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VALUING THE ECONOMIC BENEFITS OF COMPLEX INTERVENTIONS: WHEN MAXIMISING HEALTH IS NOT SUFFICIENT

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ABSTRACT

Complex interventions, involving interlinked packages of care, challenge the application of current methods of economic evaluation that focus on measuring only health gain. Complex interventions may be problematic on two levels. The complexity means the intervention may not fit into one of the current appraisal systems, and/or maximising health is not the only objective. This paper discusses the implications of a programme of work that focused on clinical genetics services, as an example of a complex intervention, and aimed to identify the following: the attributes that comprise both health and non-health aspects of benefits and whether it is possible to evaluate such an intervention using current National Institute for Health and Clinical Excellence appraisal processes. Genetic services and tests are a good example of a complex intervention and have broader objectives than just health gain, which may usefully be measured using the concept related to capability, which we have called 'empowerment'. Further methodological work is required to identify the trade-off between non-health (empowerment) and health benefits for other complex interventions. We do not advocate a move away from QALY maximisation but do suggest that there is a need for a more considered approach that can take account of the perceived value for non-health attributes for some complex interventions. Copyright © 2012 John Wiley & Sons, Ltd.

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1. INTRODUCTION

All healthcare interventions are complex, but some can be viewed as being more 'complex' than others. However, such different levels of complexity are not generally acknowledged in currently applied methods of economic evaluation used to inform the allocation of scarce healthcare resources. The UK Medical Research council (MRC) guidance distinguishes between 'simple' and 'complex' interventions to inform how best to evaluate the benefits and costs of interventions (MRC, 2008). Simple interventions are typically single technologies, such as a pharmaceutical, that have a single objective. MRC guidance describes a complex intervention as involving one or more of the following aspects: more than one interacting component; numerous and difficult behaviours required by those delivering (or receiving) the intervention; more than one group, or organisational level, that is targeted by the intervention; and numerous and variable outcomes and a degree of flexibility or tailoring of the intervention is allowed (MRC, 2008). In practice, there are many examples of complex interventions. One example is complex structured drug treatment packages for problem drug users, provided through the UK NHS, which have multiple interacting component, outcomes valued by the drug user (improvements in own health and well-being) and by society (behaviour change of the drug user, e.g. crime reduction/risk taking behaviour). This complex intervention has potential to impact improved choice, access and participation in

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healthcare, education, housing, employment and social life (Godfrey *et al*., 2004, Davies *et al*., 2009). Clinical genetic services also are a good example of a complex intervention, which offer interlinked packages of care involving the following: clinical, molecular and cytogenetic-based diagnoses; risk estimation; non-directive counselling; and expert advice/training to healthcare professionals. Clinical genetic services are integral to most healthcare systems, including the British National Health Service (NHS). Genetics services often have a co-ordinating role and refer patients to other services offering health and social care and support but rarely provide treatment themselves. 'Patients' in clinical genetics are not just one individual but include other family members who are healthy but at risk of developing or transmitting a condition. The implicit value of genetic services and tests includes the individual's and their family's valuations of gaining knowledge about the diagnosis, prognosis and risk of having a condition, which supports future decision making, both medically (treatment choices) and personally (reproductive and other lifestyle, choices).

Some healthcare services and complex interventions provided by the NHS seem to provide a broader range of benefits than health gain (Long *et al*., 2008; Byrne *et al*., 2010). If this is the case, and the relative importance of health is low for some services, it is necessary to formally identify what patients and citizens fundamentally want from their health service. However, such non-health attributes often are not well described. Drummond *et al*. (2008) suggest that it is timely, given current policy developments in health, to think more broadly about the costs and benefits of interventions. This paper discusses the implications of the Valuation and Evaluation Research Theme (VERT), a programme of work to understand if it is reasonable to value health status alone, when measuring patient benefits from a complex intervention. We define reasonable in the sense that health status has some face validity and is a logical and reasoned measurement approach underpinned by empirical data. A secondary aim was to identify if it is possible to appraise complex healthcare interventions using some or all of the current NICE processes. The programme of work used clinical genetics as a case study. Here, clinical genetics include the genetic test technology and counselling interventions and comprise a complex intervention. Genetics often is viewed as being distinct from routine health care and the term 'genetic exceptionalism' is sometimes used. However, genetic services are similar to other types of complex interventions, which may need to take account of non-health in addition to health benefits.

