European Journal of Human Genetics (2014) 22, 25–31

www.nature.com/ejhg

ARTICLE

Criteria for fairly allocating scarce health-care resources to genetic tests: which matter most?

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The use of genetic tests is expanding rapidly. Given limited health-care budgets throughout Europe and few national coverage decisions specifically for genetic tests, decisions about allocating scarce resources to genetic tests are frequently *ad hoc* and left to lower-level decision makers. This study assesses substantive ethical and economic criteria to prioritize genetic services in a reasonable and fair manner. Principles for allocating health-care resources can be classified into four categories: need-based allocation; maximizing total benefits; treating people equally; and promoting and rewarding social usefulness. In the face of scarcity, the degree of an individual's need for medical intervention is an important criterion. Also, different economic concepts of efficiency are of relevance in the theory and practice of prioritizing genetic tests. Equity concerns are most likely to be relevant in terms of avoiding undesirable inequities, which may also set boundaries to the use of efficiency as a prioritization criterion. The aim of promoting and rewarding social usefulness is unlikely to be relevant to the question of what priority a genetic test should have in clinical practice. Further work is needed to select an appropriate set of criteria; operationalize them; and assign weights before some kind of standardized priority information can be added to information sources for genetic services. Besides the substantive criteria, formal considerations like those pointed out in the framework of accountability for reasonableness need to be considered in decision making.
European Journal of Human Genetics (2014) **22,** 25–31; doi:10.1038/ejhg.2013.172; published online 7 August 2013

Keywords: health-care prioritization; genetic testing; costs and cost analysis; resource allocation; ethics

INTRODUCTION

The availability of new genetic tests as well as their use in health care is expanding rapidly.[1,2] In spite of technical improvements leading to steadily lowering laboratory costs per test,[3] the costs of bioinformatic evaluation, counselling and follow-up testing could easily outweigh the potential savings from early prevention and lead to an overall increase in health-care expenditures.[4,5] For example, it has been demonstrated that it is not economically feasible to conduct cascade testing for all monogenic disorders for which tests are available.[6] Also, a recent survey of Canadian health-care providers reported insufficient resources to fund all genetic tests that were considered desirable.[7]

If resources for health services are limited, there is a large risk that limits are set unfairly. It is well known that individuals with higher socioeconomic status experience longer lives and better health-related quality of life across Europe, as well as have better access to specialist health care.[8,9] It is also likely that individuals with higher socioeconomic status are better informed about options for genetic testing; better at arguing their case with their general practitioner or when seeking a geneticist appointment; and more able to fund tests out of pocket. Although for many things in life such as expensive cars or other luxury goods, it is widely accepted that societal allocation differs by income, for important health-care services, this is frequently considered unfair.

Fair allocation of health-care resources requires that tests are allocated according to ethically reflected criteria.[10] If resources are not sufficient to fund all desirable tests, there is a need to determine which tests are most important to provide. This is referred to as explicit 'prioritization', that is, ranking genetic tests based on their perceived importance.[11]

Frequently, decisions about health-care coverage and priority setting are associated with formal processes of national decision-making bodies. For example, the UK National Institute of Health and Clinical Excellence (NICE) recently implemented a 'Diagnostics Assessment Programme'. However, most health-care funding decisions are made in a less formalized manner at lower decision levels such as health-care organizations.[12] Adair and co-workers[7] report that most Canadian decisions on which genetic tests would be covered were local level *ad hoc* decisions and concluded that a more coordinated approach would be desirable. The development of standards for prioritization on a local and regional level could also be beneficial in Europe.

The appropriate criteria for prioritization of genetic tests may differ by clinical condition or application. The successful prioritization activities in Sweden include 'Defining the area of prioritization'.[13] Prioritization activities have been more successful in Sweden within clinical specialties than across clinical areas.[14] An approach specifically targeted at genetic tests may therefore be particularly promising.

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Received 21 December 2012; revised 29 May 2013; accepted 19 June 2013; published online 7 August 2013

The challenge for prioritizing genetic tests is to develop criteria that have a firm ethical basis, are sufficiently specific for genetic tests and that can be operationalized, measured and consistently applied across a variety of genetic tests and clinical applications.

This study provides an overview of ethical and economic concepts for prioritizing scarce health-care resources toward genetic tests. The study focuses on human genetic services that can be provided in potentially different local organizational contexts. The long-term aim is to develop structured prioritization information, which could be used to complement standardized information sources about genetic tests such the Clinical Utility Gene Cards published by the *EJHG*[15] or the Gene Dossiers developed by the UK Genetic Testing Network (UKGTN).

