Valuing preferences for the process and outcome of clinical genetics services: a pilot study

Ewan Gray[a], Martin Eden[a], Caroline Vass[a], Jordan Louviere[b], Katherine Payne*[a]

[a] Manchester Centre for Health Economics, The University of Manchester, Manchester, UK

[b] CenSoC, University of Technology Sydney, Australia

* Address correspondence to:

Katherine Payne,
Manchester Centre for Health Economics,
The University of Manchester,
Jean McFarlane Building,
Oxford Road, Manchester M13 9PL, UK

E-mail: katherine.payne@manchester.ac.uk.

Paper presented to Health Economists Study Group Meeting, Sheffield

8th to 10th January 2014

Acknowledgements: We would like to thank all survey respondents who took time to complete the DCE. We would also like to thank Leonie Burgess and Deborah Street, University of Technology, Sydney, for their advice on the design of the choice experiment.

Abstract (A024)

Background: Clinical genetics services continue to evolve, in part, as a response to technological advances in genetic-based diagnostics. Clinical genetics services comprise complex interventions such as genetic counselling and genetic based diagnostic testing. Importantly, outcomes are multifaceted and various process attributes may also shape the service users' experience of care. One useful piece of information for service providers seeking to improve, or modify, clinical genetics services is to understand whether process or outcome attributes are the key driver of preferences. The nature of the outcome attribute for clinical genetics services (informed decision making), which can not be readily quantified introduced a considerable challenge for this stated preference study.

Aim: To identify if a linked conjoint analysis (CA) and discrete choice experiment (DCE) can be used to quantify the relative importance of process and outcome attributes for a 'generic' clinical genetics service.

Methods: A two-step pilot stated preference survey, using CA followed by a DCE, was administered (online) to a purposive sample of service users (completed face to face) with experience of clinical genetic services and members of the public (via remote electronic completion). The CA component was designed to elicit which of 13 attributes influenced preferences on 'ability to make an informed decision', using a rating scale ranging from 1 to 9. The DCE that followed comprised six attributes (location of the service; pre-consultation contact; turnaround time for a test result; follow-up contact; cost of services; and a scale rating the ability to make an informed decision). The discrete choice task asked respondents to chose a service and then which, if any, they felt they would actually use in real life. An orthogonal main effects design was used to generate choice sets for the CA and DCE. A three-step approach was taken to analyse data (i) a random effects probit regression to identify preferences for the preferred service (ii) an ordered-logit model to estimate the relative value of the CA attributes (iii) a linking of the CA and DCE data through hierarchical information integration.

Preliminary Results: The stated preferences of respondents (n=37) revealed most would prefer a service better at helping them make an informed decision, to wait less time, pay less and receive pre-consultation contact. Therefore, an outcome rather than process attribute was the key driver of preferences. The implications of the linked design and findings of this pilot study will be discussed in the context of designing stated preference surveys of complex interventions.

Introduction

Advances in molecular genetics, biochemical genetics, immunogenetics and somatic cell genetics have helped to increase understanding about the causes of genetic disease. These advances have led to dramatic implications for clinical genetic service delivery with the availability of new genetic-based diagnostic technologies such as next generation sequencing and exome sequencing. These developments have resulted in a rise in the number of individuals and families for whom clinical genetics services, genetic counselling and genetic testing may be appropriate.

In the UK NHS, clinical genetics offers individuals, and their family, services for diagnosis, prevention and management of genetic disorders (McKusick 2002). Clinical genetic services (CGS) generally involve referral for a consultation with a relevant expert, sometimes including a physical examination, followed up by further consultations, genetic counselling and genetic testing, as necessary. Current NHS models of CGS delivery follow a hub and spoke model where Consultants, Specialist Registrars and Genetic Counsellors conduct clinics both in the Specialist Centre and at district hospitals throughout the health region (Donnai & Elles 2001). A crucial component of CGS is genetic counselling, which is the user interface and the aspect of the service most apparent to service users. Genetic counselling is a communication process that usually involves a range of activities (Fraser 1974).

CGS continue to evolve, in part, as a response to the technological advances in genetic-based diagnostics and the associated increase in demand for genetic counselling. Whilst it is envisaged that Regional Genetics clinics will continue to offer a comprehensive genetic counselling service to those at high risk of rare genetic disorders, it is worth considering whether some groups of individuals at low-moderate risk could be seen in other settings, such as primary or secondary care. One possibility is for genetic counsellors to act in a liaison capacity with general practices in primary care to help in the assessment of individuals who could be seen in primary care and those for whom referral to a Regional Genetics Clinic would be more appropriate. Therefore, with growing demand, it is important for decision makers to understand how to effectively and efficiently configure CGS taking into account the process of service delivery but also the effect on ‘patient’ outcomes.

CGS comprise a complex set of interventions and services such as genetic counselling and genetic testing with multifaceted and various attributes shaping the service users’ experience of care. There is a correspondingly complex set of outcomes that often offer little in the way of traditional health gains. Standard measures of clinical outcome, health status and the value of gains in health related quality of life have been considered insufficient for the rigorous evaluation of existing and new

genetics services (Clarke 1997; Payne & Thompson 2013). Unlike other areas of medicine, the 'patients' are usually healthy (Kessler & Bloch 1989) and no specific treatment is offered (Clarke et al. 1996). The users of a CGS may therefore place greater value on how the service is provided and recognise the value of the process of service delivery, such as who provides the service and where the service is located. Qualitative methodologies (grounded theory and thematic analysis) have been used to elicit service users' perceptions of what makes genetic counselling effective (Bernhardt et al. 2000; Skirton 2001). These studies concluded that key process attributes, which drove the effectiveness of genetic counselling were: a good 'connected' relationship with the genetic counsellor; time to outline their concerns and being given information. More recently, McAllister et al. (2008) used focus groups and semi-structured interviews to explore what process attributes of CGS may be highly valued by patients and service providers and identified five process attributes: (1) local and accessible services (2) open access and follow-up, (3) coordinated, tailored family care, (4) quality of the patient–clinician relationship and (5) time to talk.

