Objectives of genetic counselling: differing views of purchasers, providers and users

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Abstract

Background This study aimed to compare the views of purchasers, providers and users about the objectives of genetic counselling.

Methods A modified Delphi technique was used, incorporating two postal questionnaires that were sent to six study groups with a three-month interval: purchasers (public health doctors \( n = 37 \) and regional advisers in general practice \( n = 35 \)); providers (clinical geneticists \( n = 33 \) and genetic counsellors \( n = 25 \)); and users (out-patients \( n = 36 \) and members of genetic support groups \( n = 32 \)). The response rate for the first questionnaire was 115/198 (58 per cent) and for the second, 102/198 (52 per cent). The first questionnaire asked an open-ended question about what the objectives of genetic counselling should be and asked respondents to rank order them. The second questionnaire summarized the views expressed in the first questionnaire and asked respondents to rank order the most frequently cited and most highly ranked five objectives.

Results The five most frequently cited and highly ranked objectives from Questionnaire One were, in descending order: provide information, give support, facilitate decision-making, assess risk and achieve understanding. In response to Questionnaire Two, purchasers differed from providers and users in rating 'facilitate decision-making' more highly than did providers and users. By contrast, providers and users rated 'give information' more highly than did purchasers.

Conclusions Purchasers hold different views from providers and users about what the objectives of genetic counselling should be. This raises two questions: (1) Which views are or should be most influential in the future development of genetic counselling? (2) By what processes can more than one view on genetic counselling be integrated?

Keywords: genetic counselling, Delphi technique, consensus, genetic services

Introduction

The first step in evaluating a service is to identify appropriate outcome measures. Usually these are derived from the stated objectives of the service. This process, however, becomes difficult when there is uncertainty about the prime purposes of the service in question. This problem applies particularly to genetic counselling, where there seems to be a lack of clarity and conflicting objectives. For example, a widely quoted definition of genetic counselling by Fraser\(^1\) (p 637) stressed the importance of information giving, understanding and informed decision-making. Information-giving and understanding find echo in other guidance on genetic counselling\(^2\) but in the main there is less agreement on what is meant by informed decision-making. Descriptors such as 'voluntary' and 'appropriate' offer little guidance to the researcher trying to identify the most suitable measures of successful counselling, and when specific criteria are offered – such as the Royal College of Physicians\(^7\) view that a desirable goal is to reduce the number of births of affected persons – it does not find universal support.

When there is a lack of clarity over the objectives of a service, one way of proceeding is to try and identify the criteria on which there can be consensus. In particular, the views of the three main protagonists in the area, namely, purchasers of the service, providers of the service and users of the service, would all seem to have some claim to legitimacy. The current study was designed to establish the extent to which consensus exists between these three groups about the objectives of genetic counselling and to see how far their views concord with current guidelines. If differences between groups were found, a further aim was to seek views about whether these differences were seen as problematic.

The method chosen for the study was the Delphi technique. This is a way of eliciting and combining the opinions of a group of responders using a controlled series of information exchanges.
amongst a group who do not meet face-to-face. It involves sending individuals an anonymized questionnaire with open-ended questions, feeding back their summarized responses and asking for further responses to, and ratings of, these data in an iterative process until the extent of consensus is established. A good description of this technique has been given by Fielding. The method is suitable for areas where definitive answers do not exist and where population presents diverse backgrounds with respect to experience or expertise. It also has the advantage over other consensus methods of allowing divergence from publicly expressed views and limiting any dominating influences of particular groups or individuals and enabling all groups to feel involved. Because it is conducted by post, it avoids the logistical problems of a consensus meeting.

Method

Design

The study used a modified Delphi technique, in which two postal questionnaires were sent to six study groups with a three-month interval.