2. CURRENT EVALUATIVE FRAMEWORKS

In UK health policy terms, it is argued that the age of the economic evaluation and the incremental cost effectiveness ratio (ICER) has arrived. The Department of Health policy focuses on a combined health, public health and social care agenda in the UK. The National Institute for Health and Clinical Excellence (NICE) now leads the appraisal and assessment of healthcare technologies and public health interventions and programmes. Guidance for technologies and public health services go through discrete processes, informed by separate method guides for technology appraisal (NICE, 2008) and public health (NICE, 2009a). Separate processes are used for clinical guidelines, interventional procedures and, now in pilot phase, diagnostics. It is assumed that all healthcare interventions fit neatly into one of the guidance 'processes'. With the exception of interventional procedures (defined as invasive diagnostic procedures and appraised in terms of safety and efficacy), the measure of health effects reported in the NICE reference case for appraisal is the quality adjusted life year (QALY) (NICE, 2008).

The underlying assumption for technology appraisal is that all healthcare interventions funded by a healthcare system should improve health status, quantified as a composite index measure of the number of remaining years of life adjusted by their quality. This generic measure theoretically allows comparison across all healthcare technologies or interventions. Health status is generally defined by a multi-attribute utility function such as the five outcome domains of mobility, self-care, usual activities, pain/discomfort and anxiety/depression in the EQ-5D (measures of health-related function). Preferences for health states should be valued by an appropriate group of society (NICE, 2008).

The public health guidance is more pragmatic in its approach compared with the guide for technology appraisals. This includes using alternative outcome measures (such as life years gained, cases averted or a more

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disease-specific outcome) and cost consequences analyses (Chalkidou *et al*. 2008; NICE, 2009a). Even so, there is still an underlying assumption that the goal of the intervention is to improve or maximise health status and health-related function (as illustrated by the recommendation to use the EQ-5D for the estimation of QALYs).

The procedures and processes set up by NICE generally do work well and are internationally renowned for producing robust guidance in technology appraisals, public health and clinical guidelines. It is not clear what process should be used if the intervention funded by the healthcare system does not meet the criteria for appraisal as either a technology or public health programme. Public health programmes are described as complex interventions in the methods guide. Some healthcare interventions that are not deemed to be public health interventions also are best described as complex interventions involving a number of discrete, but interlinked, components. Clinical genetic services are not public health interventions but are specialist services providing complex interventions (MRC, 2008). Some aspects of these services, for example, a genetic diagnostic test could be appraised under the technology appraisal process. Using this sequential, discrete appraisal process assumes that the benefits from each of the component parts of the service can be summed to quantify the benefit for the whole service. For complex interventions, it cannot generally be assumed that the whole is the sum of the parts. Furthermore, if one aspect of the service, such as the genetic test, can fit into one of the appraisal systems, NICE will then assume that the objective should be to maximise QALYs (NICE, 2008).

3. LIMITATIONS WITH CURRENT EVALUATIVE FRAMEWORKS?

The QALY approach is not always viewed as ideal, and there are numerous debates in the literature focussing on issues of theoretical validity, measurement methodology and the ethics of using them to inform health policy decisions (see Coast *et al*., 2008a or Dolan, 2008, for examples). Other approaches are available. These include construction of a contingent valuation market and use of willingness-to-pay to quantify a monetary benefit for use in cost–benefit analysis (Smith, 2003) or discrete choice experiments to quantify the trade-offs between health, non-health and process attributes (de Bekker-Grob *et al*., 2012). These methods, and their application, are described in the health economics literature but not currently advocated, or being used, in UK health policy decisions. The possible consequences of using inappropriate or incomplete measurements of health gain for some interventions, such as genetic screening and testing, can produce misleading findings for decision makers (Mooney and Lange, 1993; Hall *et al*., 1998; Grosse *et al*., 2008). Mooney (1994) directly questions the assumption that patients only want to gain health from their use of health services, suggesting the relative importance of health in a person's utility function depends on the type of service. To date, there is no published empirical evidence to identify whether QALYs systematically under-value aspects of services, such as complex clinical genetics services, by emphasising health outcomes and excluding other aspects of patient benefit.