Attempts to define an outcome for CGS has been a contentious issue. A simplistic outcome measure for CGS that has been employed to suggest genetic counselling is effective is 'recall of information'(Abramovsky et al. 1980). Measuring the effect of decisions about future reproductive behaviour, and associated decision to terminate a pregnancy, is a contentious outcome measure that has been used in the past (Kessler & Levine 1987; Frets et al. 1990). Considering the number of terminations of pregnancy as an outcome has distinct eugenic overtones. Recent work has identified the primary goal of CGS as 'empowering' patients to improve their ability to make an informed decision. A key aim of genetic counselling is to help counselees adjust to their situation and cope emotionally with the disease or risk of disease in their family. McAllister (2008) developed a new outcome measure (CGOS-24) designed specifically to capture 'empowerment' and the key domains of a genetic counselling episode: (1) able to make important life decisions in an informed way (decisional control); (2) has sufficient information about the condition, including risks to oneself and one's relatives, and any treatment, prevention and support available (cognitive control); (3) able to make effective use of the health and social care systems for the benefit of the whole family (behavioural control); (4) able to manage one's feelings about having a genetic condition in the family (emotional regulation); (5) can look to the future having hope for a fulfilling family life, for oneself, one's family, and/or one's future descendents (hope). In the economics literature, the concept of empowerment has been viewed as one operationalisation of Sen's notion of capability (Payne et al. 2013) by capturing '(cap)ability to make an informed decision' and, therefore valuing cognitive capability.

Although different models of CGS have been proposed, such as moving to primary care, a notable feature of the literature to date has been the absence of information about preferences for such service delivery models. Information on preferences may provide useful evidence for service providers seeking to improve, or modify, CGS is to understand whether the process or outcome of the service are the key driver of preferences. Stated preference methods can be usefully applied to inform the development of new models of service delivery (Ryan & Farrar 2000). Discrete choice experiments (DCEs) are a particularly useful form of stated preference method because they can identify the trade-offs that individuals make between the process and outcome-focused attributes of a new service (Ryan & Gerard 2003). The attributes in a DCE can relate to the clinical outcome of the service or the process of providing the service. The ability to incorporate both outcome and process attributes is one of the key advantages of this method. The nature of the outcome attribute for CGS (ability to make an informed decision), which is subjective and complex poses a challenge for the design of a feasible and useful stated preference study. For this reason, other DCEs which have looked at preferences for genetic services have focussed mainly on process attributes such as information provision, how results are disclosed and location rather than the outcomes of the service (Hall et al. 2006; Peacock et al. 2006; Regier et al. 2009).

This study aimed to pilot a tool to elicit preferences and understand whether patients make trade-offs between process attributes and the primary outcome of a CGS. An important output for this study was to explore whether a modified DCE framework could be used to elicit preferences for a service with a subjective and complex outcome (informed decision making).

Methods

This study used an online survey combining a CA and discrete choice experiment (DCE) task.
The online survey comprised five tasks:

Task 1: Making a genetics healthcare decision. Respondents each read a scenario that described a genetic consultation (designed with input from 3 genetic counsellors) and use of a standardised scale (Bradley et al. 1996; Entwistle et al. 2001) to rate the level of involvement with decision making they would prefer. This task was included to understand the level of involvement each respondent perceived they might want when taking part in a genetic counselling episode.

Task 2: Effect of service on ability to make decisions. Respondents completed four CA questions and were asked to rate how much they prefer each scenario that described different types of information that could be made available to inform decision making from the clinical genetics service, using a 9-

point scale (see Figure 1 for an example). The purpose of the CA was to identify which factors are important to patients when making decisions during a clinical genetics consultation.

Task 3: Importance of service characteristics. This task was used as a warm-up exercise and asked respondents to rank the six attributes used to describe a generic CGS in order of importance.

Task 4: Choosing your preferred genetics service. Respondents completed five questions that involved making a discrete choice between two scenarios describing a generic CGS. The purpose of the DCE was to identify if respondents made trade-offs between process and an outcome attribute.

Task 5: Background. The final task asked respondents ten questions about themselves including their use of CGSs. Respondents were also asked for their views about the survey or CGS, in general.

Sampling frame

The pilot study aimed to elicit preferences from the public and a population of patients over 16 years of age who have experience of using a CGS. Patients who attended for a CGS appointment were identified when they came to clinic for a pre-arranged appointment at the Clinical Genetics Department, St Mary's Hospital, Manchester. Information sheets were mailed to respondents prior to the clinic visit. Individual consultants/genetic counsellors checked the daily clinic appointment lists to screen out patients who are ineligible for the study. The study excluded people with learning disabilities and those who were severely ill. A researcher (KP) was present in the waiting room and recruited respondents while they were waiting for their clinic appointment.

Designing the CA and DCE

The first step in the design of a CA or DCE is to identify the attributes to include in the questionnaire. The process attributes included in the CA and DCE were informed by a previous qualitative study using focus groups and face-to-face interviews to explore the views of patients, health care professionals and user representatives (see McAllister et al. 2008). The outcome measure (ability to make an informed decision) used in the DCE was informed by an extensive programme of work that culminated in the development of the empowerment scale (see for example: McAllister met al. (2008) and McAllister et al. (2011)). Table 1 shows the attributes included in the CA exercise. Each attribute was assigned two levels; present or absent. Figure 1 shows an example CA question.