Participants

The sample comprised UK National Health Service purchasers and providers and users of genetic counselling (total n = 198). Purchasers were public health doctors (n = 37) and general practitioners (GPs) (n = 35). Regional public health faculty advisers nominated two public health doctors per region and the president of the Faculty of Public Health Medicine nominated seven experts in the area. Regional advisers in general practice nominated one or two associate advisers per region. Although these GPs were not selected as direct purchasers of secondary care (although some may have been fund-holders at the time), they were indirect commissioners of genetic services through the referral process and they have therefore been assigned to the ‘purchaser’ group for this study. Providers were clinical genetic consultants (n = 33) and genetic nurse counsellors (n = 25) from four regional genetics centres. Users of genetics services were out-patients from one regional centre who had agreed to take part in a previous study (n = 36) and co-ordinators of patient support groups affiliated to an umbrella organization representing the interests of families affected by genetic disease (n = 32). The study was carried out in 1995–1996.

Procedure

Participants were sent a questionnaire with a tear-off slip which they were asked to return separately once they had returned the questionnaire. This allowed non-responders to be followed up whilst ensuring that responses were anonymous. A reminder letter and further questionnaire were sent to all those who had not replied after four weeks. A second questionnaire was sent to respondents after three months using the same procedure.

Measures

The first questionnaire outlined the nature of the study and asked respondents the question ‘What do you think the objectives of genetic counselling should be?’ They were asked to list and then rank the objectives in order of importance. Responses were coded and used as the basis for the second questionnaire. Categories were generated by the second author by grouping together responses to all questionnaires. The adjusted list contained 15 categories. A further 30 questionnaires were independently coded by the first two authors, to determine inter-rater reliability. As this was high (kappa >0.90), reliability was not checked on a larger sample.

The second questionnaire listed the five objectives which were most frequently mentioned and most often ranked as most important by the whole sample in the first questionnaire. The frequency with which each of the six participating groups mentioned these objectives was illustrated using histograms. Participants were asked to rank the five categories in order of importance in the light of this information. They were also asked ‘To what extent do you consider there to be a consensus (agreement) between the groups?’, using a five-point rating scale with 1 as ‘no consensus at all’ to 5 as ‘complete consensus’.

Analysis

To ensure adequate numbers for analyses, the six study groups were combined to make three: purchasers, providers and users. In the first questionnaire the three groups were compared in terms of their ordering of objectives (Mann–Whitney U) and the numbers of time particular objectives were mentioned (x^2). In the second questionnaire the scores given to each objective were compared across the three groups (Mann–Whitney). There were insufficient data to perform a Kruskal–Wallis test.

Results

Response rates to the first questionnaire were as follows: public health doctors 27/37 (73 per cent); regional advisers in general practice 19/35 (54 per cent); clinical geneticists 22/33 (67 per cent); genetic counsellors 7/25 (28 per cent); out-patients 17/36 (47 per cent) and Genetic Interest Group members 23/32 (72 per cent). The overall response rate was 58 per cent (115/198). Response rates to the second questionnaire were: public health doctors 23/37 (62 per cent); regional advisers in general practice 17/35 (48 per cent); clinical geneticists 19/33 (58 per cent); genetic counsellors 6/25 (24 per cent); out-patients 13/36 (36 per cent) and Genetic Interest Group members 24/32 (75 per cent). Overall, 52 per cent replied to both questionnaires (102/198).

The respondents to the first questionnaire identified distinct objectives for genetic counselling. Ninety-nine respondents gave a ranking, 11 ranked all objects as equal and five gave no ranking. The results are shown in Table 1 (full definitions of the objectives are available from the corresponding author). There...
The mean ranks from the purchaser, provider and user groups are given in Table 2. Purchasers valued ‘facilitate decision-making’ more highly than users (Mann–Whitney \( U = 414; p = 0.005 \)). Both users and providers valued ‘giving information’ more highly than purchasers (respectively, Mann-Whitney \( U = 438 \) and \( U = 207; p = 0.01 \) and \( p = 0.0005 \)).