There is evidence that users of health care, and society, more generally, expect and demand broader benefits from health care than health function (Coast *et al*., 2008a, 2008b). These require the extension of outcome measures to include the value of the process of delivering health care, non-health outcomes and capability to participate equally in life. There is conflicting evidence about the importance of process utility, defined as the satisfaction from the process of care (Birch *et al*., 2003; Donaldson and Shackley, 1997). A recent review of service user priorities and preferences for the outcomes of treatment for psychosis (Byrne *et al*., 2010) included improved social activity and inclusion; improved functional ability and participation in life; independence, confidence and empowerment; and improved ability to manage as well as more traditional functional outcomes associated with symptom control and management. Coast *et al*. (2008a) also use examples such as a need to feel safe or retain dignity and self-respect and suggest that these broader perspectives of gain can be discussed in the context of the capabilities approach first proposed by Sen (Sen, 1993). Sen (1993) was not prescriptive in how he defined the components of capability. Furthermore, Sen viewed the capability concept as something that should be distributed across society rather than something to be maximised, making it fundamentally different to the QALY maximisation approach. To date, capability has been implemented in

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the context of valuing health benefits for older people using the ICECAP measure (Coast *et al*., 2008c) and also people in chronic pain (Kinghorn, *et al*., 2007). It has been suggested as a useful measure for public health interventions (Lorgelly *et al*., 2010). The capability approach aims to measure the extent to which a person has the ability to function (capability) if they choose to. Coast *et al*. (2008b) cautiously advocate the use of the capability approach in health economics, which suggests a move away from utility as the metric, because it may prove useful for evaluating health promotion and public health.

4. CLINICAL GENETICS SERVICES: A COMPLEX INTERVENTION

Clinical genetic services cover all core genetic activities including population screening programmes (DH, 2010). The current focus is on single gene disorders in which there is a clear association with a particular gene (the person's genotype) and the expression of harmful characteristics in that individual (their phenotype). There are conditions with more complex genetic associations in which multiple genes result in the expression of an affected phenotype, for example, learning disabilities. Genetic tests are sometimes offered to patients for diagnosis, predictive or carrier testing, but they are not available for all genetic conditions, and patients may not always want to have a genetic test.

In the UK, clinical genetic services and associated tests are currently funded from a separate budget for specialised services with different commissioning arrangements to other NHS services. The level of funding is not known. However, they do compete directly with other healthcare services for scarce resources with, as yet unidentified, opportunity costs attached to diverting funding to them. The future is likely to see an expansion of the service from single gene disorders to more complex conditions with the advent of techniques such as high-throughput sequencing (more than one gene identified at a time) and whole genome sequencing (the complete genome examined rather than just a short section as achieved by current genetic tests). The use of these techniques has the potential to detect 'incidental findings', providing information on the condition being considered and could reveal the chance of having, or developing, currently un-related (and unknown) conditions. These developments offer the prospect of additional information to guide clinical decision making, potentially leading to innovative management strategies and new pathways of care. However, they also could cause psychological harm by revealing incidental findings. In this climate of expansion in clinical genetic services, and potential impact on NHS resources, it is essential to evaluate the impact of new genetics-related services, including the genetic tests, on patients, their families and service provision.

5. ECONOMIC EVIDENCE FOR CLINICAL GENETICS SERVICES

There are limited numbers of economic evaluations of genetics *services*, and these have presented cost–consequences analysis (Torrance *et al*., 2006; Brain *et al*., 2000). The examples have evaluated models of genetic service delivery for inherited forms of breast cancer and used psychosocial outcomes such as general anxiety, perceived risk, breast cancer worry and knowledge of familial breast cancer, rather than valuing a composite measure of health gain, such as the QALY. The majority of previous economic evaluations in genetics have tended to focus on the genetic *test* rather than the *service* Hall *et al*. (1998). The outcomes used are summarised in subsequent systematic reviews (Carlson *et al*., 2005; Jarrett and Mugford, 2006; Rogowski, 2006; Rogowski, 2007). These evaluations focussed on cancer (breast, ovarian or colorectal), neonatal and prenatal screening for cystic fibrosis and Downs syndrome, hereditary haemachromotosis and familial hypercholesterolaemia. Most evaluations were cost-effectiveness analyses using outcomes such as the number of cases detected, life-years gained and, in a limited number of instances, QALYs. Termination of the 'affected' pregnancy was sometimes used in the evaluation of prenatal screening programs (Piggott *et al*., 1994; Chamberlain, 1978; Sadovnick and Baird, 1981) and measured as the number of cases avoided (Cuckle *et al*., 1995) to estimate a cost per affected pregnancy detected.