Table 1: Attributes in conjoint analysis exercise

Attribute	Levels
Information about genetic risks	Yes / No
Information from genetic test results	Yes / No
Information about a diagnosis	Yes / No
Information about possible treatment or management options	Yes / No
Information tailored to your ability to understand it	Yes / No
Information tailored to your specific circumstances	Yes / No
Counsellor tells you what s/he thinks you should do	Yes / No
Counsellor explores with you the possible consequences of the different options you could take	Yes / No
Counsellor allows you time to talk	Yes / No
You feel like you have a good relationship with the counsellor because they seem to understand you and your problems	Yes / No
Counsellor helps you work out which option best fits with your values and life goals	Yes / No
Counsellor helps you make sense of your existing knowledge	Yes / No
Counsellor explores how your previous experience and coping strategies could influence your decision	Yes / No

Table 2 shows the attributes included in the DCE. The DCE included two "value" attributes: cost of the service and turnaround time. These value attributes were included to allow comparison between respondents by calculating marginal rates of substitution. The level range for each attribute was set to represent clinically meaningful options and be sufficiently wide to encourage respondents to take account of each attribute and limit the possibility of dominant preferences. This consideration was particularly important for the two value attributes. The wording of the survey was pre-piloted in a sample of ten patients who were sent a postal version of the survey. Further pre-pilot work in another sample of ten patients helped to further refine the framing of the attributes and levels and the general survey design. A key output of this pre-pilot work was to use the phrase 'ability to make an informed decision' rather than the overarching concept 'empowerment'.

Experimental Design

The CA exercise was designed using an orthogonal main effects plan for thirteen 2-level attributes, which was combined with a foldover (mirror image) of the original design to maintain orthogonality and potentially significant 2-way interactions. Producing choice sets in this way provides protection from bias in the main effects. The DCE binary choice sets were created using Street & Burgess (2007) methods for an $8 \times 4^2 \times 2^2$ design. Online software was used to check the design could estimate main-effects and compare the design efficiency with the optimal design for a binary choice set. Six versions of the survey were created that contained 9 choice binary sets (eg. See Figure 2).

Figure 1: Example of CA question

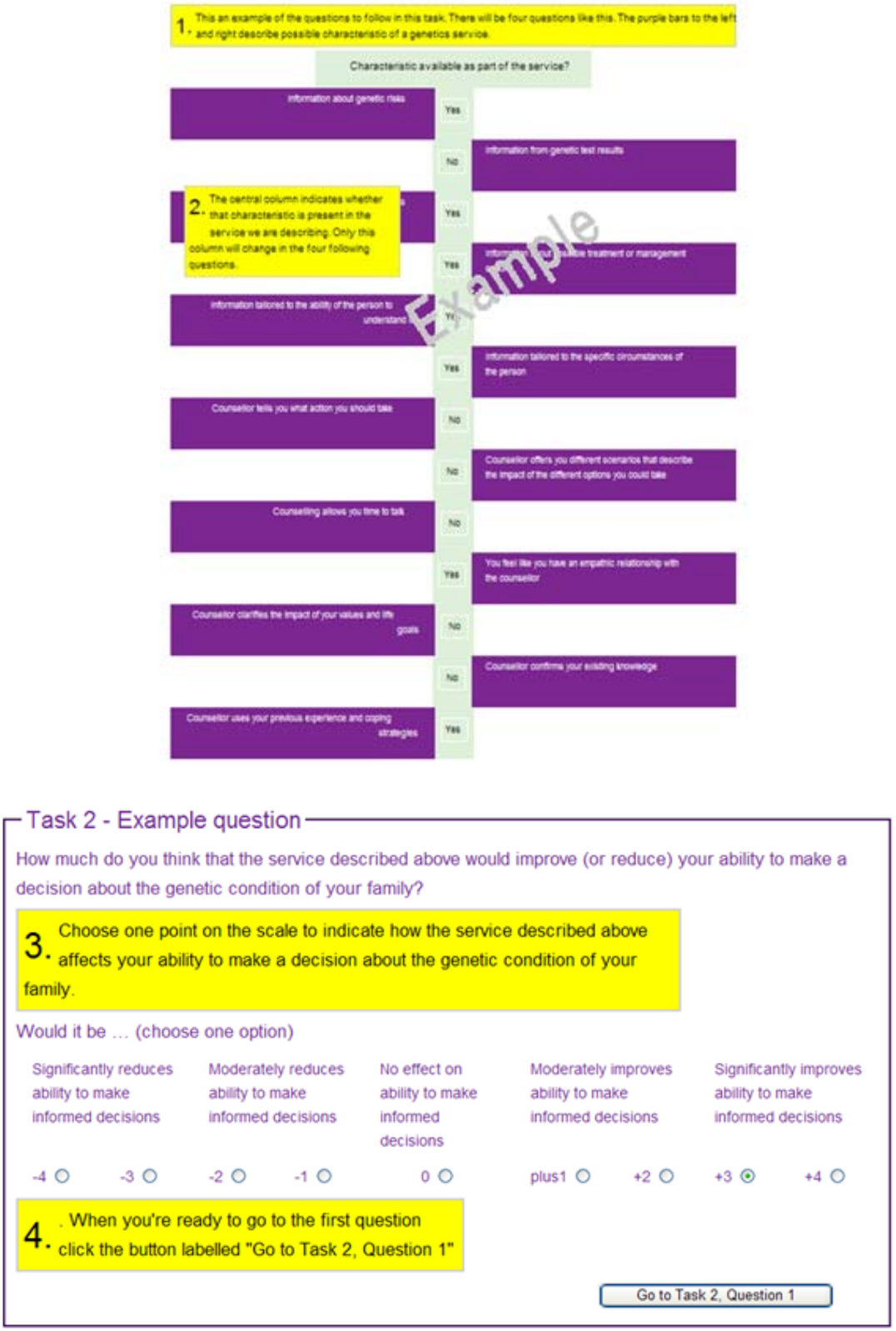
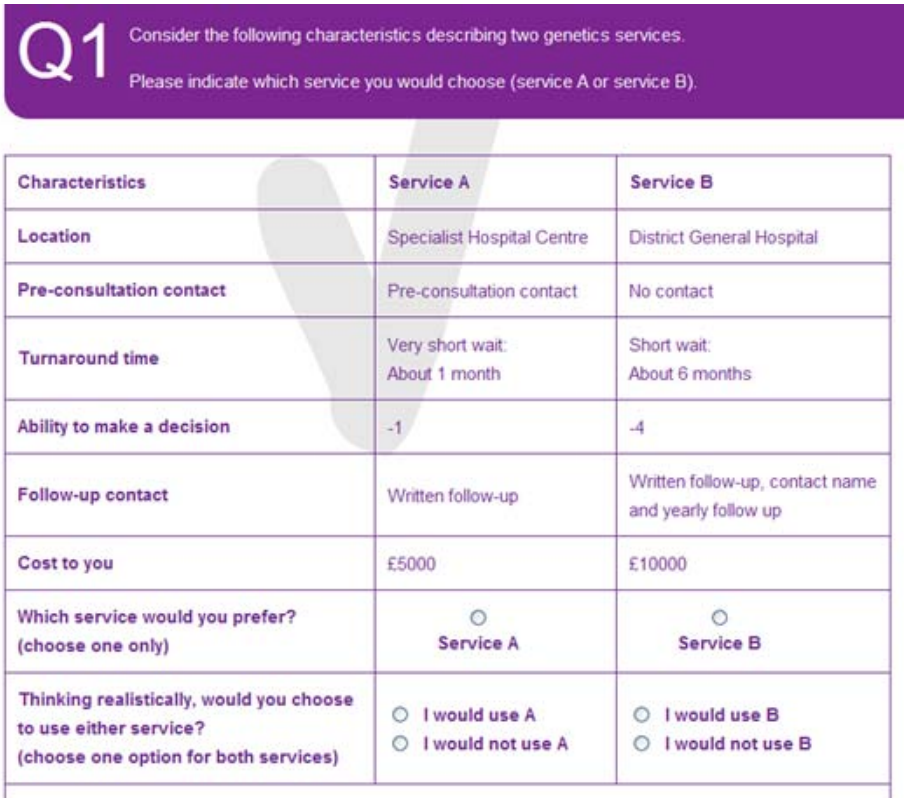


