Analysis of problems in making the reproductive decision after genetic counselling

Petra G Frets, Hugo J Duivenvoorden, Frans Verhage, Bernadina M T Peters-Romeyn, Martinus F Niermeijer

Abstract
A follow up study of 164 couples to evaluate reproductive decision making two to three years after genetic counselling showed that 43% had problems making the reproductive decision. These couples (1) had experienced difficulty in the decision making process, (2) had doubts about the decision they had made, or (3) had been unable to make a decision. Using logistic regression analysis we identified the following factors as independently and significantly associated with problems in the decision making process: (1) no postcounselling relief, (2) anticipation of a high risk level, (3) relatives’ disapproval of decision, (4) a decision against having children, and (5) the presence of an affected child. Interestingly, of the couples that decided to have children, 45% of those who were eligible for prenatal diagnosis experienced the decision making process as difficult compared with 23% of those for whom prenatal diagnosis was not available (p<0.05). Problems in the decision making process may become apparent after genetic counselling rather than in the course of it. We suggest a structured follow up three to six months after genetic counselling to identify couples that would benefit from additional supportive counselling.

In 1975, the Ad Hoc Committee on Genetic Counselling of the American Society of Human Genetics determined that the aim of genetic counselling is to inform consultands about the nature of a mental or physical handicap in the family and its risk of occurrence or recurrence. Furthermore, the Committee advocated discussion of various options to prevent the birth of an affected child, such as refraining from having children, prenatal diagnosis and selective abortion, or alternative methods of conception. Genetic counselling should facilitate informed reproductive decision making, allowing for personal and social considerations.

Problems in the postcounselling reproductive decision making process can be divided into three categories: (1) experiencing the decision making process as particularly difficult, (2) unresolved doubt about a decision once taken, and (3) inability to make a decision. Factors related to difficulty with the decision making process after genetic counselling have been assessed only occasionally. The absence of a healthy child, the inability to share the responsibility for the decision with others, and the fear of not being able to cope with an affected child complicated the decision making process, while others found that the availability of prenatal diagnosis facilitated the decision making process.

Factors associated with persistent reproductive uncertainty after genetic counselling have been reported in several studies. Lubs et al.

The study reported here is part of a larger one concerning the decision making process after genetic counselling. Other aspects included analysis of single factors or a combination of factors influencing the reproductive decision. A model was developed capable of identifying the reproductive decision correctly in more than 90% of the cases.

In the present study, we investigated which factors were associated with postcounselling difficulty in making the reproductive decision, unresolved doubts after the decision had been made, or persistent reproductive uncertainty.

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Insight into the factors which are related to problems with the decision making process might indicate which couples would benefit from additional counselling.

Material and methods

STUDY POPULATION

The study population comprised 500 couples seen for genetic counselling at the Department of Clinical Genetics in 1984, most of whom had complex family histories of mental or physical handicap or both. This counselling entailed diagnosis, family history taking, estimation of the risk level, and discussion of various options to prevent the birth of an affected child.

Only those couples were entered into the study who had requested genetic counselling for their own offspring ($n=421$). Couples were excluded from the study if they had a history of genetic counselling at another clinical genetics centre ($n=63$), uncompleted genetic counselling ($n=30$), separation of the couple ($n=13$), insufficient command of Dutch ($n=5$), personal circumstances ($n=6$), and assessment in the pilot study to test the questionnaire or to train the research assistants ($n=13$).

Three levels of genetic risk were distinguished. Category I involved a genetic risk of less than 5%, category II entailed a genetic risk of 5 to 15%, and category III consisted of a genetic risk of higher than 15%. The lowest risk category was overrepresented in the whole population. Therefore, couples from this risk category were randomly selected to achieve equal representation of the three risk categories, thereby excluding 101 couples.

Of 190 couples eligible for the study, 16 refused to participate because they were not interested ($n=8$) or because the subject was too emotional ($n=8$). Five couples could not be traced and three couples were excluded because the husband would not participate. Two couples failed to understand the questions and were therefore excluded. Altogether, 164 couples were enrolled in the study. A detailed description of the educational level and religious affiliation of the study population has been reported elsewhere.9

PROCEDURE

Between 1986 and 1987, the couples under study were interviewed at home by a psychologist or one of three senior medical students trained for this purpose.