CONCEPTS AND METHODS

A wide range of definitions of genetic testing appear in international recommendations, guidelines and reports.[16] Genetic testing is defined here as the application of a laboratory test or assay (the analysis of human DNA, RNA, chromosomes, proteins or certain metabolites to detect alterations related to a heritable disorder or to specific reactions to medical treatments (see http://www.genetests.org/servlet/access?id=8888891&key=Wt-CocgvbZFre&fcn=y&fw=FNL-&filename=/concepts/primer/primerwhatistest.html (downloaded on 22 December 2009; pharmacogenetics included)) to a defined clinical context. A given genetic assay may have multiple applications, including diagnosis, population screening or cascade testing of family members. The prioritization of coverage of a given laboratory assay may differ depending on the specific type of clinical application.

The provision of genetic tests involves a range of ethical aspects such as questions of privacy, unnecessary worry or under what conditions a termination of pregnancy is ethically acceptable. Ethical considerations can provide important constraints to the practice and organization of genetics services. These issues cannot be addressed here. This paper is restricted to established genetics services that are considered by most observers to be ethically acceptable and to provide more good than harm. In particular, we exclude from this study carrier implantation, preimplantation and prenatal testing for the purpose of family planning as well as population screening. These tests involve a range of very specific ethical issues that have been discussed elsewhere.[17–20]

The study is based on a large number of exploratory literature searches complemented by a systematic search for existing prioritization frameworks (Supplementary Appendix available on request).

Criteria for allocating health-care resources to genetic tests

Prioritization of scarce health-care resources has been addressed by the health economic and ethical scientific communities for many years. Persad *et al*[21] classify normative criteria for allocating medical interventions into four categories: need-based allocation, benefit maximization, equity, and promotion and reward of social usefulness. The following sections discuss different normative frameworks for health-care prioritization that account for these criteria and their application to the use of available genetic tests. Table 1 provides an overview of the most relevant criteria, their rationales as well as examples for illustration.

Need-based allocation. As illustrated by the proverb 'A healthy person has many wishes, but the sick person has only one', 'health' is a good of specific importance. It is a prerequisite for the pursuit of happiness rather than one among a multitude of options to choose from. Therefore, medical need in terms of severity of disease and need for medical services to alleviate diseases are important criteria for decision making in the face of scarce health-care resources. However, the term 'need' in the context of prioritizing health services requires further specification. 'Need' can be defined as the gap between an actual state experienced by an individual and a norm that prescribes something desirable. Different attributes of need can be used to illustrate this gap in genetics.

Health-related need

A 'health-related need' constitutes a gap between actual and desirable health states and is independent of whether there is anything that can be done to reduce the gap. In the 'fair innings' approach, measuring this gap involves a comparison of the total health individuals experience over their lifespan with an average amount they could have expected. This can relate to the life expectancy at birth, or to life expectancy adjusted by a weight for health-related quality of life.[22]

A second example of health need, which is likely to be relevant independent from whether effective treatment is available, is the *a priori* probability of having the disease. According to this dimension of need, first-degree relatives of a known mutation carrier may be considered to have a higher need for genetic testing than individuals in an average population. Also, high-risk individuals from population groups where a hereditary condition is relatively common may be considered to have a higher need than do individuals from the average population.

Intervention need

Different from the health-related need, the concept of 'intervention need' requires that an intervention is available for which there is scientific evidence that it can reduce the gap between an actual and the desirable health state[23] (p 131ff). In a situation in which no effective medical treatment exists, no intervention need exists according to this definition.

Given that different notions of 'need' can lead to different orders of priority, the question arises of what type a benefit has to be to be considered meeting an intervention need. For example, if need is defined in terms of 'degree of ill health', it can be argued that a threat to life is the most severe form of ill health and that life-saving or life-prolonging interventions should therefore take priority over life-enhancing ones.[24] Taking Huntington disease (HD) as an example, according to this definition, there would be no intervention need for HD testing.