Table 2: Attributes and levels used in the DCE exercise

Attributes	**Levels**
Location of the service	Specialist Hospital Centre District General Hospital
Pre-consultation contact	Pre-consultation contact No contact
Turnaround time for test result and/or diagnosis,	Very short wait : 1 month Short wait: 6 months Medium wait: 12 months Long wait: 24 months
Rating of how much the service improves your ability to make an informed decision.	Your rating is measured on a line 1 2 3 4 5 6 7 8 9
Follow-up after the consultation.	Written follow-up summary letter only Written follow-up summary letter & one personal phone call follow-up Written follow-up summary letter & given a contact name & phone number Written follow-up summary letter and given a contact name, phone number and you are contacted yearly
Total cost of the service.	£50 £2,000 £5,000 £10,000

Figure 2: Example of a DCE question



Q1 Consider the following characteristics describing two genetics services.

Please indicate which service you would choose (service A or service B).

Characteristics	Service A	Service B
Location	Specialist Hospital Centre	District General Hospital
Pre-consultation contact	Pre-consultation contact	No contact
Turnaround time	Very short wait: About 1 month	Short wait: About 6 months
Ability to make a decision	-1	-4
Follow-up contact	Written follow-up	Written follow-up, contact name and yearly follow up
Cost to you	£5000	£10000
Which service would you prefer? (choose one only)	○ Service A	○ Service B
Thinking realistically, would you choose to use either service? (choose one option for both services)	○ I would use A ○ I would not use A	○ I would use B ○ I would not use B

One additional question was included to check the internal validity of the DCE which involved a dominance check. The internal validity question set the levels of the attributes such that the 'dominant' option should be obvious to the respondent.

Mode of administration

Each eligible patient was offered two options: (i) to complete the survey when they return home by accessing a secure website with the address shown in the cover letter (ii) to complete the survey while they are waiting in clinic on a laptop located in the clinic waiting area under the supervision of the researcher. The members of the public completed the survey online via a secure website.

Data Analysis

Responses to Tasks 1, 3 and 5 were analysed using descriptive summary statistics.

Conjoint Analysis: Responses to task 2 were modelled using ordered logit model used to estimate attribute effects on the allocated rating score. In the rating task respondents could rate their answer on a scale between 1 and 9. This was labelled from (-4) "Significantly reduces ability to make an informed decision" through a mid-point (0) "No effect on ability to make an informed decision" to (+4) "Significantly improves ability to make an informed decision". Response values were coded to a 9-point scale. Standard errors were adjusted for clustering of responses within individuals.

