amniocentesis?

Materials and Methods

Definitions

For the purpose of the review, DSTs were defined as: ‘Interventions designed to help people make specific and deliberative choices among options by providing information on the options and outcomes relevant to a person’s health status’ (O’Connor et al., 2003). Further, four essential criteria allowing the distinction between information leaflets and DSTs were identified in the Cochrane review (O’Connor et al., 2003) and in the domains and items of the International Patient Decision Aids Standards Instrument (IPDASi) (Elwyn et al., 2006). To be classified as a DST, an intervention should:

(i) state the decision to be addressed and deliberated upon,
(ii) provide information about the options, their harms, benefits and the associated probabilities of the decision outcomes,
(iii) allow patients to express and clarify their values, attitudes, preferences with regard to the decision,
(iv) provide structured guidance in coming to a decision (step-by-step way to make a decision).

Two out of the four criteria (criteria 1 and 2) were used in selecting the DSTs for review. Accordingly, interventions that stated that to have amniocentesis was the key decision to be considered and provided information about the harms, benefits and outcomes probabilities associated with each option were included in the review.

Literature search strategy

Ten electronic databases were searched until January 2008: medline (1966–2008), Medline In-Process (January 2008), PubMed (2008), Embase (1980–2008), British Nursing Index (1994–December 2007), CINAHL (1982–December 2007), all EBM Reviews (2008), PsycINFO (1806–2008), Science Citation Index Expanded (1970–2008) and Social Sciences Citation Index (1970–2008). A list of key words and subject headings (MeSH words in PubMed) was written in Ovid in order to be combined and run in each database (see Table I). DSTs for amniocentesis where reports have not been published in peer-reviewed journals or have not been evaluated in a trial were identified through manual check of reference lists from published papers, internet search and manual check of the A–Z list of decision aids developed by the Ottawa Health Decision Centre (http://decisionaid.ohri.ca/AZinvent.php). All major DST developers such as the Ottawa Health Decision Centre, Healthwise, Mayo Clinic, Midwives Information and Resource Service (MIDIRS), Foundation for Informed Medical Decision Making (FIMDM) and Intelihealth (Harvard Medical School) were contacted.

Table I. Literature search strategy.

<table>
<thead>
<tr>
<th>Amniocentesis</th>
<th>DST</th>
</tr>
</thead>
<tbody>
<tr>
<td>Amniocentesis</td>
<td>Decision support technique</td>
</tr>
<tr>
<td>Prenatal diagnosis</td>
<td>Patient decision aid</td>
</tr>
<tr>
<td>Antenatal diagnosis</td>
<td>Decision aid</td>
</tr>
<tr>
<td>Prenatal testing</td>
<td>Decision explorer</td>
</tr>
<tr>
<td>Antenatal testing</td>
<td>Decision tool</td>
</tr>
<tr>
<td></td>
<td>Decision support</td>
</tr>
<tr>
<td></td>
<td>Decision</td>
</tr>
<tr>
<td></td>
<td>Decision making</td>
</tr>
<tr>
<td></td>
<td>Software</td>
</tr>
<tr>
<td></td>
<td>Decision support systems</td>
</tr>
<tr>
<td></td>
<td>Computer assisted</td>
</tr>
<tr>
<td></td>
<td>Information systems</td>
</tr>
<tr>
<td></td>
<td>Computer assisted decision support systems</td>
</tr>
<tr>
<td></td>
<td>Genetic counselling</td>
</tr>
</tbody>
</table>

All terms in the first column were combined with terms in second column.
Study inclusion and exclusion criteria

First, studies were included if they (i) considered DSTs that focused on the decision to undertake amniocentesis (regardless of age and pathway of entry) or (ii) considered the decision to undertake amniocentesis as well as other prenatal screening tests or other available diagnostic tests such as chorionic villus sampling (CVS). CVS is a procedure to remove a small amount of tissue from the placenta to test the tissue in the laboratory and check for chromosomal abnormalities (Brun et al., 2003). CVS is generally conducted between 11 and 14 weeks of pregnancy and carries a 2% risk of miscarriage (Caughey et al., 2006).

Second, only interventions that were classified as DSTs, as opposed to information leaflets, were included in this review (as described, see Definitions section).