A questionnaire was constructed listing 91 items, partly multiple choice and partly open ended. The questionnaire enquired whether the couple had been worried in the months before genetic counselling, whether they expected their risk to be high or low and how they felt shortly after genetic counselling (relief, worry, disappointment, etc.). They were also asked about their recollection of the risk level given by the counsellor, indicated as the recalled risk. Risk recall was considered correct if both spouses gave a figure which fell into the correct category (I, II, or III).

Couples were also asked whether they had subsequently come to a reproductive decision or had remained undecided ($=\text{UNDEC}$). If they had made a decision they were asked whether they had experienced particular difficulty in making this decision or whether it had taken a great deal of deliberation ($=\text{DIFF}$). Whether the decision, once made by the couples, had left unresolved doubts was based on the answers to the following questions: (1) does your decision bother you, or (2) have you been wondering recently whether you made the right decision? Unresolved doubt ($=\text{DOU}$) was indicated when either of these questions was answered in the affirmative.

The questions about difficulty with the decision making process and subsequent doubtfulness had to be answered by yes or no. Difficulty with the decision making process or post decision doubt were considered to be present when the consultand had experienced these feelings more than once after genetic counselling. In two cases consultands gave unequivocal answers regarding unresolved doubts, requiring a senior psychologist’s judgement.

Table 1 is a schematic presentation of the variable outcome of the postcounselling decision making process in relation to problems experienced in making that decision. DIFF and DOU couples are classed separately because unresolved doubt about such an important decision was considered more serious than

<table>
<thead>
<tr>
<th>Genetic counselling ($n=155$)</th>
<th>Decision making process difficult ($n=65$)</th>
<th>Decision making process not difficult ($n=90$)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Undecided (UNDEC) ($n=18$) (12%)</td>
<td>Decided (DIFF*) ($n=47$) (30%)</td>
<td>Decided $n=90$</td>
</tr>
<tr>
<td>Doubtful (DOU) ($n=12$) (8%)</td>
<td>Not doubtful ($n=35$)</td>
<td>Not doubtful ($n=89$)</td>
</tr>
</tbody>
</table>

*The DIFF couples are those who had made a decision and had experienced the decision making process as difficult.
difficulty experienced in making a decision that leaves consultands at peace. Factors that contributed to both
difficulty in the decision making process and post-
decision doubt were not assessed separately.

Both husband and wife were required to participate
in the follow up study. In the case of disagreement
between spouses, the answer which might indicate
worries concerning the health of a future child was
selected. For example, if one spouse had not felt
relieved after counselling, this would become a
concern for the couple in their decision making
process. Thus the couple would be considered to have
felt no relief after genetic counselling even if one
spouse had felt relieved. Interpersonal differences
might also appear in respect to other factors relevant
to problems in the decision making process. Evalu-
ation of the influence of disagreement between spouses
on problems in making the reproductive decision will
be investigated at a later date.

DIFF couples were compared with those who did
not experience the decision making process to be
difficult. Similarly, DOU couples were compared
with those who had no doubts about the decision they
had made. Couples who were undecided were
compared with those that had decided to have
children. The latter group is described in detail
elsewhere. 9 10

A previous publication lists the disorders involved
in the study. 9

STATISTICAL ANALYSIS
To identify differences, the relative risk was estimated
by the odds ratio with the levels of significance (p
values) being two tailed. In case of trichotomies, such
as the genetic and recalled risk level, the $\chi^2$
test for
trend was applied. 11 If cell entries in the table
equalled zero, 0·5 was added to each cell to estimate
the odds ratio. 12