Discrete Choice Modelling: The analysis of DCE data aimed to identify the weight attached to each attribute included in task 4. Data were coded as the difference in attribute values between the two alternatives in each choice pair ($X_a - X_b$). Choice was coded as 1 if the respondent chose alternative A in the choice set and 0 if they chose B. A random effects (RE) probit model was used to allow for the clustering of responses within individuals under the assumption random effects assumption that individual specific effects were uncorrelated with the explanatory variables.

Marginal Rates of Substitution and willingness-to-pay: To obtain willingness-to-pay (WTP) estimates for CA attributes it was necessary to obtain predictions for the distribution of scores given each attribute being 'on' or 'off'. These were estimated using single explanatory variable ordered logit models. The experimental design should ensure that these uncontrolled regressions produce unbiased estimates of the attribute effects on score. How the predicted mean differences in "ability to make an informed decision" score were obtained can be explained starting from the specification of the ordered logit model.

The ordered logit models can be described by a latent variable specification in which latent score is a linear function of the explanatory variables:

$$y_{ij}^{*} = \beta X_{ij} + \mu_{ij}$$

X_{ij} is a vector of attributes presented to individual i in choice j. β is a vector of coefficients and μ_{ij} is a random error term with the distribution Logistic(0,1).

The probability of each ordered outcome is modelled as the probability of the latent variable exceeding estimated cut-offs for the outcome:

$$Pr\big(outcome_j = k\big) = Pr\big(\tau_{k-1} < y_{ij}^{*} \leq \tau_k\big)$$

The cut-offs are $\tau_1, \tau_2, \ldots, \tau_{j-1}$ where j is the number of alternative outcomes. In this choice experiment there are 3 outcomes and 2 cut-offs. The predicted probability of each outcome given the presence of each an element attributes in the sub-experiment is given by:

$$\Pr(outcome = k) = \Pr\big(\tau_{k-1} < \beta_i + \mu_j \leq \tau_k\big) = \Pr\big(\tau_{k-1} - \beta_i < \mu_j \leq \tau_k - \beta_i\big)$$

$$= \frac{1}{1 + e^{-(\tau_k - \beta_i)}} - \left(1 - \left(\frac{1}{1 + e^{-(\tau_{k-1} - \beta_i)}}\right)\right)$$

This can be used to predict the average increase in 'ability to make an informed decision' given the addition of an attribute by taking the difference in the expected value of the outcome when the attribute is 'on' and 'off'.

$$\Delta y_i = E(y|X_i = 1) - E(y|X_i = 0)$$

$$= \sum_{1}^{J} \Pr(outcome = j|X = 1)\, j - \sum_{1}^{J} \Pr(outcome = j|X = 0)\, j$$

The marginal rates of substitution of each CA attribute with the DCE cost attribute were calculated by concatenating the results of ordered logit model (CA) and the RE probit model (DCE). The MRS for DCE attributes was calculated by dividing the estimated parameter coefficient for the attribute of interest by the estimated parameter coefficient for the cost attribute.

$$WTP = -\frac{\partial U/\partial X}{\partial U/\partial P} = -\frac{\beta^{x}}{\beta^{P}}$$

Consequently, the MRS for CA attributes is given by:

$$WTP = -\frac{\Delta y_i \beta^{Ability}}{\beta^{P}}$$

Bootstrapping (case re-sampling with 1000 repetitions) was used to estimate 95% confidence intervals for the MRS. This analysis allows a direct comparison of the relative size of the MRS, but note that the absolute size of the MRS for any CA attribute will depend on the levels of other attributes. When this analysis is used to inform an economic evaluation it will be important to know

policy relevant sets of attribute values. Intuitively, the value of additional information or counselling in improving the ability to make an informed decision depends greatly on the existing information and counselling services.

Results

The final sample comprised those who had completed all sections of the survey (n=37). Table 3 summarises the respondents' main characteristics. The majority of the sample were female (76%, n=28). In terms of ethnicity, almost all participants (95%, n=35) described themselves as 'White-British'. There was a fairly broad range of ages represented in this small pilot study; two participants were aged between 16 and 24 years and three were older than 65 years. Forty-one percent (n=15) of the sample was aged over 45 years. The majority of this sample (68%, n=25) were either married or living with a partner. Nine (24%) were single/divorced and three (8%) were widowed. Of those married or living together 64% (n=16) were less than 45 years old.

All but two participants (5%) were in paid employment at the time of completing the survey; one participant was retired and one student completed the survey. Thirty-two percent (n=12) of the sample was employed in education or academia and a further 24% (n=9) were public sector employees. Thirty-eight percent (n=14) of the sample was employed in the private sector. Most participants (92%, n=34) had attained an educational qualification equivalent to GCSE-level or a professional qualification. Nearly half of the sample (49%, n=18) had attained a degree and of these seven participants – representing 19% of the total sample – has gone on to study at post-graduate level. Sixty-two percent (n=23) reported a monthly household income of greater than £1000. Six participants (16%) had a monthly household income of between £250 and £1000 and eight (22%) declined to answer. The sample comprised those with ex ante experience of CGS (51%, n=19) and those without (49%, n=18). Of those had used a clinical genetics service, 52% (n=11) had used the service multiple times, and 89% (n=17) had done so within the last 12 months. In addition, twelve participants – representing 32% of the total sample (n=12) – had visited a clinical genetics service within the three months preceding completion of the survey.