Studies were excluded if they (i) considered DSTs that exclusively focused on prenatal screening tests or CVS without addressing the decision to undertake amniocentesis and (ii) addressed a choice between amniocentesis and CVS (see Fig. 1).

DST assessment

After having identified interventions that met our inclusion criteria, we contacted the DST developers to obtain a copy of the DST and information on its current use and implementation in clinical settings. Information about the three aspects of DSTs was collected in order to address our research questions.

First, a content analysis of the DSTs was carried out using the IPDASi in order to quantify the quality of DSTs components for amniocentesis. The IPDASi (www.ipdasi.org) is currently being validated (Elwyn et al., 2008) by an international group of researchers working to assess the quality of DSTs (Elwyn et al., 2006). The author was trained to perform IPDASi ratings before assessment. IPDASi is a set of 48 quality criteria (or items) addressing 10 domains: information, test, probabilities, values, guidance, development, evidence, disclosure, plain language and evaluation. The ‘information’ domain assesses the quality of information provided on the index decision, the options available and the positive and negative features of each option (eight items). The ‘test’ domain assesses the extent to which specific features of the diagnostic or screening investigation (e.g. rate of false positive or false negative test results) are described in the DST (nine items). The ‘probabilities’ domain evaluates the way in which probabilistic information is presented and framed (eight items). The ‘values’ domain assesses whether the DST facilitates the expression and clarification of the expectant parents values in regards to the decision (five items). The ‘guidance’ domain investigates whether the DST provides structured guidance in helping expectant parents come to a decision (two items). The ‘development’ domain appraises the quality of the DST’s development process which should ideally involve professional/parents needs assessment, expert review and field-testing (six items). Field testing consists of showing the DST to potential users (expectant parents) who comment on its content and usability in order to amend the DST accordingly. The ‘evidence’ domain assesses the quality of the research evidence used in developing the DST (five items). The ‘disclosure’ domain appraises the transparency of the funding and author disclosure (two items). The ‘plain language’ domain assesses the DST clarity and readability levels (one item). Finally, the ‘evaluation’ domain assesses the impact of the DST on decision outcomes (one item). Each item was rated on a scale from 1 = strongly disagree to 4 = strongly agree. Domain and total IPDASi percentage scores were calculated by summing relevant items and dividing by the number of items per domain, which accounted for the unequal number of items per domain.

It has been found that the majority of DSTs are developed without theoretical foundations (Durand et al., 2008). We therefore examined the use of theory in the development of DSTs for amniocentesis by reviewing the relevant publications and by contacting the DST developers to determine whether theory had been used in the development

Figure 1: Decision support technology search of 10 databases.
process. The efficacy of the DSTs was determined by assessing evaluation methods and outcomes. Finally, we examined the current use and implementation of the DSTs in clinical settings.

Results

Selection of DSTs

The literature search and contact with authors identified 11 interventions (Fig. 1). After assessment of their content and/or related available publications, five interventions were excluded. Three DSTs did not focus on the decision to undertake amniocentesis. The DST by Heckerling et al. was excluded because it compares amniocentesis testing and CVS (Heckerling et al., 1994, 1999; Verps and Heckerling, 1995). The DSTs by Harris et al. (2001) and Kuppermann et al. (2004) were excluded because they focus on prenatal screening tests without specifically addressing the decision to undertake amniocentesis. Two interventions failed to be classified as DSTs (did not meet criteria 1 and 2 of Definitions section) and were identified as information leaflets after content analysis. These two are the web-based intervention produced by InteliHealth (2005) and the information leaflet produced by MIDS: ‘Is my baby alright?’ (MIDS, 2005).

How many DSTs for amniocentesis exist and what are their aims?