Need can also be interpreted in terms of 'immediacy of ill health', which includes reduced health-related quality of life. Untreatable disorders may incur substantial emotional distress and mental health challenges. In case these can be ameliorated effectively by psychosocial care, an intervention need may exist for mutation carriers even in the absence of a life-extending medical treatment. Intervention need can also be defined broadly as the 'potential to benefit from health care'.[24] The latter definition is of particular relevance for the use of genetic tests because like in the case of HD, the perceived benefits of genetic tests can extend beyond health outcomes: a patient at risk of HD can benefit in terms of information for making choices such as whether to start a family or not. Such non-health benefits can contribute much to the value patients derive from genetic testing.[25] Currently, a range of instruments are in development to demonstrate such other benefit and, thus, intervention need, within clinical studies.[26]

The role of 'need' in decision making

Generally, the proven ability of a medical technology to ameliorate intervention needs is likely to be the important criterion in explicit decision making about medical technology: most agencies for health technology assessment and for coverage decision-making assess the scientific evidence of a technology's effectiveness if explicit coverage decisions are made.[12]

Also for genetic testing, scientific evidence of intervention need appears to be the most frequently addressed criterion. A number of groups have developed evidence-based approaches to evaluating the benefits of specific genetic tests and weighing them against potential harms, for example, the Evaluation of Genomic Applications in Practice and Prevention (EGAPP) initiative in the United States of America.[27] Also, a survey about decision making about scarce resources for the use of genetic tests in the Canadian health care system reported that 'evidential basis' and 'availability of preventative strategies' were among the criteria for decision making.[7]

Apparently following the concept of intervention need, the Department of Health of New South Wales, Australia, distinguishes genetic tests of high and low priority based on their expected benefit. For example, diagnostic testing is assigned high priority 'When confirmation of a clinical diagnosis will lead to changes in management of an affected person' and low priority

Table 1 Substantive principles for prioritizing genetic tests

Category	*Criterion for prioritization*	*Rationale for the criterion*	*Example of priority order if criterion is applied*	*Methods*	*Comments*
Allocation according to greatest need	Health need, for example, fair innings	Those at risk of a severe disease if left untreated are worse off than those at risk of a mild condition, and thus have a stronger claim on scarce health-care resources	FDR screening test for HNPCC ranked above FDR screening test for hereditary periodontal disease because the health loss associated with HNPCC is larger	Tools for describing and ranking different types of health and intervention need (few available)	Most important ethical claim Evidence appraisal widely accepted
	Intervention need	Claims on scarce health-care resources depend on, additional to health need, a medical intervention that can decrease the health need. Different types of benefit can be distinguished: reduction of mortality and morbidity or potential to benefit without tangible impact on health	FDR screening test for HNPCC ranked above FDR screening test for HD because for HNPCC, health can be improved by increased colonoscopic surveillance	Well-developed tools of evidence appraisal to establish intervention need	Limited possibilities to rank
Allocation to maximize benefit	Maximize health	A new genetic test consumes health-care resources, which alternatively would have been spent for other purposes in the health-care system. It should only be introduced if its health gains exceed the gains from the intervention forgone elsewhere	Genetic test is only reimbursed if the cost per QALY falls below a threshold value, which represents the price of the health gain forgone	Cost–utility analysis and other methods of health economic evaluation well developed	Used by some decision makers Limited sensitivity to other than health outcomes
	Maximize welfare	A new genetic test consumes resources that alternatively would have been spent for other purposes, for example, health-care, education or private consumption. It should only be introduced if its benefits, measured in terms of WTP, exceed the opportunity costs	Genetic test is only reimbursed if the mean willingness to pay exceeds the test costs (or funding is left to the market so that only those whose WTP exceeds the service costs receive the test)	CBA to measure WTP established method Markets to measure WTP practically	CBA rarely used by decision makers Markets may cause fairness concerns
Equitable allocation	Promote equality	Every citizen has equal claims on scarce health-care resources Problem to determine 'equality of what' – health, health-care spending, access to care according to equal need?	All of the above are of equal priority. If resources are not sufficient to fund all tests, a lottery decides The decision is based on cost per QALY, but gains to individuals in very bad health receive higher weight, which improves cost-effectiveness	Lotteries and random assignment (like for organs) Equity-weights in CUA (which still face methodological difficulties)	Lotteries ignore too much info to be used in practice Rather, allocate equitably according to the criteria above
	Avoid inequity	As no conclusive concept of equity exists, legal or ethical boundaries define which types of inequality are unacceptable; efforts are made to avoid or overcome these	Predictive HH test is offered both to men and women even if cost-effectiveness is much worse in females to avoid sex discrimination	Methods to measure specific types of (in)equity	Boundaries to rather than tool for priority setting

Abbreviations: CBA, cost–benefit analysis; CUA, cost–utility analysis; FDR, first-degree relatives; HD, Huntington's disease; HH, hereditary haemochromatosis; HNPCC, hereditary nonpolyposis colorectal cancer; QALY, quality-adjusted life year; WTP, willingness to pay.