Table 3: Characteristics of survey respondents

Respondent Characteristics	Number of participants (%)
Qualifications	
No qualifications	3 (8%)
O-level / GCSE	10 (27%)
A-level	3 (8%)
Degree	11 (30%)
Higher degree	7 (19%)
Professional qualification	3 (8%)
Monthly household income	
£251 to £500	1 (3%)
£501 to £1000	5 (14%)
£1001 to £2000	3 (8%)
Over £2000	20 (54%)
Do not want to answer	8 (22%)
Number of times CGS used (n=19)	
One visit	8 (42%)
Two visits	4 (21%)
Three to ten visits	5 (26%)
More than 10 visits	2 (11%)
Time of last CGS used (n=19)	
In the last 3 months	12 (63%)
In the last 6 months	3 (16%)
In the last 12 months	2 (11%)
More than 2 years ago	2 (11%)

Understanding respondent's desire to be involved in decision-making

Four (11%) of the 37 participants who completed the DCE task indicated that they would prefer a doctor to tell them what to do in the situation described. The majority of this sample (89%, n=33) would, however, prefer to have at least some input into the decision. Of the 33 favouring active involvement, six participants expressed a preference for making the decision together with the doctor and 27 indicated that they would prefer to be the ones who ultimately make the decision on whether or not to take the test. Whilst most participants would want to arrive at their own decision, answers to task 1 (see Table 4) show how the provision of information from the doctor would be valued by everyone in the situation described; no-one would prefer to arrive at a decision without input from the doctor.

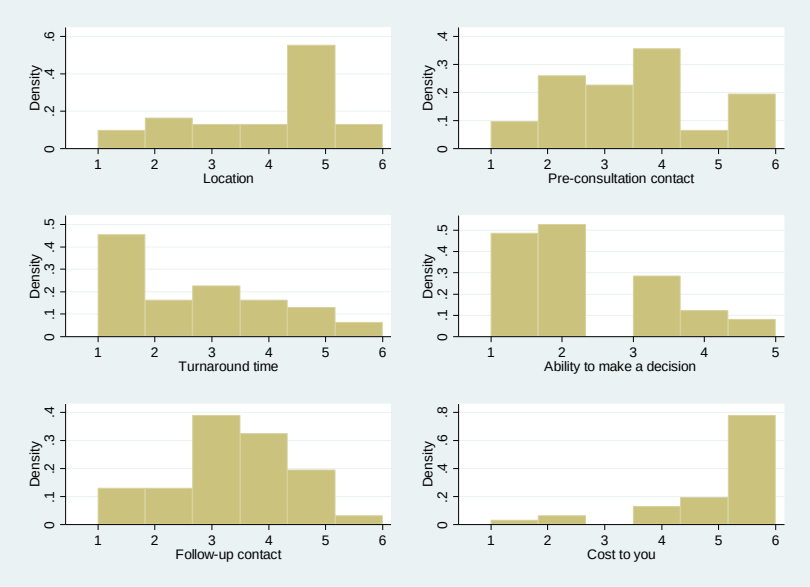
Appendix Table 1: Desire to be involved in shared decision making

Statements: I prefer that…	Number (%) of respondents who agreed with statement
my doctor tells me what to do	1 (3%)
my doctor tells me what to do & explains the choice briefly	0 (0%)
my doctor tells me what to do & explains the choice, including other choices & consequences	3 (8%)
my doctor and I make the decision together	6 (16%)
I make the decision with the doctor giving me both information & a recommendation	23 (62%)
I make the decision with the doctor giving me information but no recommendation	4 (11%)
I make the decision without any information or recommendation from the doctor	0 (0%)

The ranking task

Respondents were asked to rank six attributes of a CGS in order of importance with '1' equating to the 'most important' attribute and '6' indicating the 'least important' (Figure 3). Time taken to obtain genetic test results was considered 'most important' by the largest number of people (38%, n=14). A clinic's propensity to facilitate informed decision-making was also cited as 'most important' by a similar proportion of the sample (32%, n=12). The only attribute not cited as 'least important' was the clinic's propensity to facilitate informed decision-making. Only one person (2.7%) considered the extent of follow-up contact to be 'least important'. Out-of-pocket cost associated with clinic attendance was considered 'least important' by the largest proportion of the sample (65%, n=24).

Figure 3: Importance rating scores by attribute



*The CA and DCE*The data elicited from the CA and DCE were modelled (1) separately and (2) jointly to investigate the influence of CA and DCE attributes on choice of service. Two respondents failed the dominance internal validity test question but were included in the final analysis of this pilot study. Table 4 reports the results of an ordered logit model using the CA data and WTP estimates for each attribute in the CA derived by integrating the ordered logit results with the 'bridging' DCE task. Results of a RE probit model analysing the DCE data to estimate the effect of attributes are presented in Table 5. *CA:* Eleven of the thirteen attributes in the CA were positively associated with the ability to make an informed decision. The exceptions were "Counsellor tells you what action should take" and "You feel like you have an empathic relationship with the counsellor". The information provided by the CGS seemed to be an important determinant of ability to make an informed decision; all information attributes had positively signed coefficients and three of these were statistically significant at the 5% level ("Information from genetic test results", "Information about a diagnosis" and "Information tailored to the circumstances of the person"). The only other attribute to have an effect significant at the 5% level was "Counselling allows you time to talk".

DCE: Table 5 shows the results from the DCE analysis. Location of CGS and degree of follow-up contact did not significantly influence the respondent's choice of service. Attributes with negative coefficients were turnaround time; reflecting respondents' aversion to longer waits for results, and price which demonstrates, unsurprisingly, the respondents preferred to pay less. Patients favoured CGS that offered some pre-consultation contact with a genetic counsellor. They also preferred services which would improve their ability to make an informed decision.