Six DSTs for amniocentesis were examined in the review: (i) A Decision Analytic Consultation by Bekker et al. (2004), (ii) a DST developed by Drake et al. combining an audiotape and a booklet entitled Making Choices: Prenatal Testing (Drake et al., 1999; Hunter et al., 2005), (iii) The Amniocentesis Report, a booklet downloaded from the internet produced by Ferber and Sicherman (2001), (iv) a web-based DST for amniocentesis developed by the Healthwise group entitled Should I have an amniocentesis? (Healthwise, 2006), (v) a DST for prenatal testing developed by Nagle et al. (2008) and (vi) a decision analytic model developed by Pauker and Pauker (1979, 1987). Of the six DSTs identified, three were developed in the USA (Pauker and Pauker, 1979; Ferber and Sicherman, 2001; Healthwise, 2006), one in Canada (Drake et al., 1999), one in Australia (Nagle et al., 2008) and one in the UK (Bekker et al., 2004). Two out of the six interventions were available on internet (Ferber and Sicherman, 2001; Healthwise, 2006), although one of the DSTs’ availability was subjected to payment (Ferber and Sicherman, 2001). On the basis of the Ottawa A–Z inventory and contact with authors, it was ascertained that two of the six DSTs were ordered as a hard copy. The DST is divided into six sections: (i) What is amniocentesis and how is it done, (ii) Nature and accuracy of the amniocentesis results, (iii) What are the benefits of amniocentesis, (iv) What are the costs of amniocentesis (relevant to the USA), (v) Making the amniocentesis decision and (vi) Alternative procedures. The DST does not explicitly enable pregnant women/couples to clarify their values with regard to the decision.

The web-based DST produced by Healthwise is an interactive website entitled Should I have An Amniocentesis. The DST intends to help expectant parents understand their choices regarding amniocentesis testing. The DST includes four sections (introduction, medical information, your information and wise health decision) and is equivalent to seven printed pages. The third section ‘your information’ compares the reasons to have or to decline amniocentesis, whereas the last section ‘a wise health decision’ provides a worksheet for patients to clarify their ideas and values with regard to the amniocentesis decision.
<table>
<thead>
<tr>
<th>Authors</th>
<th>Name of DST</th>
<th>Decision</th>
<th>Format, use and access</th>
<th>Theoretical framework</th>
<th>Outcome measures</th>
</tr>
</thead>
<tbody>
<tr>
<td>Bekker et al. (2004)</td>
<td>Decision analysis consultation</td>
<td>Prenatal diagnostic test (amniocentesis and CVS)</td>
<td>Routine consultation structured by decision analysis Method used in an additional information consultation offered to women after they received a screen-positive maternal serum screening test result for Down syndrome DST developed in the UK</td>
<td>Decision analysis</td>
<td>Consultation length Informed decision-making</td>
</tr>
<tr>
<td>Drake et al. (1999)</td>
<td>Making choices</td>
<td>Prenatal testing</td>
<td>Workbook and audiotape DST developed in the UK DST used is a genetic counselling consultation and includes a discussion with a genetic counsellor DST developed in Canada</td>
<td>Ottawa decision support framework</td>
<td>Test choice Knowledge Anxiety</td>
</tr>
<tr>
<td>Hunter et al. (2005)</td>
<td>Prenatal testing</td>
<td>–Maternal serum screening</td>
<td></td>
<td></td>
<td>Decisional conflict Intervention satisfaction</td>
</tr>
<tr>
<td></td>
<td></td>
<td>–Amniocentesis</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td>–Chorionic villus sampling</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Ferber and Sicherman (2001)</td>
<td>The amniocentesis report</td>
<td>Amniocentesis Testing</td>
<td>Web-based DST <a href="http://www.amniocentesis.org">www.amniocentesis.org</a> DST can be downloaded in PDF format online or shipped worldwide Minimal fee: $5.05 DST developed in Canada</td>
<td>No theory</td>
<td>No evaluation</td>
</tr>
<tr>
<td>Healthwise (2006, last update)</td>
<td>Should I have an amniocentesis?</td>
<td>Amniocentesis testing</td>
<td>Web-based DST <a href="http://www.webmd.com/baby/should-i-have-an-amniocentesis">www.webmd.com/baby/should-i-have-an-amniocentesis</a> Open Access (free of charge) DST developed in the USA</td>
<td>No theory</td>
<td>No evaluation</td>
</tr>
<tr>
<td>Nagle et al. (2006)</td>
<td>A Decision aid for Prenatal Testing of fetal abnormalities</td>
<td>Prenatal testing:</td>
<td>24-page booklet DST given to women in early pregnancy by their GP DST developed in Australia</td>
<td>Ottawa decision support framework</td>
<td>Informed choice Decisional conflict Anxiety Depression Attitudes to the fetus/pregnancy Satisfaction with the DST Assessed cost of elective miscarriage</td>
</tr>
<tr>
<td>Pauker and Pauker (1979)</td>
<td>A decision analytic model to counsel patient about amniocentesis</td>
<td>Amniocentesis Testing</td>
<td>Routine consultation structured by decision analysis Method used in a routine genetic counselling session for prenatal diagnosis DST developed in the USA</td>
<td>Decision analysis</td>
<td>Assessed cost of spontaneous miscarriage Actual decision Decision suggested by decision analytic model</td>
</tr>
</tbody>
</table>
The DST for prenatal testing by Nagle et al. (2008) is a 24-page booklet containing varied graphical design elements (diagram, images, charts and dot points) and information about maternal serum screening, second-trimester ultrasound scan, CVS and amniocentesis testing. The DST aims to assist women in making an informed choice about amniocentesis and reduce decisional conflict. It provides information on the reasons for being offered testing, the range of prenatal tests available and the results and implications of such tests. The DST contains scenarios of women’s experience when deciding about testing, a worksheet to weigh up the pros and cons of each option and a list of further information resources available. Finally, it includes a risk-report sheet presenting the risk estimates of having a baby affected with Down’s syndrome, based on the expectant mother’s age and gestation.