There was no significant difference between users and providers in their judgements of consensus but purchasers viewed the results as reflecting less consensus than did either users or providers (2.0 (SD 1.1) for purchasers, 2.8 (SD 0.8) for providers and 2.6 (SD 0.9) for users; Mann–Whitney \( U = 730, p = 0.0006 \)).

### Discussion

The first round of the Delphi process produced a reasonable response and consensus across the three groups as to the most important objectives. The top five objectives were consistent with those stated in existing guidelines\(^6\) and with a common definition of genetic counselling.\(^7\) They did not, however, include the Royal College of Physicians’ performance indicator for a genetic testing service, the reduction in the number of births of affected persons. These results give some support to the legitimacy of using these five objectives in future evaluations of genetic counselling services. It should be noted, however, that the very low response rate of genetic counsellors calls into question the representativeness of the respondents in this group.

There were some differences between the three groups in the emphasis they gave each objective. Perhaps reflecting their public health orientation, purchasers were more keen on risk assessment than the other two groups, whereas the other groups valued providing support more highly. Such emphases do not

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**Table 1** Number of times objectives were stated* (with rank orderings for each group)

<table>
<thead>
<tr>
<th></th>
<th>Purchasers ( n = 46 )</th>
<th>Providers ( n = 29 )</th>
<th>Users ( n = 40 )</th>
</tr>
</thead>
<tbody>
<tr>
<td>Facilitate decision-making</td>
<td>26 (1 = 1)</td>
<td>18 (3)</td>
<td>18 (3)</td>
</tr>
<tr>
<td>Risk assessment</td>
<td>28 (1 = 1)</td>
<td>9 (7)</td>
<td>10 (5)</td>
</tr>
<tr>
<td>Give information</td>
<td>24 (3 = 1)</td>
<td>21 (2)</td>
<td>28 (2)</td>
</tr>
<tr>
<td>Support</td>
<td>24 (3 = 1)</td>
<td>25 (1)</td>
<td>32 (1)</td>
</tr>
<tr>
<td>Achieve understanding</td>
<td>10 (5)</td>
<td>14 (5)</td>
<td>15 (4)</td>
</tr>
<tr>
<td>Facilitate coping</td>
<td>9 (6 = 1)</td>
<td>16 (4)</td>
<td>9 (3)</td>
</tr>
<tr>
<td>Professional education/liaison</td>
<td>9 (6 = 1)</td>
<td>4 (8)</td>
<td>2 (11 =)</td>
</tr>
<tr>
<td>Advice</td>
<td>6 (8 = 1)</td>
<td>1 (11 =)</td>
<td>5 (7 =)</td>
</tr>
<tr>
<td>Quality of service</td>
<td>6 (8 = 1)</td>
<td>3 (9)</td>
<td>4 (9)</td>
</tr>
<tr>
<td>Diagnosis</td>
<td>5 (10 =)</td>
<td>11 (6)</td>
<td>3 (10)</td>
</tr>
<tr>
<td>Public health</td>
<td>5 (10 =)</td>
<td>2 (10)</td>
<td>1 (13 =)</td>
</tr>
<tr>
<td>Treatment</td>
<td>2 (12)</td>
<td>1 (11 =)</td>
<td>0 (15)</td>
</tr>
<tr>
<td>Public education</td>
<td>1 (13 =)</td>
<td>1 (11 =)</td>
<td>2 (11)</td>
</tr>
<tr>
<td>Research</td>
<td>1 (13 =)</td>
<td>0 (15)</td>
<td>1 (13 =)</td>
</tr>
<tr>
<td>Tests</td>
<td>0 (15)</td>
<td>1 (11 =)</td>
<td>5 (7 =)</td>
</tr>
</tbody>
</table>

*All those who ranked the objective first are included. In addition, those who ranked two objectives as most important were also included (but not more than two).