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This focus on genetic tests means that evaluations have generally not considered the potential role of the service or clinician providing the counselling information in the supply of genetic (or DNA-based) testing and related diagnostic services for genetic conditions. It is important to broaden the focus in economic evaluations for two reasons. The benefit of the test is inextricably linked to that of the service (Payne, 2009). Genetic services use non-directive counselling to help the patient and family to understand the genetic condition and make informed choices themselves. Even if a test is an integral component of a particular genetic service, such as testing for Huntington disease in a family, testing does not always become part of the counselling process. In some instances, an individual, or the family, may decide not to pursue the testing option. The counselling process can have a potentially strong influence and determine whether benefit is gained by the patient from having/not having the test. Importantly, testing is not always possible. This may change in the future as high-throughput sequencing, and whole genome sequencing become available. At present, the clinical genetics service involves family history taking to identify modes of inheritance of a condition to establish, or aid, a clinical diagnosis. Even if genetic testing is not possible, the family can still be given risk information to help their decision making. Counselling interventions are offered to help the family adjust to this new information. This process fits the capability paradigm, as the service aims to enhance the patients'/families' capability to make an informed choice and provide freedom of choice, a key aspect of outcome proposed by Sen (Sen, 1993). However, it focuses on capability in a behavioural rather than functional sense.

Hall *et al*. (1998) present the value of information from genetic testing as the key outcome of interest, which is relevant to the individual who provides DNA for the test and other family members, the unborn/aborted foetus, and could affect social relationships and interactions (Hall *et al*., 1998). They concluded that current evaluations of genetic tests are not measuring the appropriate benefits for that intervention, and methodological improvements in the measurement of outcome are necessary. However, to date, no one has suggested an empirical approach that describes the nature of the benefits, from genetic services and tests, and how such benefits should be measured.

6. VALUING THE BENEFITS OF CLINICAL GENETICS SERVICES

The VERT programme of work was designed to collect empirical data using a range of methodological approaches. The overall aim was to define health and non-health benefits from clinical genetic services. Published outputs include the following: a systematic review to identify existing validated outcome measures used in genetics services (Payne *et al*., 2008); Delphi survey of users (n = 72) and providers (n = 115) of genetics services to identify the degree of consensus about the relevance of existing outcome measures (Payne *et al*., 2007) and qualitative research (focus groups and semi-structured interviews) to explore the outcomes valued by patients, patient representatives and service providers (n = 52) (McAllister *et al*., 2007a, 2007b; McAllister *et al*., 2008a, 2008b). The systematic review identified 67 outcome measures, which capture 19 outcome domains (see Appendix A). The majority were classed as subjective measures, with three objective (countable) measures of outcome. No measures to value the utility of genetic service outcomes were identified. A measure of health status, the SF-36, was included in one evaluation (Trask *et al*., 2001). The majority of outcome measures identified did not quantify health *per se* but focussed on the psychological aspects of counselling. A two-round Delphi survey, comprising definitions of these 19 outcome domains was posted to a panel of experts, (patients, patient support group members and genetics clinicians and counsellors), who rated each outcome domain as useful or not useful on a seven-point rating scale. The Delphi resulted in nine potentially useful outcome domains. These findings were then compared with the qualitative findings. The qualitative findings indicated the following: (i) measures of process and outcome are necessary to fully describe the benefits of a clinical genetics service; (ii) process utility is a useful concept to consider; and (iii) five important process attributes were identified to be inextricably linked with outcome (Figure 1). Furthermore, rather than simply representing separate components of a utility function, the process attributes are potential moderators of the benefit a patient or family may derive from a clinical genetics service. Ten emotional effects and 11 social or societal effects of