The decision analytic DST developed by Pauker and Pauker (1979) is used to counsel patients about amniocentesis testing. The DST was designed to help parents assess their values and attitudes about the outcomes of options and make a logical decision (guided by decision analysis) about amniocentesis. The decision analytic DST uses a lottery technique and considers the following outcomes: miscarriage, detection of chromosomal abnormality and being faced with diagnostic errors. Prospective parents are asked to assign a utility (on a scale from 0 to 100) to the potential outcomes of both available options: undertaking or declining amniocentesis.

Do DSTs for amniocentesis meet published quality standards?

IPDAS evaluation criteria

Five out of the six DSTs were rated against all IPDAS domains to assess their quality (Table III). The evaluation of one DST (Nagle et al., 2008) was exclusively based on the publications since the developers declined to provide a copy of the intervention.

The provision of different types of information was variable across domains. Scores on the ‘information’ domain reached 73.7% on average. Most DSTs for amniocentesis showed very little variation regarding the provision of information. Scores on the ‘test’ domain (evaluating the specific features of a diagnostic test) were on average lower (61.6%) than scores on the information domain. Three DSTs (Ferber and Sicherman, 2001; Bekker et al., 2004; Healthwise, 2006) did not include full information on the false positive and false negative results associated with the tests and scored significantly lower than other DSTs evaluated. Scores on the ‘probabilities’ domain reached 55% on average. Most DSTs provided very little information about the outcome probabilities associated with the options. Scores on the ‘plain language’ domain reached 40% on average. Most DSTs did not use plain language throughout and did not report readability levels.

There were also IPDASi score differences in domains clarifying factors and ways of making the decision. The average score on the ‘values’ domain was 53%. Three interventions only (Drake et al., 1999; Bekker et al., 2004; Healthwise, 2006) explicitly enabled parents to express and clarify their values. In contrast, average scores on the ‘guidance’ domain reached 80%. Three DSTs (Drake et al., 1999; Bekker et al., 2004; Healthwise, 2006) reached the highest score by providing ‘a step-by-step way to

Table III. DSTs rated on the International Patient Decisions Aids Standards (IPDAS) instrument.

<table>
<thead>
<tr>
<th>Name of IPDAS Domains</th>
<th>Name of DST</th>
<th>Average scores per domain</th>
</tr>
</thead>
<tbody>
<tr>
<td>Information</td>
<td>Test</td>
<td>68.7a</td>
</tr>
<tr>
<td></td>
<td>Probabilities</td>
<td>55.6</td>
</tr>
<tr>
<td></td>
<td>Values</td>
<td>55.0</td>
</tr>
<tr>
<td></td>
<td>Guidance</td>
<td>55.0</td>
</tr>
<tr>
<td></td>
<td>Development</td>
<td>55.0</td>
</tr>
<tr>
<td></td>
<td>Evidence</td>
<td>55.0</td>
</tr>
<tr>
<td></td>
<td>Disclosure</td>
<td>55.0</td>
</tr>
<tr>
<td></td>
<td>Plain Language</td>
<td>55.0</td>
</tr>
<tr>
<td></td>
<td>Evaluation</td>
<td>55.0</td>
</tr>
<tr>
<td></td>
<td>Average score per DST</td>
<td>70.8</td>
</tr>
</tbody>
</table>

Scores on the IPDAS instrument in percents.
make a decision’, and worksheets designed to structure the expectant parents’ decision-making process.

