RAPID PRENATAL DIAGNOSIS TESTING FOR DOWN SYNDROME
ONLY OR LONGER WAIT FOR FULL KARYOTYPE: THE VIEWS
OF PREGNANT WOMEN

Background

Prenatal diagnosis by invasive testing is currently offered to all women at increased risk of fetal chromosome abnormality. Women currently wait on average 13 days, and sometimes up to 21 days, for the results from prenatal tests for Down syndrome (NEQAS, 2001). This ‘comprehensive test’ analyses the entire chromosome compliment of the fetus, also allowing detection of gender and any chromosomal abnormalities. The level of stress experienced by prospective parents upon receipt of a screen positive result and during the subsequent wait for results of further tests has been well documented (Cederholm et al., 1999).

Current developments allow rapid detection of the presence of Down syndrome, reporting results within two days of sampling. This ‘simple test’ does not identify the gender of the fetus nor other rare chromosomal conditions that may be present.

In recent years some centres have offered rapid prenatal diagnosis alongside full karyotype analysis (Tepperberg et al 2001; Levett et al 2001). The Scottish Executive now funds this additional test for “at risk” pregnancies in Scotland. With over 30,000 amniocenteses performed in the UK annually, this policy has cost implications. As the health service faces limited resources and as the rapid testing has been proven to reliably detect the most common abnormalities, there is now a debate about whether rapid diagnosis should be used as the sole diagnostic tool for certain referral groups (e.g. those at risk of Down syndrome on biochemical screening) (UK Department of Health, 2003).

This study used a discrete choice experiment (DCE) to help address the question. The DCE was used to estimate the trade-offs individuals were willing to make between waiting time and information. Willingness to pay (WTP), a monetary measure of benefit, was also estimated for ‘simple and quick’ versus ‘comprehensive and longer’ prenatal diagnosis programmes. For more information on this study see Ryan et al (2005).
Subjects and methods

50 pregnant women attending Aberdeen Maternity Hospital were asked to take part in the study. Women were randomly approached as they attended for booking, and nobody refused to complete the questionnaire.

The policy question identified the attributes to be included in the experiment (Table 1). Respondents were advised that if they received ‘comprehensive’ information they would be told whether or not the fetus has Down syndrome, gender and whether other rare chromosomal conditions were present. In contrast, ‘simple’ information would only inform them of whether the fetus had Down syndrome. Respondents were informed that ‘cost’ was included in the experiment as a method to value the alternative screening programmes in monetary terms (in terms of willingness to pay).

<table>
<thead>
<tr>
<th>Attributes</th>
<th>Levels</th>
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</thead>
<tbody>
<tr>
<td>Level of Information</td>
<td>Comprehensive, Simple</td>
</tr>
<tr>
<td>Number of days wait for results</td>
<td>2, 8, 16, 30</td>
</tr>
<tr>
<td>Cost to you</td>
<td>20, 100, 250, 500</td>
</tr>
</tbody>
</table>

Table 1: Attributes and levels in the DCE

From the attributes and levels defined in Table 1 there were 32 possible screening programmes. Experimental design techniques were used to reduce this to 8 discrete choices. Each respondent was presented with the 8 choices and asked, for each, which screening programme they would choose. Respondents were also given the option of neither (implying they preferred not to be screened). An example of such a choice is shown in Figure 1.

Responses were analysed using conditional logit regression techniques, and the following benefit equation was estimated:

\[ B = \beta_1 \text{SIMPLE} + \beta_2 \text{COMP} + \beta_3 \text{WAIT} + \beta_4 \text{COST} \]  

(1)

where B is the benefit derived from a screening test as opposed to no screening, SIMPLE refers to having simple information rather than no information, COMP to having comprehensive information rather than no information, WAIT is the number of days waiting for results, and COST the cost of screening to the patient.

Coefficients \( \beta_1 \) to \( \beta_4 \) show the relative importance of the attributes on choice. When interpreting these coefficients it is important to be aware of the unit of measurement: \( \beta_1 \) and \( \beta_2 \) show the change in benefit in moving from not being screened to having simple information (\( \beta_1 \)) or comprehensive information (\( \beta_2 \)). Therefore, \( \beta_2 - \beta_1 \) shows the value of comprehensive information over simple information. \( \beta_3 \) indicates the value of a one day reduction in waiting time for results.

The ratio of any two coefficients shows how individuals trade between the different attributes. The denominator is what the value can be estimated in terms of. For example, if we want value in terms of waiting time, \( \beta_2 / \beta_3 \) indicates willingness to wait (WTW) for ‘simple’ information (rather than have no information) and \( \beta_2 / \beta_3 \) WTW for ‘comprehensive’ information (rather than have no information). From this it is possible to estimate how much longer respondents are willing to wait to have comprehensive rather than simple information: \( (\beta_2 - \beta_1) / \beta_3 \).