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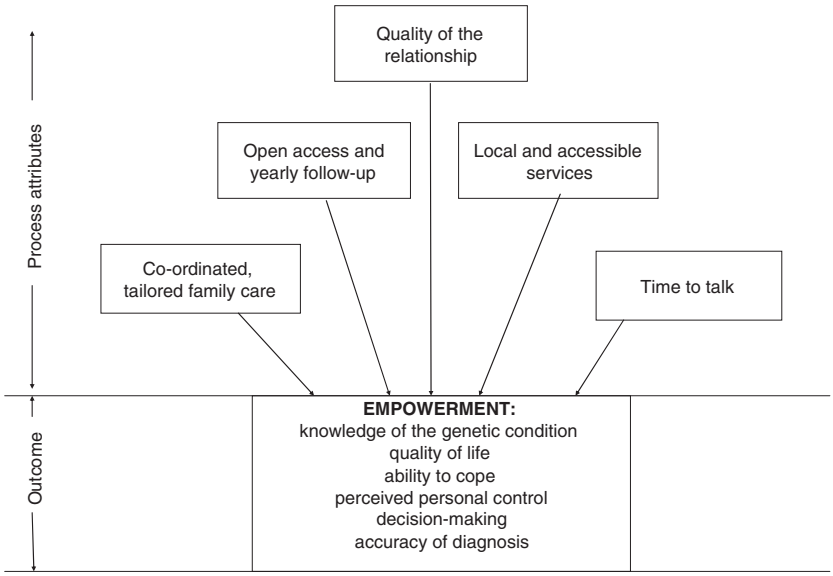

Figure 1. Components of the clinical genetics service utility function

genetic diseases on individuals and families also were identified. It was clear from the findings of the qualitative aspects of this study that patients' value genetic risk information both for themselves and for their relatives and future generations. Indeed, some patients seemed to value the potential benefits of this information to their children more highly than for themselves.

Comparing the findings of the qualitative research and Delphi survey produced six outcome domains considered useful and relevant (Figure 1). Health status was not rated as one of these, although a broader quality of life domain (see Appendix A for a definition) was perceived as useful and relevant. A unifying concept of empowerment was developed from the findings of the qualitative research. Grounded theory was used to develop an inductive model or explanatory theory 'grounded' in the data (Glaser and Strauss, 1967). This is a progressively focused analytic approach, to identify a 'core category'. This is a theme or concept that emerges from the analysis as the central phenomenon, around which all other themes are integrated (Strauss and Corbin, 1990). Empowerment in this analysis describes the patient benefits from using genetic services. In relation to clinical genetics services, it is '*a set of beliefs to enable a person affected by a genetic condition, or at risk for developing or transmitting a genetic condition, to feel that they have some degree of control over and hope for the future*' (McAllister *et al*., 2008b).

The VERT programme suggested that multiple objectives were desired, which include maximising non-health benefits (Payne *et al*., 2008; Payne *et al*., 2007; McAllister *et al*., 2007a, 2007b; McAllister *et al*., 2008a, 2008b). These findings challenge the appropriateness of current healthcare assessment and appraisal processes that focus on health gain as the only priority for patients and healthcare professionals. The multiple goals of clinical genetics services are not consistent with the single goal of maximising health status in isolation. Additionally, the overriding concept that individuals seek to maximise when using clinical genetics services is empowerment.

Empowerment, in the context of genetic services, was defined explicitly as being able to cope with having a genetic condition in the family and includes a component of being able to look at the future with hope (future orientation). This concept underpins other complex interventions and the priorities of people using healthcare services for mental health (Byrne *et al*., 2010), problem drug use (Godfrey *et al*., 2004; Davies *et al*., 2009), communication problems following stroke (Long *et al*., 2008) and other chronic diseases. Additionally, the concept has analogies with people at risk of conditions that have a less obvious genetic component, such as risk of cardiovascular disease or diabetes. Empowerment can be viewed as a type of capability that measures the

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'ability of a person to function' (Coast *et al*., 2008a). The current applications of the capability concept focus on physical aspects (Coast *et al*., 2008c; Kinghorn, *et al*., 2007; Lorgelly *et al*., 2010). Empowerment focusses on the capability to make informed decisions relating to the genetic condition but not limited to health treatment decisions and incorporates aspects of being able to cope with the future. Empowerment is potentially relevant to all healthcare decision making and, as an attribute, could theoretically replace, or supplement, health status in the utility function for a complex intervention. However, the VERT programme only described the potential non-health attributes to be considered using one example of a complex intervention: clinical genetic services. Before recommending that health is replaced, or supplemented, by empowerment, the next key step is to identify how people trade off between empowerment and health objectives. The relevant importance of empowerment compared with health may well differ between conditions and between complex interventions. Further research on identifying such trade-offs is necessary.