From the analysis of single factors significantly
associated with the criteria DIFF, DOU, or UNDEC,
no insight can be obtained into the overlap of the
significant single factors on the criteria. Therefore,
stepwise logistic regression analysis was used to
identify those factors independently and significantly
associated with the criteria (DIFF, DOU, or UNDEC).
This analysis allows for associations with multiple
factors simultaneously. It selects each factor in terms
of its strength of independent association with the
criteria (DIFF, DOU, and UNDEC), while control-
ling other associated factors. Furthermore, it provides
an estimate of the odds of the association between
each factor and the criteria. The factors selected in the
final analysis are presented with the unstandardised
regression coefficient, standard errors, and the
coefficient divided by the standard error. This latter
measure indicates the significance of the association
between the factors and the criteria (a value of >2·0
implies a statistical significance level of p<0·05). This
analysis excludes those subjects who left at least one
item of the questionnaire unanswered. To enable
comparison between single factors in relation to the
criteria and the results of the logistic regression
analysis, those couples who left at least one question
answered were excluded from the statistical
analysis.

Results
Of the 164 couples in the study, 137 had made a
reproductive decision: 109 (66%) had decided to have
(more) children, while 28 (17%) had decided to
refrain from having children. Eighteen couples (11%) were
undecided at the time of the follow up. The
remaining nine couples were excluded from the
statistical analysis, six cases because of failure to
answer all questions (all being couples who had
decided to have children) and the other three cases
because the reproductive decision was basically
different: two couples opted for artificial insemination
by donor, and one (1%) couple waited for prenatal
diagnosis to become available in the near future.
Thus, the statistical analysis applied to 155 couples.

Table 1 shows that 47 couples (30%) who had made
a decision were DIFF couples. Twelve (8%) of those
were also DOU couples. One DOU couple (1%) did
not experience the decision making process as diffi-
cult. Thus, of those who had made a decision, 13
(12+1=9%) were DOU couples. Eighteen couples
(12%) had not made a decision (UNDEC). Of all 155
couples eligible for the study, 43% ((47 DIFF+1
DOU+18 UNDEC)/155) had problems in making the
reproductive decision.

Table 2 lists the factors which were significantly
related to DIFF, DOU, or UNDEC. The strength of
the relationships is indicated as the relative risk (RR).
The more the relative risk deviated from 1·0, the
stronger the association with the criteria. For example,
a couple anticipating a high risk level before counselling
was 2·67 times more likely to become a DIFF couple
than when the risk level was expected to be low (RR
2·67). Couples who did not feel relieved after genetic
counselling were more likely to become a DIFF,
DOU, or UNDEC couple than those who felt relieved
(RR 8·02, 5·10, and 14·36, respectively).

The higher the genetic risk, the stronger the
association with the criteria (DIFF, DOU, UNDEC).
Couples with a genetic or recalled risk >15% were
most likely to become DOU couples (RR 9·09 and
19·92, respectively). Of the couples that had decided
to have children, those who were eligible for prenatal
diagnosis were more likely to become DIFF or DOU
couples than couples for whom this was not available

*In the 43% experiencing problems in the decision making process,
the overlap of DIFF and DOU couples was excluded.
Table 2 Problems in making the reproductive decision in 155 couples: factors significantly related to difficulty in the decision making process (n=47), unresolved doubts (n=13), or persisting reproductive uncertainty (n=18).

<table>
<thead>
<tr>
<th></th>
<th>Difficult*</th>
<th>Doubtful*</th>
<th>Undecided†</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>RR‡</td>
<td>RR</td>
<td>RR</td>
</tr>
<tr>
<td>Anticipated high risk level</td>
<td>2.67‡</td>
<td>NS</td>
<td>NS</td>
</tr>
<tr>
<td>No postcounselling relief</td>
<td>8.00‡</td>
<td>5.10‡</td>
<td>14.365‡</td>
</tr>
<tr>
<td>Genetic risk: 5-15%</td>
<td>3.38‡</td>
<td>3.33‡</td>
<td>5.92‡</td>
</tr>
<tr>
<td>Recalled risk: 5-15%</td>
<td>3.55‡</td>
<td>9.09‡</td>
<td>6.86‡</td>
</tr>
<tr>
<td>Risk interpreted as high</td>
<td>9.67‡</td>
<td>19.92‡</td>
<td>5.08‡</td>
</tr>
<tr>
<td>Couples opting to have children:</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Prenatal diagnosis available</td>
<td>2.68p</td>
<td>6.70p</td>
<td>—</td>
</tr>
<tr>
<td>Had child(ren) during genetic counselling</td>
<td>NS</td>
<td>4.55p</td>
<td>NS</td>
</tr>
<tr>
<td>Presence of affected child</td>
<td>1.23‡</td>
<td>6.76‡</td>
<td>NS</td>
</tr>
<tr>
<td>Personal acquaintance with disorder</td>
<td>10.71‡</td>
<td>NS</td>
<td>NS</td>
</tr>
<tr>
<td>Decided against having children</td>
<td>NS</td>
<td>5.72‡</td>
<td>—</td>
</tr>
<tr>
<td>Relatives' disapproval of decision</td>
<td>5.35‡</td>
<td>NS</td>
<td>—</td>
</tr>
</tbody>
</table>