When cost is included as the denominator willingness to pay (WTP), a monetary measure of benefit, can be estimated for different screening programmes. For example, assuming everything else equal, respondents are willing to pay \( \beta_1 / \beta_4 \) for ‘simple’ information and \( \beta_2 / \beta_4 \) for ‘comprehensive’ information. Following this, the marginal WTP for comprehensive rather than simple, assuming everything else is equal, is \( (\beta_2 - \beta_1) / \beta_4 \) and the WTP for a day’s reduction in waiting time is \( \beta_3 / \beta_4 \).

From this, WTP for the ‘simple’ test with results available in 2 days (compared to not being screened) can be estimated as:

\[ \text{WTP}_{\text{simple}} = (\beta_1 - (\beta_3 \times 2)) / \beta_4 \]  

(2)

and WTP for the ‘comprehensive’ information with results available in 21 days (compared to not being screened) can be estimated as:

\[ \text{WTP}_{\text{comprehensive}} = (\beta_2 - (\beta_3 \times 21)) / \beta_4 \]  

(3)
Forty-nine questionnaires were returned. See Ryan et al (2005) for information on characteristics of respondents. Six respondents did not complete the choices, resulting in 43 usable DCE responses. Table 2 presents the regression results. The negative and significant signs on both ‘number of days to wait for result’ and ‘cost’ show respondents prefer both these attributes to be lower. The positive signs of ‘simple’ information and ‘comprehensive’ information indicate that respondents prefer having information to not having information. Further, given that the coefficient on comprehensive information is greater than that on simple, then, everything else equal, respondents prefer ‘comprehensive’ information to ‘simple’ information. These results all conform to a priori expectations, providing support for the theoretical validity of the model.

<table>
<thead>
<tr>
<th>Attribute</th>
<th>Coefficient</th>
<th>WTP(^1) (£)</th>
<th>WTW(^1) (days)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Simple</td>
<td>1.966</td>
<td>690.54</td>
<td>38.28</td>
</tr>
<tr>
<td>Comp</td>
<td>2.252</td>
<td>791.01</td>
<td>43.85</td>
</tr>
<tr>
<td>Waiting Time</td>
<td>-0.051</td>
<td>18.04</td>
<td></td>
</tr>
<tr>
<td>Cost</td>
<td>-0.003</td>
<td></td>
<td></td>
</tr>
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Table 2: Regression Results

Assuming equal waiting time respondents were willing to pay £691 for simple information and £791 for comprehensive information. This implies that the value of the additional information is £100 (£791 - £691), suggesting that the greatest value in the information received is entailed in the results concerning Down syndrome.

Whilst respondents prefer comprehensive to simple information, they also prefer lower waiting times and were willing to pay £18 (0.07 / .0016) to reduce waiting time by one day.

From equation (2) above, WTP for a test providing simple information within 2 days is

\[
WTP_{\text{simple}} = \frac{1.965688 - 0.0513463(2)}{0.0028466} = £654.46
\]

and, from equation (3) above, WTP for a test which provides ‘comprehensive’ information in 21 days is

\[
WTP_{\text{comp}} = \frac{2.251685 - 0.0513163(21)}{0.0028466} = £412.22
\]

That is, whilst respondents were willing to pay more for comprehensive information, given the value of reducing waiting time, they gain more benefit from a simple test delivering results in 2 days than a comprehensive test delivering results in 21 days.

**Results**

Forty-nine questionnaires were returned. See Ryan et al (2005) for information on characteristics of respondents. Six respondents did not complete the choices, resulting in 43 usable DCE responses. Table 2 presents the regression results. The negative and significant signs on both ‘number of days to wait for result’ and ‘cost’ show respondents prefer both these attributes to be lower. The positive signs of ‘simple’ information and ‘comprehensive’ information indicate that respondents prefer having information to not having information. Further, given that the coefficient on comprehensive information is greater than that on simple, then, everything else equal, respondents prefer ‘comprehensive’ information to ‘simple’ information. These results all conform to a priori expectations, providing support for the theoretical validity of the model.

<table>
<thead>
<tr>
<th>Level of Information</th>
<th>TEST A</th>
<th>TEST B</th>
<th>NEITHER</th>
</tr>
</thead>
<tbody>
<tr>
<td>No. of days to wait for result</td>
<td>16 days</td>
<td>2 days</td>
<td></td>
</tr>
<tr>
<td>Cost to you</td>
<td>£500</td>
<td>£250</td>
<td></td>
</tr>
</tbody>
</table>

Which test would you choose (tick one box only)
Discussion

Advances in the techniques for prenatal diagnosis have made it possible to rapidly detect specific chromosome abnormalities. However, these rapid tests provide less information. This raises important questions concerning whether the ‘simple’ information, ‘comprehensive’ information, or indeed both tests, should be provided. This study found that whilst pregnant women would prefer the ‘comprehensive’ information test, given the longer waiting times associated with such a test, the ‘simple’ information test would be preferred. Indeed, respondents would prefer the ‘simple’ information test as long as results are received 6 days sooner than with the comprehensive test. These results have implications for the resources dedicated to providing a rapid prenatal diagnostic service. More specifically, where there are limited resources, rapid testing may be sufficient to meet women’s expectations.

References


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