7. A NEW EVALUATIVE FRAMEWORK?

A vital first stage in the evaluation of any public programme is to be clear about its objectives. This means it is then possible to be explicit about which benefits should be valued. The UK Treasury suggests willingness to pay (or accept) as the preferred measure of benefit to evaluate non-health public programmes (HM Treasury, 2001). In contrast, economic evaluation of healthcare interventions uses measures of health gain, with the QALY recommended by many as a metric. This is relevant if the stated objective of a healthcare intervention is to maximise (health) benefits subject to the resource constraint, which is the NHS budget. Existing decision rules and guidelines are unclear about how to proceed if the majority of the benefits fall outside this benefit maximisation and budget constraint.

A key question then is how best to produce guidance for the effective and cost-effective use of complex healthcare interventions that do not fit into the current NICE appraisal processes. Genetic tests, as one component of a complex intervention, could potentially fall under the remit of the pilot NICE Diagnostics Assessment Programme (DAP). However, two issues arise. Almost 99% of genetic tests do not fall within the focus of the NICE DAP because they are not formally licensed by a regulatory body (Payne, 2009). More importantly, the current methods guide for diagnostics is unclear about how to measure outcomes. The current methods guide refers to the existing NICE Reference Case for valuing outcomes, adding that the EQ-5D might not be sensitive enough to detect the impact of using a diagnostic and to understand the psychological consequences (NICE, 2010). A new five-level version of the EQ-5D is available to increase sensitivity and reducing ceiling effects of the current three-level version (Herdman *et al*., 2011), but the five domains within the new EQ-5D-5 L remain the same. Other multi-attribute health utility indices, such as the Health Utilities Index Mark 2 (HUI-2) or Mark 3 (HUI-3), could be considered as options to improve sensitivity (Horsman *et al*., 2003). However, using the EQ-5D-5 L, HUI-2 or HUI-3 maintains the focus on 'health status' and may still not adequately capture the impact of complex interventions on informed decision making or empowerment.

So how can the evaluative framework for complex interventions, such as clinical genetics services, be developed? One option is to take the (perhaps) defeatist attitude that is it just too hard to develop methods that robustly enumerate the benefits and costs from clinical genetics services and similar complex interventions. This leaves them to be funded (or not) without any stringent forms of evaluation. Such an approach does not support decision makers who have to make difficult decisions about resource allocation. Option 2 is to fund such interventions outside the healthcare service and view them as non-health public sector services. This means that valuation methods, such as willingness to pay, could be used (HM Treasury, 2001). Such an approach may be practically difficult because clinical genetics services and many other examples of complex interventions with a potential impact on informed decision making are integral and funded components of the NHS, with a conceptual health base. It also would make such complex interventions both theoretically and practically non-comparable with other healthcare technologies because using money as a valuation metric assumes a welfarist perspective.

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An option to keep clinical genetics services comparable with other services is to supplement the existing evaluative frameworks and valuation metrics of health status (the EQ-5D weighted by public values) with guidance on the use of patient-reported outcome measures for a subset of specified procedures (DH, 2008). In this approach, maximisation of health status remains an objective for genetic services that involve referral to an interventional treatment. For example, individuals with an inherited condition affecting the heart (hypertrophic cardiomyopathy) can be offered genetic testing and management as part of a clinical genetics service. Some individuals will have a significantly increased risk of sudden cardiac death, in which case an implantable cardioverter defibrillator can be inserted to correct the life-threatening arrhythmias. This intervention has clear benefits, measurable using the QALY metric of additional quantity and quality of life. Similar gains in quantity and quality of life can be seen for *BRCA1/2* testing to identify women at increased risk of inherited forms of breast cancer, which can allow them to have preventive and potentially life-saving surgery or intensive screening, which can detect cancer at an early, treatable stage. However, this approach ignores the implications of empirical evidence that patients and healthcare professionals do not perceive the QALY metric as useful for the vast majority of current clinical genetics services, which do not directly aim to improve health or extend life. This means any resulting ICER comparing different models of services or testing for genetic conditions, single or multiple-gene, is likely to have a denominator that tends to zero and an ICER that tends toward infinity, if measured by the EQ-5D.