WTP for CA attributes by integration with DCE: The WTP estimates obtained using the concatenated models are shown in table 5. These should be interpreted as the willingness-to-pay for the attribute to be present averaged across scenarios presented in the choice task. In policy evaluation the level of other attributes should be set to reflect conditions under alternative policy choices when estimating WTP for use in economic evaluation. The rank order of WTP values mirrors the order of coefficient values. The 95% confidence intervals are very wide and asymmetric around the point estimates. The asymmetry comes from combining two non-linear models.

Table 4: Conjoint Analysis Model Results – Ordered Logit and Integrated WTP

Attribute	**Coefficient (s.e.)**	**'Average' WTP(£)**	**WTP 95% CI(£)**
Information about genetic risks	0.495	824	-187 to 5533
	(0.318)		
Information from genetic test results	0.680*	1137	29 to 7314
	(0.297)		
Information about a diagnosis	0.740*	1460	21 to 9829
	(0.336)		
Information about possible treatment or management options	0.527	947	35 to 6055
	(0.270)		
Information tailored to the ability of a person to understand it	0.405	625	-504 to 3393
	(0.249)		
Information tailored to the circumstances of the person	0.692**	1310	53 to 8586
	(0.265)		
Counsellor tells you what action you should take	-0.148	-128	-3714 to 1573
	(0.347)		
Counsellor offers you different scenarios that describe the impact of different	0.388	880	-368 to 5965
	(0.342)		
Counselling allows you time to talk	0.763*	1233	11 to 7706
	(0.327)		
You feel like you have an empathic relationship with the counsellor	-0.153	-373	-4042 to 1362
	(0.270)		
Counselllor clarifies the impact of your values and life goals	0.404	511	-678 to 3915
	(0.287)		
Counsellor confirms your existing knowledge	0.730	1122	-508 to 8021
	(0.405)		
Counsellor uses your previous experience and coping strategies	0.065	90	-2039 to 2394
	(0.299)		
cut1 Constant	0.054(0.734)		
cut2 Constant	1.355(0.700)		
cut3 Constant	1.956**(0.756)		
cut4 Constant	2.583**(0.802)		
cut5 Constant	3.269***(0.796)		
cut6 Constant	3.892***(0.815)		
cut7 Constant	4.787***(0.931)		
cut8 Constant	5.945***(1.079)		
Observations	148		

**Significant at 5% or better level*
*** Significant at 1% or better level*
**** Significant at 0.1% or better level*

Table 5: Discrete choice experiment model results for respondents – binary response

	Coefficient (s.e.)	WTP (£)	95% CI (£)
Cost (£)	-0.0000468*	n/a	n/a
	(0.0000197)		
Degree of Follow-up Contact	-0.0657	-1404	-8284 to 2512
	(0.0753)		
Turnaround time (months)	-0.0470***	-1004	-5017, -512
	(0.00877)		
Pre-consultation contact	0.428***	9145	2197 to 49250
	(0.123)		
Ability to make an informed decision	0.0732**	1564	285 to 9044
	(0.0232)		
Constant	-0.176 (0.133)	n/a	n/a
lnsig2u Constant	-13.18 (30.32)		
Observations	185		

Significant at 5% or better level; ** Significant at 1% or better level; * Significant at 0.1% or better level*

Discussion

This pilot study aimed to understand if a hybrid stated preference approach, using CA with a integrated DCE, could be used to understand trade-offs between process and outcome attributes for a complex intervention; CGS involving a subjective measure of outcome 'informed decision making.' The approach used has been termed hierarchical CA with integrated choice experiments and previously has been suggested as a solution to understanding preferences in complex situations (Rungie et al. 2012; Oppewal et al. 2013). This pilot study suggested that the hybrid stated preference approach was able to determine the preferences of a small sample of patients and potential patients of CGS.

The CA indicated that information about a diagnosis and from test results would facilitate informed decision making. Information tailored to the individual would also help in this respect. Being given the time to talk within a genetic counselling session was also considered an important driver of informed decision making by these participants. The DCE revealed that lower out-of-pocket costs, quick turnaround of test results and pre-consultation contact would be preferred attributes of a CGS along with the service's ability to facilitate informed decision making. The DCE results suggested

that these participants would be willing to pay over £1500 to for a service that improved their ability to make an informed decision by one level and £1,004 for a month's reduction in waiting time, with all other things equal.

The available sampling frame, and resulting sample size, for this pilot study placed limitations on the design of the choice tasks. The DCE design could only include main effects. Extending the survey to a larger sample frame and size would allow the DCE design to be modified and include two-factor interactions, but this design would require more scenarios. This preliminary analysis used a simple approach to integrating the results of the CA and DCE tasks to obtain WTP estimates for CA. A more detailed plan of analysis to investigate preference heterogeneity through a combination of structured choice modelling and hierarchical information integration is suggested in Appendix 1.

References

Abramovsky, I. et al., 1980. Analysis of a follow-up study of genetic counseling. *Clinical genetics*, 17(1), pp.1–12.

Bernhardt, B. a, Biesecker, B.B. & Mastromarino, C.L., 2000. Goals, benefits, and outcomes of genetic counseling: client and genetic counselor assessment. *American journal of medical genetics*, 94(3), pp.189–97.

Bradley, J.G., Zia, M.J. & Hamilton, N., 1996. Patient preferences for control in medical decision making: a scenario-based approach. *Family medicine*, 28(7), pp.496–501.

Clarke, A., 1997. Outcomes and process in genetic counselling. In *Genetics, society and clinical practice*. pp. 165–178.

Clarke, A., Parsons, E. & Williams, A., 1996. Outcomes and process in genetic counselling. *Clinical genetics*, 50(6), pp.462–9.

Donnai, D. & Elles, R., 2001. Integrated regional genetic services: current and future provision. *BMJ (Clinical research ed.)*, 322(7293), pp.1048–52.