IPDASi scores concerning the process of DST development and evaluation were in the mid to higher range. On the ‘development’ domain reached 60% on average. The development process generally lacked rigour and rarely involved expectant parents. According to the IPDAS standards, the development process should ideally involve a needs assessment with expectant parents and professionals, the parents and professionals review of the DST and field testing before evaluation. Most DSTs were based on literature reviews, reviewed by researchers or occasionally health professionals and omitted expectant parents’ needs assessment or review. Finally, only two out of the six DSTs were explicitly field tested with patients (Drake et al., 1999; Nagle et al., 2008). Scores on the ‘evidence’ domain (assessing the scientific validity of the DST) reached 66% on average. The development of three DSTs (Drake et al., 1999; Bekker et al., 2004; Healthwise, 2006) was explicitly based on research evidence (i.e. citations to studies used). Scores on the ‘evaluation’ domain reached 60% on average. Four DSTs (Pauker and Pauker, 1987; Drake et al., 1999; Bekker et al., 2004; Nagle et al., 2008) were evaluated in a trial, including the DST by Nagle et al. (2008). Two DSTs out of the three (Drake et al., 1999; Bekker et al., 2004) were reported to help patients make a decision that was ideally consistent with their values and preferences. Finally, most DSTs acknowledged sponsors and contributors. The average score on the ‘disclosure’ domain was 72.5%. All DSTs reached high scores on this domain by displaying funding information and credentials of those developing the DST.

Theoretical foundation

Four out of the six interventions mentioned the contribution of a theoretical framework in developing the DST (Pauker and Pauker, 1987; Drake et al., 1999; Bekker et al., 2004; Nagle et al., 2008). Decision analysis (expected utility theory) guided the design and use of two DSTs for amniocentesis (Pauker and Pauker, 1979; Bekker et al., 2004). Two DSTs (Drake et al., 1999; Nagle et al., 2008) relied on the Ottawa decision support framework (O’Connor et al., 1999). The Ottawa decision support framework combines social support and cognitive psychology theories such as the expectancy value model (Fishbein, 1975), prospect theory (Kahneman and Tversky, 1979), the conflict theory model of decision-making (Janis and Mann, 1977) and the theory of reasoned action (Fishbein, 1975). There is as yet no evidence about the influence that theory-based design has on outcomes. However, two out of the three ‘theory based’ DSTs (Drake et al., 1999; Bekker et al., 2004) have higher IPDASi scores (70.8 and 70.5) than DSTs that do not rely on theory.

What is the effectiveness of existing DSTs?

Three out of the six DSTs were evaluated using randomized controlled trials (Bekker et al., 2004; Hunter et al., 2005; Nagle et al., 2006). One DST was evaluated in a pilot study (Pauker and Pauker, 1979).

The decision analysis DST by Bekker et al. was evaluated in a randomized controlled trial of 117 women offered amniocentesis testing and randomized to a routine consultation or a decision analysis consultation. The risk perception of the screening test result, subjective utility (generated by the decision analytic method), knowledge, consultation quality, decisional conflict and anxiety were measured. The intervention reduced decisional conflict, improved informed decision-making and led to a more realistic evaluation of information. However, the DST was unable to predict the final decision: the decision suggested by the decision analysis was inconsistent with the decision that parents finally made. The DST did not significantly impact on consultation satisfaction, knowledge nor reduced anxiety when compared with the control group. The latter finding is consistent with the results of similar evaluations (O’Connor et al., 2003).