A further related issue is whether the ICER is an acceptable value below a pre-defined threshold that represents 'the implicit social valuation of health' Claxton (2007). In the UK, NICE uses a range of incremental cost per QALY gained thresholds. Special reasons are needed to recommend interventions that exceed this threshold (NICE, 2008, 2009b). Because the QALY ignores key benefits, it is unlikely that being able to raise the threshold incremental cost per QALY under specially defined circumstances would be sufficient to ensure that genetic tests or services are funded. The ICER threshold, as currently measured, only provides a decision maker with the 'social valuation of health' and excludes non-health attributes. This raises a potential problem, given that clinical genetics services are currently funded from the health budget and not the non-health budget.

A final option is to define a sub-set of services to evaluate with a modified framework using different valuation metrics. There is already a precedent with different NICE appraisal processes for technologies compared with public health programmes. One consideration is the extent to which objective criteria can be defined to identify a programme as eligible for evaluation under a complex intervention remit rather than as a single technology. Further research is needed to introduce empowerment as a valuation metric to supplement (or replace) the QALY.

A number of potential measures of empowerment are available for use in health care [Anderson *et al*., 2000; Rogers *et al*., 1997; Bulsara *et al*., 2006; Hibbard *et al*., 2005] but do not use either a consistent definition or conceptualisation of empowerment [Herbert *et al*., 2009]. A measure of empowerment has been developed for clinical genetics services (McAllister *et al*., 2011a, 2011b). The availability of multiple measures of empowerment introduces the first challenge for researchers who must, therefore, generate empirical evidence to support which domains, and associated empowerment measure, are necessary and sufficient to compare across different complex interventions. Once the appropriate measure of empowerment has been established, a useful methodological approach is to measure both health status and empowerment in economic evaluations. This enables comparisons with past and current analyses and consideration of whether it is appropriate to incorporate other valuation methods in the future (Lipscomb *et al*., 2009; Drummond *et al*., 2008).

8. CONCLUSION

Clinical genetics services, and other complex interventions, compete for healthcare resources with interventions that more clearly aim to maximise health gain. Genetics services have evolved to be an integral part of the UK NHS but pose special challenges for robust economic evaluations of the technology component (genetic tests) and also the service component (genetic counselling). An empirical research programme (VERT) demonstrated

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that genetic services and tests have broader objectives than just health gain. Patients and healthcare professionals defined the stated objective for the majority of genetic services and associated genetic tests as empowerment of patients rather than simply improvement in health status. Empowerment is a feature of other complex and behavioural interventions in health care and is consistent with the Department of Health's strategic objectives. Research is required to identify an empowerment state descriptive system and apply an appropriate metric to value each empowerment state. This methodological work will support empirical economic evaluations, which, in the early stages, use a health status valuation method, such as the EQ-5D, alongside the empowerment status valuation method. We do not advocate a move away from QALY maximisation, which is a sufficient and useful method for evaluating the majority of competing healthcare interventions that aim to improve health alone. We do suggest that there is a need for a more considered approach taking account of the findings from this study that support the perceived value of non-health attributes. This concurs with the conclusions of Claxton *et al*., 2007 that maximising health gain is not a sufficient objective to achieve once costs and benefits outside the healthcare sector are recognised. Such a considered approach requires collecting empirical data with concurrent use of alternative outcome measures, to identify the balance between health and non-health gains for interventions that do not aim to cure but offer people adjustment and hope for the future for incurable conditions, which cannot be treated using pharmacological or surgical interventions.