'Where the clinical diagnosis is confirmed by other means and genetic testing will not alter the patient's management or options' (http://www.health.nsw.gov.au/policies/gl/2007/pdf/GL2007_013.pdf (download on 19 January 2010)). A similar guidance has been adopted by the Human Genetics Society of Australia.

However, approaches that assess genetic tests for one disorder at a time have limitations for prioritizing all genetic tests. First, they typically only determine whether or not a test is needed but do not prioritize among recommended tests. Also, to address the problem of escalating health-care costs, this approach has limitations, for example, because 'no evidence of effectiveness' does not necessarily imply 'evidence of no effectiveness' and funding may not be sufficient to establish effectiveness of all available technologies[28] – particularly in the field of genetic testing.[27] Currently, a widespread application of this criterion would exclude far most of the genetic tests available and used in clinical practice because only few have been covered by evidence reviews.

Apart from intervention need/evidence of effectiveness, different notions of health-care need have been used as criteria for prioritization across Europe, alongside further criteria.[29] For example, in Norway the first report of the Lønning–Komitee gave highest priority to acute life-saving technologies and low priority to technologies that are likely to improve health and quality of life, but do not incur serious damage if they are withheld.[30] According to the prioritization guidelines of the Danish Ethics Council, 'need' should be taken into account, which was defined as the gravity and prognosis of the disease, urgency and capacity to benefit.[29] However, these additional criteria have hardly been operationalized and used in a structured, evidence-based manner and no such framework could be identified for genetic tests. Therefore, there is a need for methodological work before such a needs-based approach can be used in prioritization practice.

Benefit maximization. Purely need-based decision making can lead to counter-intuitive results. For example, although many would agree that a

patient at immediate risk of death should be treated before a second patient who is suffering from a non-lethal disease, few would agree that all available health-care resources should be devoted to emergency rescue medical services. The important question is how many resources should be allocated to different health and intervention needs and to what extent higher needs justify higher resource spending. The two major health economic schools of thought provide different points of orientation for answering this question.

Welfare economics

Welfare economics correspond with a view of patients as autonomous citizens and consumers whose welfare is to be maximized. The best judges of how much genetic tests contribute to welfare are assumed to be consumers who assess the expected value from accessing different types of tests. Consumers care about many things, not just health, and may be willing to trade off lower health gains for higher satisfaction of other desires. Whether scarce resources should be spent for a genetic test depends on whether the consumers' willingness to pay (WTP) exceeds the cost of the test. In theory, consumers choose a bundle of products that maximize their expected utility. The WTP for a genetic test measures the expected benefit relative to alternative choices. If adding another good to the bundle than a genetic test would provide more benefit for the same cost, the consumer should prefer buying that bundle.[31]

If microeconomic assumptions for perfectly competitive markets held, for example, perfect information, market sale of genetic tests would lead to an efficient and optimal provision of those genetic tests where WTP exceeds the cost. In the absence of well-functioning markets, as is the case in health care,[32] economists seek to simulate market outcomes by assessing consumer WTP values from stated preference questionnaires. The WTP values can then be compared in a cost–benefit analysis (CBA) to the costs associated with providing the tests[33] (p 292ff).

It has been suggested that CBA may be of particular relevance for the economic evaluation of genetic tests because much of the utility a patient derives from testing is personal rather than clinical: the purpose of many genetic tests is to assist patients in making decisions about how to plan their life rather than to guide clinical interventions.[25,34] According to the welfarist perspective, focusing just on health outcomes will likely understate the economic benefits of genetic testing.

Extra-welfarist economics

Critics of the welfarist perspective argue that normative principles other than individual preferences should be the starting point for assessing public resource spending.[35] Many health-care decision makers base their decisions on aggregate health outcomes[12] and may explicitly reject the idea that WTP should guide the allocation of scarce health-care resources.[36] Those frameworks that include benefit measures other than individual welfare are labelled 'extra-welfarist'.[36]

One particularly influential approach is the model of a societal decision maker who spends scarce health-care resources to maximize health gains for the covered population. If the overall health-care budget is fixed, the opportunity cost of adding a new technology consists of the health foregone from reductions in other services that are displaced because of the need to reallocate scarce resources currently spent on their provision. The health gain per additional euro spent of these services forgone can be used as a threshold to decide whether a new service should be funded.[37] Health economic evaluation provides a tool to assess the value of novel health technologies through a comparison of the incremental cost-effectiveness ratio with the threshold value.