Entwistle, V. a, Skea, Z.C. & O'Donnell, M.T., 2001. Decisions about treatment: interpretations of two measures of control by women having a hysterectomy. *Social science & medicine (1982)*, 53(6), pp.721–32.

Fraser, F.C., 1974. Current Issues in Medical Genetics. , pp.636–659.

Frets, P.G. et al., 1990. Factors influencing the reproductive decision after genetic counseling. *American journal of medical genetics*, 35(4), pp.496–502.

Hall, J. et al., 2006. What influences participation in genetic carrier testing? Results from a discrete choice experiment. *Journal of Health Economics*, 25(3), pp.520–37.

Kessler, S. & Bloch, M., 1989. Social system responses to Huntington disease. *Family process*, 28(1), pp.59–68.

Kessler, S. & Levine, E.K., 1987. Psychological aspects of genetic counseling. IV. The subjective assessment of probability. *American journal of medical genetics*, 28(2), pp.361–70.

McAllister, M., Payne, K., Macleod, R., Nicholls, S., Dian Donnai, et al., 2008. Patient empowerment in clinical genetics services. *Journal of health psychology*, 13(7), pp.895–905.

McAllister, M. et al., 2011. The Genetic Counseling Outcome Scale: a new patient-reported outcome measure for clinical genetics services. *Clinical genetics*, 79(5), pp.413–24.

McAllister, M., Payne, K., Macleod, R., Nicholls, S., Donnai, D., et al., 2008. What process attributes of clinical genetics services could maximise patient benefits? *European journal of human genetics : EJHG*, 16(12), pp.1467–76.

McKusick, V., 2002. History of medical genetics. In *Principles and Practice of Medical Genetics*. pp. 3–36.

Oppewal, H., Louviere, J. & Timmermans, H., 2013. Processes Modeling Hierarchical Conjoint Experiments Integrated Choice. *American Marketing Association*, 31(1), pp.92–105.

Payne, K., Mcallister, M. & Davies, L.M., 2013. Valuing the economic benefits of complex interventions: When maximising health is not sufficient. *Health Economics*, 22, pp.258–271.

Payne, K. & Thompson, A., 2013. Economics of pharmacogenomics: rethinking beyond QALYs? *Pharmacogenomics*, In Press.

Peacock, S. et al., 2006. A discrete choice experiment of preferences for genetic counselling among Jewish women seeking cancer genetics services. *British Journal of Cancer*, 95(10), pp.1448–1453.

Regier, D.A. et al., 2009. Valuing the benefit of diagnostic testing for genetic causes of idiopathic developmental disability: willingness to pay from families of affected children. *Clinical genetics*, 75(6), pp.514–521.

Rungie, C.M., Coote, L. V. & Louviere, J.J., 2012. Latent variables in discrete choice experiments. *Journal of Choice Modelling*, 5(3), pp.145–156.

Ryan, M. & Farrar, S., 2000. Using conjoint analysis to elicit preferences for health care. *BMJ (Clinical research ed.)*, 320(7248), pp.1530–3.

Ryan, M. & Gerard, K., 2003. Using discrete choice experiments to value health care programmes: current practice and future research reflections. *Applied Health Economics and Health Policy*, 2(1), pp.55–64.

Skirton, H., 2001. The Client ' s Perspective of Genetic Counseling — A Grounded Theory Study. , 10(4).

Street, D. & Burgess, L., 2007. *The Construction of Optimal Stated Choice Experiments: Theory and Methods*, London: Wiley.

Appendix 1: *Future Analysis Plan*

Structural choice modelling (SCM) is a potentially method of analysis for data collected from a hierarchical CA with integrated DCE. In theory, CGS could be tailored to individuals belonging to a pre-identified preference segment in terms of (i) preferred information provision and (ii) counselling. SCM is based on a general econometric model which combines factors and structural equations (Walker 2001). This approach is consistent with random utility theory. The commonly used conditional logit and mixed logit (with random coefficients) specifications are special cases of this general model. In SCM a random coefficient choice model is specified such that the random coefficients are functions of factors and the factors may be functions of each other (Rungie et al 2012)[1]. When the SCM approach is combined with HII it will be possible to identify common factors underlying preference heterogeneity in different choice tasks.

Following approach of Rungie et al (2012) we propose a number of candidate models to fit the data:

1. The conditional logit. In this model coefficients are set as constants and there are no additional factors. This model does not provide any information on preference heterogeneity but is important to include as a comparison with subsequent models.
2. The mixed effects logit with random coefficients. The vector of coefficients has a unique value for each individual. The distributions of coefficient values are assumed to be independent and normal. Conclusions about preference heterogeneity can be made by considering the standard deviations of the coefficients, large standard deviations indicate substantial preference heterogeneity. The limitation of this specification in exploring preference heterogeneity lies in the assumption of independence of the distributions of the random coefficients. Correlations between attribute coefficients are not identified and we cannot attempt to define latent population 'segments'.
3. Factor models. These models provide structure to the covariance matrix of the random coefficients. Several specifications may be compared. Initially a factor model with a single factor would be considered. Factor loading would provide information on the degree to which preference heterogeneity in each attribute explains overall preference heterogeneity. Important research questions would relate to whether preference heterogeneity for the attribute "ability to make an informed decision" explains a high proportion of overall heterogeneity and how this is correlated with other attributes.

Model selection between conditional logit, mixed logit and factor models is made using goodness-of-fit statistics.

[1] For a complete mathematical explanation of SCM see Rungie et al 2012. It is not possible to recap this here due to page limits. Candidate models are therefore also described without reference to equations.