APPENDIX A: IDENTIFIED DOMAINS WITH THEIR DESCRIPTION AND EXAMPLES OF OUTCOME MEASURES

Domain	Description Used in Delphi	Examples of Outcome Measure Containing the Domain	Primary Source of Ouctome Measure
Knowledge of the genetic condition	Knowledge about the genetic condition in the person who attended the clinic. The aspects of knowledge measured include knowledge about the risks of the condition to them and other members of their family.	Breast Cancer Genetic Counselling Knowledge Questionnaire Genetic Knowledge Index Knowledge about genetic risk for breast cancer	Erblich *et al*. (2005) Furr and Kelly (1999) Donovan and Tucker (2000)
Anxiety	Whether using the service had changed how anxious the person felt.	Cancer Anxiety and Helplessness Scale General Health Questionnaire Hopkins Symptom Checklist	Kash *et al*. (1992) Goldberg and Williams (1988) Derogatis *et al*. (1974)
Depression	Whether using the service had changed how depressed the person felt.	Beck Depression Inventory Hospital Anxiety and Depression Scale Self-Rating Depression Scale	Beck *et al*. (1988) Zigmond and Snaith (1983) Zung (1986)
Worry	Whether using the service had changed how worried the person felt.	Worry Interference Scale Breast Cancer Worry	Trask *et al*. (2001) Lerman *et al*. (1991)
Health status	Whether using the service had changed the health of the patient. The aspects of health measured include ability to care for oneself, ability to perform day-to-day activities, pain, ability to get around, anxiety and depression.	Medical Outcomes Short-Form Survey (SF-36; SF-12)	Ware (1993)
Quality of life	Whether using the service had changed the quality of life of the patient. The aspects of quality of life measured include physical and mental well-being, social and family relationships and attitudes to the future.	Subjective Quality of Life Profile Functional Assessment of Cancer Therapy-General	Dazord (1995) Cella *et al*. (1993)

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APPENDIX A. (*Continued*)

Self esteem	Whether using the service had changed the self-esteem of the patient. The aspects of self-esteem measured include feelings about body image and relationships with family and other people.	Rosenberg Self-Esteem Scale	Rosenberg (1965)
Family environment	Whether using the service had changed the family environment of the patient. The aspects of family environment measured include closeness, communication and relationships within the family.	Family Environment Scale	Moos and Moos (1994)
Spiritual well-being	Whether using the service had changed the spiritual well-being of the patient.	Spiritual Well-Being Scale Subjective Quality of Life Profile	Ellison and Smith (1991) Dazord (1995)
Coping	Whether using the service had changed whether the patient felt able to cope with living with a genetic condition.	Monitoring Blunting Style Scale Psychological Adaptation to Genetic Information Scale Utrecht Coping List	Miller (1987) Read *et al*. (2005) Westbrook (1979)
Mood	Whether using the service had changed the mood of the patient. The aspects of mood measured include tension, anxiety, depression, dejection, anger, hostility, vitality, fatigue, inability to motivate, confusion and bewilderment.	Profile of Mood State Beck Depression Inventory Breast Cancer Worry	McNair *et al*. (1981) Beck *et al*. (1988) Lerman *et al*. (1991b)
Satisfaction with service	How satisfied overall the patient was with the clinical genetics service	Genetic Counseling Satisfaction Scale Patient Satisfaction with Genetic Counselling Satisfaction with Decision Scale	Tercyak *et al*. (2001) Shiloh (1990) Holmes-Rovner *et al*. (1996)
Meeting of expectations	Whether the experience of using a clinical genetics service achieved what the person using the service thought they wanted.	Prostate cancer genetic screening survey	Doukas (2004)
Perceived personal control	Whether using the service had changed how much control the patient felt they had over the situation. It assumes that people who feel in control are better able to adjust to the genetic condition in their family.	Perceived personal control	Berkenstadt *et al*. (1999)
Decision making	Whether the use of the service had changed the patient's ability to make decisions linked to the genetic condition in the family. The aspects of decision making measured include: how comfortable the patient feels about making decisions; whether the patient has the information to make the decision; whether the patient feels the service has supported them enough.	Decision Evaluation Scales Decision-making process	Stalmeier *et al*. (2005)

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Table. (*Continued*)

Domain	Description Used in Delphi	Examples of Outcome Measure Containing the Domain	Primary Source of Ouctome Measure
Perception of risk	The accuracy of a patient's risk perception and how they believe the risk relates to them personally.	Health Beliefs Model (screening and breast cancer)	Kash *et al*. (1992)
		Multidimensional Impact of Cancer Risk Assessment	Cella *et al*. (2002)
		Perceived Risk of Breast Cancer	Lerman *et al*. (1997)
Rate of terminated pregnancies*	How many times the information provided by a clinical genetics services resulted in a decision not to continue with a pregnancy.	Not appropriate	Not appropriate
Accuracy of tests *	How accurate the tests used by the clinical genetics service are.	Not appropriate	Not appropriate
Accuracy of diagnosis*	How accurate the diagnoses, using a clinical examination or test result, made by the clinical genetics service are.	Not appropriate	Not appropriate

*Objective outcome measure.

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CONFLICT OF INTEREST

The authors declare no conflict of interest.

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