\( ^* \) denotes tied ranking.
detrap from the general importance accorded these objectives but they do suggest that purchasers and providers at least might usefully share their views on why exactly genetic counselling is being provided.

The second questionnaire, which assessed the re-ranking of the different objectives after respondents had considered the views expressed by all groups in the first round, showed some changes. The inevitable response attrition between the first and second questionnaires may account for some of the change, though the numbers being lost to follow-up (13 people) were relatively small.

The main change from first to second questionnaire was in the emphasis accorded to 'giving information'. Purchasers ranked it last of the five objectives whereas both providers and users moved it into first position; these differences were confirmed by the significantly different range of scores given this item across the three groups. In many ways the differences between some of the objectives selected is one of degree. For example, 'giving information' and 'achieving understanding' are closely related, the latter presumably presupposing the former, yet respondents did seem able to distinguish between them and apply different judgements. Accordingly, the fact that purchasers emphasized different levels of information transfer than other groups might reflect their different perspectives on the genetic counselling service. In many ways, achieving understanding as a prelude to better decision-making would seem desirable but the fact that providers and users valued giving information more highly may reflect either a belief that information on its own speaks for itself or that this is all that can realistically be achieved in the consultation. Given that information transfer is such a central part of genetic counselling it would seem important that this difference in why information is being imparted is further explored.

Overall, the Delphi exercise seemed to be a useful process to establish a consensus about appropriate outcome measures for genetic counselling. Broad agreement on the five key objectives gives legitimacy to using these as the basis of any evaluation of the service. Nevertheless, differences between the groups do exist, and were perhaps heightened by the iterative Delphi process rather than diminished. These may have implications for how the service is actually provided.

This study shows that purchasers hold different views from providers and users about what the objectives of genetic counselling should be. There are several possible explanations for our findings. It may be that the differences revealed are semantic and that purchasers are simply responding to the question in a different way but the values held by all groups are in fact very similar. It is also possible that those responding are unrepresentative of their groups. The response rate of 52 per cent is not uncommon for this type of study and we have no way of knowing whether there was a systematic bias operating across the groups that might account for the observed group differences. Alternatively, the concordance between providers and users may be because of their shared direct experience of genetic counselling consultations. Their views may therefore reflect current practice more than those of purchasers, who perhaps draw on different data to make their judgements. Purchasers' evidence-based medicine and financial perspectives may lead to a greater consideration of outcome (the achievement of understanding and informed decisions) rather than the process or activity of giving information, which was given priority by providers and users.

If the differences are more real than apparent, as is suggested by the difference in the guideline literature, purchasers may be applying different criteria to the purchasing of services than those valued by users and providers. Some of the respondents added comments about the results which reflected this concern:

'The shape of the curve of the graphs for outpatients, Genetic Interest Group, geneticists and counsellors is quite consistent but the public health doctors (who are likely to influence purchasing) is different. This is important to note if providers are to meet family needs.' Provider (genetic nurse counsellor).

'The highest consensus was that genetic counselling should provide support: the highest proportion gave this in each group except for the public health doctors. This is worrying because public health doctors have considerable influence on purchasing decisions and yet were obviously out of tune with all other groups (I am speaking as a public health doctor myself.)' Purchaser (public health doctor).

'Those specifically working in genetics and those affected by genetic conditions seem to be in greater agreement than the public health doctors and GPs. Is there a lack of communication here?' User (Genetic Interest Group member).

Two questions arise from these concerns: (1) Which views are or should be most influential in the future development of genetic counselling? (2) By what process can more than one view on genetic counselling be integrated into purchasing decisions?

Acknowledgements

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References

4 Health Council of the Netherlands, Committee on Genetic


7 Royal College of Physicians; Clinical Genetics Committee. *Purchasers' guidelines to genetic services in the NHS: an aid to assessing the genetic services required by the resident population of an average Health District.* London: Royal College of Physicians, 1991.


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