*137 couples had made a decision.
†109 couples opted to have children and were compared with those who remained undecided.
‡RR=relative risk estimated by odds ratios.
§p<0.001, ¶p<0.01, *p<0.05, NS=not significant.

Table 3 Stepwise logistic regression for couples experiencing difficulty with the decision making process or subsequent doubts.

<table>
<thead>
<tr>
<th>DIFFICULTY</th>
<th>Coefficient</th>
<th>Standard error</th>
<th>Coeff/SE*</th>
<th>Adjusted relative risk†</th>
</tr>
</thead>
<tbody>
<tr>
<td>(1) Relieved by counselling</td>
<td>-2.11</td>
<td>0.45</td>
<td>-4.71</td>
<td>0.12</td>
</tr>
<tr>
<td>(2) Anticipated high risk level</td>
<td>0.91</td>
<td>0.44</td>
<td>2.04</td>
<td>2.48</td>
</tr>
<tr>
<td>(3) Relatives disapproving the decision</td>
<td>1.96</td>
<td>0.62</td>
<td>3.16</td>
<td>7.07</td>
</tr>
<tr>
<td>Constant</td>
<td>-0.44</td>
<td>0.40</td>
<td>-1.10</td>
<td>0.64</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>DOUBTFULNESS</th>
<th>Coefficient</th>
<th>Standard error</th>
<th>Coeff/SE*</th>
<th>Adjusted relative risk†</th>
</tr>
</thead>
<tbody>
<tr>
<td>(4) Decided against having children</td>
<td>-1.58</td>
<td>0.63</td>
<td>-2.49</td>
<td>0.21</td>
</tr>
<tr>
<td>(5) Presence of affected child</td>
<td>1.73</td>
<td>0.65</td>
<td>2.64</td>
<td>5.64</td>
</tr>
<tr>
<td>Constant</td>
<td>-2.02</td>
<td>0.63</td>
<td>-3.23</td>
<td>0.13</td>
</tr>
</tbody>
</table>

*Coefficient divided by standard error. †Adjusted for the other selected variables.
For all factors 1=yes and 0=no.

(RR 2.68 and 6.70, respectively). Of the couples who decided to have children, 45% of those who were eligible for prenatal diagnosis became DIFF couples and 12% DOU couples, compared with 23% and 2% of those for whom prenatal diagnosis was not available.

Couples with an affected child (irrespective of whether this child had died or survived) were more likely to become DIFF or DOU couples than those without an affected child (RR 2.23 and 6.67, respectively). Couples that refrained from having children were more likely to become DOU couples than those opting to have children (RR 5.72). Couples whose relatives disapproved of their decision were more likely to become DIFF couples than those whose relatives did not disapprove (RR 5.35).

No significant associations were found regarding DIFF, DOU, or UNDEC couples and the following single factors: death or survival of an affected child, the presence of healthy and affected children or only healthy child(ren), the presence or absence of mental retardation in the disorder, the type of and perceived severity of the disorder, and parental age.

Any unresolved doubts experienced by couples who had decided to undertake a pregnancy after genetic counselling were not related to the subsequent health of this child.