The DST by Drake et al. (Drake et al., 1999; Hunter et al., 2005) was evaluated in a randomized controlled trial of three counselling methods for prenatal diagnostic (Hunter et al., 2005) and one pre-to-post study (Drake et al., 1999). First, a randomized controlled trial of three counselling methods for prenatal diagnosis (individual counselling, group counselling and use of a DST by Drake et al.) assessed the following outcomes measures: knowledge, state-trait anxiety, decisional conflict and intervention satisfaction in a sample of 350 women (and 225 partners) who had been offered prenatal diagnostic testing because of advanced maternal age (Hunter et al., 2005). Compared with other counselling methods (group and individual counselling), the DST was least efficient at improving prospective parents knowledge (although knowledge increased compared with the pre-counselling phase). The DST did not significantly diminish state anxiety. However, decisional conflict was significantly reduced. The satisfaction with the DST was high but not higher than alternative counselling methods (i.e. group counselling). Second, the pre-to-post study (Drake et al., 1999) evaluated the DST’s impact on knowledge, decisional conflict, state-trait anxiety and intervention acceptability in a sample of 21 women (and 17 partners). The DST was reported to have significantly increased knowledge and decreased decisional conflict but did not modify state anxiety level. However, the findings were not compared with a control group.

The DST by Nagle et al., which was not available for IPDASi rating, was evaluated in a cluster randomized controlled trial where 55 general practitioners were randomized to provide women (n = 338) with the DST (intervention group) or a pamphlet (control group). The following outcome measures were assessed: informed choice and decisional conflict (primary outcome measures), anxiety, depression, attitudes to the fetus/pregnancy and satisfaction with the DST or pamphlet. The results showed that more women made an informed choice when given the DST than when given the pamphlet. The satisfaction with the intervention was significantly higher in the intervention group (use of DST). The level of decisional conflict was low in both intervention and control groups. There were no significant differences on the secondary outcomes: anxiety, depression or attitudes to the pregnancy/fetus.

The decision analysis DST by Pauker et al. was evaluated with 90 women (35 couples) who were offer amniocentesis. The evaluation consisted of comparing women’s (or couples) attitudes on miscarriage and termination of pregnancy. Most couples made a decision that was consistent with their attitudes towards miscarriage and elective pregnancy termination. However, choices made by some couples conflicted with their stated values. The findings showed that the actual choice bore no relation to the
choice suggested by the decision analysis. The reliability of the findings could be questioned by the absence of a control group. IPDASi scores reached a total average score of 58.1%.

Finally, two DSTs (Ferber and Sucherman, 2001; Healthwise, 2006) were not evaluated but low scores on IPDASi suggest domains that could be improved (Table III). The amniocentesis report produced by Ferber et al. scored the lowest on IPDASi (total adjusted score: 44.8%). In brief, the evaluation of four out of the six DSTs was of variable quality and did not result in significant changes in the selected decision outcomes (knowledge, anxiety and decisional conflict).

Discussion

Six DSTs for amniocentesis were identified; their quality was variable and they did not score highly against recently developed standards. The evaluations conducted on the DSTs had considerable scope for improvement. This reflects the emergent nature of this field and highlights the need for a DST capable of addressing parents’ information and decision support needs.

Existing DSTs for amniocentesis are a small proportion of the total number of DSTs (over 500) developed in other healthcare contexts worldwide (O’Connor et al., 2007), and there may be several reasons to account for the scarcity of DSTs in this medical context. First, deciding about amniocentesis might not be perceived as generating as much anxiety and decisional conflict as other screening or treatment decisions surrounding directly life-threatening conditions. However, there is much evidence that amniocentesis generates peak levels of anxiety, at a time of particular emotional vulnerability (Robinson et al., 1984; Sarkar et al., 2006), and that such maternal stress could be associated with poor outcomes (i.e. gestational complication, fetal growth retardation) for the mother and fetus (Reading, 1983; Nakamura et al., 2008). Second, the difficulty to assess the decision-making process surrounding amniocentesis testing may account for the small proportion of amniocentesis DSTs available. Only 1–5% of women who undertake prenatal screening tests will be offered an amniocentesis (Benn et al., 2006; Gidiri et al., 2007). The number of women concerned with this decision is therefore few and combined with high levels of stress and anxiety, it is difficult to approach women at the early stage of the decision-making process in order to gain the necessary data to inform DST development. Finally, there is much variation in patient preference for sole or shared decision-making (Schneider et al., 2006) and a lack of DSTs in this highly emotive context. This may reflect the fact that some patients prefer to hand over decision-making to their physicians even though they ultimately must make the decision.