STEPWISE LOGISTIC REGRESSION ANALYSIS
All factors that had a statistically significant relationship with DIFF, DOU, or UNDEC were entered into the stepwise logistic regression analysis.

Table 3 shows the factors independently and significantly associated with problems in the decision making process: no postcounselling relief (1), anticipation of a high risk level (2), relatives' disapproval of the decision (3), a decision not to have children (4), and the presence of an affected child (5).

Couples who felt relieved after genetic counselling were less likely (8.33 times=1/0.12) to become a DIFF couple than those who did not feel relieved (1).
Twenty-three of the 35 DIFF couples stated that counselling had not brought relief because the risk was perceived to be high (n=13) or genetic counselling had not produced a definite mode of inheritance in their case in the absence of a postnatal diagnosis (n=10). Couples who anticipated a high risk were more likely (2-48 times) to become a DIFF couple than those anticipating a low risk level (2). Couples whose relatives subsequently disapproved of their decision were more likely (7-07 times) to become a DIFF couple than those who did not meet disapproval (3).

Couples who decided against having a(nother) child were more likely (4-76 times=1/0-21) to become a DOU couple than those who decided to have children (4). Seven DOU couples decided not to have children, in four cases because they felt they had no choice. Reasons given were: "We did not want another affected child, therefore we could not undertake another pregnancy" and "Rationally we know that we have made the right decision, but emotionally it is very difficult to accept". Couples who had an affected child were more likely (5-64 times) to become a DOU couple than those who did not have an affected child (5). Couples with an affected child who decided against a subsequent pregnancy had found the care of their affected child very demanding.

The anticipation of a high risk level in DIFF couples was not related to precounselling worries or postcounselling relief, or genetic or recalled risk category, or risk interpretation.

Couples feeling relieved after genetic counselling were less likely (14-36 times) to remain undecided than those who had not experienced relief. This appeared to be the only factor that contributed significantly to the differentiation between couples that remained undecided and those opting to have children.

Discussion
Altogether, 43% of the study population had experienced problems in the postcounselling reproductive decision. Our finding that 12% of the couples remained undecided after counselling was in agreement with other published reports, after correction for variations in the follow up intervals (table 4). Only Emery et al13 found no reproductive uncertainty at two years' follow up. This may be because at that stage counsellors had been assessed three times, which might have provided additional support in the decision making process.

The postcounselling decision is never easy to make,2 but some couples seem to experience more difficulty than others. The logistic regression analysis indicated several factors which were independently and significantly associated with problems in the decision making process. These were: no postcounselling relief, anticipation of a high risk level, relatives' disapproval of the decision, a decision not to have children, and the presence of an affected child. Jointly these factors can substantially differentiate couples that will develop problems in the decision making process from those who will not.

Where genetic counselling had not brought relief because of the unavailability of a precise diagnosis, couples tended to experience additional difficulty in the decision making process. Kessler et al16 pointed out that feelings of guilt and self-blame tend to increase when the circumstances surrounding the cause and nature of the defect are ambiguous. Because of this ambiguity the couple may be preoccupied with the guilt feelings so that their decision making is shrouded in emotion.16,17 Therefore, it is important to explore the emotional as well as rational aspects of the reproductive decision during counselling. Such exploration might facilitate the decision making process.

The absence of postcounselling relief appeared to be the only factor contributing to the inability to make a decision. The reasons given for remaining undecided showed some similarity with the reasons given by couples who had decided not to have children, such as high risk interpretation or fear of not being able to cope with another affected child.10

It is possible that the desire to have children was stronger for couples who remained undecided than for those who had decided against having children. At any rate the desire was not strong enough for the latter group to counterbalance the fears of having a(nother) affected child.

Couples who expected their risk to be high tended to experience additional difficulty in the decision making process. The reason for this remains unclear. At the very beginning, the genetic counsellor needs to find out whether the consultand is anxious about an anticipated high risk. Talking about this might bring relief, even if the counsellor cannot provide a risk level at that stage.