DSTs for amniocentesis are generally not used in routine clinical practice. Interventions that are primarily developed by academics (lack of field testing and needs assessment) might be unable to meet the practical requirements and decision support needs of patients and professionals who are expected to use it and would justify the consistent difficulty to implement DSTs into practice (Holmes-Rovner et al., 2000; Silvia et al., 2008). These results point to the need to develop DSTs that have better goodness-of-fit with the clinical situations they are meant to support.

The IPDASi evaluation emphasized the variable quality across DSTs and domains. Most DSTs are effective and reliable information resources because they provide adequate information on the amniocentesis decision, on the features of a diagnostic test, guide expectant parents in making a decision and use scientific evidence. However, the communication of outcome probabilities, the expression and clarification of values, the development process, the evaluation and the use of plain language could be significantly improved. Lower scores on those domains may reflect the complexity and specialization of the domains’ requirements combined with the recent development, inexperience and implementation difficulties in the field of shared decision making (and DSTs development). Our analysis subsequently revealed that most DSTs had a theoretical origin, a finding inconsistent with existing DST reviews (Bekker et al., 1999; Bowen et al., 2006; Durand et al., 2008).

The DSTs evaluations in trials of varying size and method revealed poor quality experiments and pointed to the difficulty to assess the DSTs’ effectiveness. The match between clearly stated goals and the results of the evaluation was poor. In majority, DSTs were shown to facilitate information processing but failed to reduce emotional burdens associated with the amniocentesis decision (e.g. anxiety) and could not predict actual decisions. This may reflect a growing tendency to develop DSTs in short timeframes and promote their use on internet without rigorously evaluating their impact on decision-making outcomes. This raises concerns as to the use of poor quality DSTs by expectant parents who are expected to make high-stake decisions at a time of considerable emotional upheaval.

The present study is the first to have reviewed DSTs for amniocentesis. However, systematic reviews conducted in other healthcare contexts (Williams et al., 2002; Evans et al., 2005; Volk et al., 2007) corroborate our findings. Systematic reviews of prostate cancer screening (Evans et al., 2005; Volk et al., 2007) revealed that DSTs increased knowledge but did not impact on other decision outcomes nor reduce emotional burdens. A systematic review of interactive decision aids for breast cancer genetic testing (Williams et al., 2002) supports the present findings by identifying a very small number of poor quality interventions rarely evaluated and available to users. The IPDASi scores of DSTs for breast cancer genetic testing reached lowest scores on the same domains as DSTs for amniocentesis (communication of outcome probabilities, value expression and clarification, development process and evaluation).

Strengths of the study were the comprehensive search and the multidimensional analyses performed. Limitations need to be considered in interpreting the findings. All the evaluations, including the IPDASi rating, were undertaken by a single researcher (M.A.D.). However, decisions, especially those where uncertainty arose, were discussed with co-authors to arrive at agreed resolutions.

To conclude, few DSTs for amniocentesis are implemented in practice and do not seem to provide sufficient decision support for people facing this difficult decision (Hunt et al., 2005). There is a need for further improvement of existing interventions. The next generation of DST for amniocentesis should be guided by the IPDAS criteria, using patient and professional involvement as a central component to the DST development and rigorous field test and evaluation should be conducted (Evans et al., 2007). Compared with other healthcare contexts (breast cancer, heart disease), little attention has been given to the decision to undertake
amniocentesis, or other difficult decisions surrounding the area of reproduction and infertility. Whether deciding about prenatal testing or infertility treatments, the complexity of the information provided, the high stakes of the decision, emotional strain and high anxiety levels associated makes it difficult and distressing for couples or expectant parents to engage in a decision-making process. This emphasizes the need for high-quality DSTs.

Funding

This study was funded by the Sir Halley Stewart Trust, Willingham, Cambridge, UK.

References


Healthwise. Should I have an amniocentesis. Internet site. 2006.


Submitted on April 10, 2008; resubmitted on July 15, 2008; accepted on July 24, 2008.