Couples whose relatives disapproved of the decision tended to experience difficulty in the decision making process, as if they had sensed that disapproval would be forthcoming. This may not have occurred in the consultand's consciousness. The influence of the

<table>
<thead>
<tr>
<th>Time between counselling and follow up</th>
<th>Undecided (%)</th>
<th>Reference</th>
</tr>
</thead>
<tbody>
<tr>
<td>2 years</td>
<td>0</td>
<td>13</td>
</tr>
<tr>
<td>5 months-15 years</td>
<td>19</td>
<td>14</td>
</tr>
<tr>
<td>About 3-8 years</td>
<td>9</td>
<td>6</td>
</tr>
<tr>
<td>6 months</td>
<td>30</td>
<td>7</td>
</tr>
<tr>
<td>7-10 days</td>
<td>24</td>
<td>15</td>
</tr>
<tr>
<td>2-3 years</td>
<td>12</td>
<td>Present study</td>
</tr>
</tbody>
</table>
Analysis of problems in making the reproductive decision after genetic counselling

reaction of other people on the decision making process was also stressed by others. It is therefore important to find out during counselling whether the consultands anticipate disapproval of any decision from relatives.

Couples who decided to refrain from having children tended to have unresolved doubts about their decision. If genetic counselling seems to leave no other option in the perception of the consultands than to refrain from having children, additional counselling may be beneficial. In couples refraining from having children, one or both parents might have experienced the birth of their affected child as a punishment. Taking the entire responsibility for the affected child, this spouse may be bolstering his/her self-esteem by denying him/herself the pleasure of having another child.

Wertz et al. and Sorenson et al. found that couples who had an affected child tended to remain undecided. Our findings of unresolved doubt in the presence of an affected child are similar.

Couples with unresolved doubts have been analysed as a separate group, even though nearly all these couples had experienced the decision making process to be difficult. The factors associated with post-decision doubtness are different from those related to experiencing the decision making process as difficult.

Some single factors need to be mentioned, even though these were not independently and significantly related to problems in the decision making process. Interestingly, the availability of prenatal diagnosis appeared to increase rather than decrease difficulties and doubtfulness for those couples opting to have children. This contradicts the notion that prenatal diagnosis could provide the easy way out. Other investigators have stressed the period of anxiety while awaiting the result of prenatal testing and the burden of selective abortion in the case of fetal abnormality.

The availability of prenatal diagnosis of a specific disorder, such as cystic fibrosis or a neural tube defect, appeared to facilitate the reproductive decision making process because of the widened scope of choices. The present study does not contradict such findings because in our assessment of problems experienced in the decision making process the burden of selective abortion in case of fetal abnormality was incorporated.

In agreement with other published reports we found that couples who interpreted their risk as high tended to experience the decision making process as difficult or to remain undecided.

Owing to the retrospective design of this study, no firm conclusions can be drawn from the reports of precounselling feelings. The recollection might have been distorted by the outcome of genetic counselling or the birth of an affected child after counselling, a mechanism called anchoring. In our study, however, couples who perceived the disorder in their family to be severe seemed to have a particularly vivid memory of the period before and immediately after genetic counselling. Considering the fact that this group did not differ from the entire study population in other respects, we feel that the recollection of precounselling emotions would be similar for the whole study population. A prospective study is required to substantiate this premise.

The emotional impact of the factors which can substantially differentiate couples who are developing problems in the decision making process from those who will not can be discussed during counselling. However, the emotional impact of these factors might only become apparent after counselling rather than in the course of it. Therefore, we will develop a second questionnaire based on these factors, to be used three to six months after genetic counselling for couples requesting genetic counselling with respect to future offspring. This questionnaire will be tested in a forthcoming study. This second questionnaire also gauges the need for additional support, although probably not all couples that need support will accept the offer.

When consultands are still undecided nine months after genetic counselling, they may benefit from additional counselling provided by a psychosocial worker or psychologist familiar with their specific problems.

The authors are indebted to the Foundation of Clinical Genetics, Rotterdam, and to Professor H Galjaard for instigating this study. They also wish to thank Sophie van de Berge and Eva Ketzer for conducting the interviews with great dedication and accuracy, Alice Ribbink for editing the manuscript, and Mirko Kuit for the illustration.

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