To consider simultaneously quality of life and mortality, outcomes can be expressed in terms of quality-adjusted life-years (QALYs).[38] In the calculation of QALYs, life-years are weighted by an index between one (representing full health) and zero (representing death). To assist decision makers in making rational decisions incorporating other criteria than health outcomes, methods of multicriteria decision making are available which can establish a priority score across various dimensions simultaneously.[39] It should be noted that the benefits and harms of genetic testing include not just the impact on the individual being tested but the health effects for family members as a result of cascade screening.[34]

The more severe a disease, the higher the QALY gains theoretically possible in case the disease can be cured completely. However, the concern for severity independent of potential health gains is not incorporated in QALY maximization models, even if tests for severe diseases without effective treatment are of particular importance for genetics. Finally, there are ethical concerns about benefit maximization for the allocation of scarce health resources because individual claims rather than measures of (aggregate) benefit may be a more adequate basis for priority setting in health care.[40]

The role of benefit maximization in decision making

Extra-welfarist economic evaluation is applied in decision-making procedures by the English NICE including the recently implemented 'Diagnostics Assessment Programme'. Health technologies are considered for inclusion or exclusion of the services provided by the United Kingdom's National Health Service, based on their cost-effectiveness[41] (with a 'weak' threshold area of between £20 000 and 30 000 per QALY). Recently, NICE issued a clinical guideline supporting genetic cascade screening for familial hypercholesterolemia, which concluded that genetic testing was the most cost-effective strategy, with an incremental cost-effectiveness ratio of £2700 per QALY.[42]

A framework for prioritizing genetic tests by Kroese and co-workers[43] for the United Kingdom appeared to be based on a benefit maximization approach, although it did not explicitly account for costs. A work group for the UKGTN successfully developed and piloted a multicriteria decision-making framework. Aiming to maximize the gains from limited resources, it was based on five dimensions of clinical utility: 'Reduction in morbidity and mortality', 'Information to provide reproductive choice', 'Improvement in the process of care', 'Deliverability of pathway of care' and 'Providing additional information not relevant to other criteria'. The criteria were measured for a set of genetic tests, weighted and a rank order measuring overall benefit was established.[43]

Evidence of cost-effectiveness is also considered by the respondents of the Canadian survey[7] as well as a range of other decision bodies but typically not as a sole criterion but alongside with other criteria.[44]

Benefit maximization in terms of individual WTP takes place any time a customer acquires a genetic test out of his or her own pocket in the direct-to-consumer testing market or if patients seek private appointments with geneticists. However, it is unclear whether test users and potentially affected relatives are sufficiently informed about the potential harms that can arise from genetic testing.[45,46] Also, it is unclear whether direct-to-consumer testing may result in high follow-up costs for health-care systems as individuals seek additional medical advice and further potentially unnecessary testing.[47]

Equity. Besides medical need and benefit maximization, also equity is a widely acknowledged criterion of fair prioritization.

Equity as a positive aim

Equity can be formulated as a positive aim, in the sense that some kind of equality is desired. However, it is not only difficult to define 'need' but also establishing a universally agreed definition of desirable 'equality', which should guide fair allocation of resources is an unresolved challenge (see 'Justice and Access to Health Care', Stanford Encyclopedia of Philosophy, available online at: http://plato.stanford.edu/entries/justice-healthcareaccess/#WheAccCarEqu (download on 27 September 2011)). This is on the one hand, because not all types of inequality are necessary unfair: for example, if an individual has lower income because he or she chose to devote more time to leisure activity than to earning money.

Also, equity can relate to different concepts, for example, equal health-care spending per capita (across regions); equal health; equal access to health care; or equitable allocation according to medical need.[48] Also, the World Health Organization (WHO)'s concepts of horizontal equity (health care to all individuals with the same medical need) and vertical equity (preferential health care for those with greatest need)[49] illustrates that using equity as a prioritization criterion involves determining an ethically justified dimension of equality like medical need.

One approach to account for equity concerns in resource allocation is to address trade-offs between resource allocations, which maximize benefit and those which prioritize certain health needs. This could be done, for example,

by assigning special weight to health gains that accrue to more severely ill patients. In genetics, special weight could be assigned to interventions with higher health need according to the concepts described above (eg, to diagnostic rather than predictive tests). However, equity weights are still subject to a range of methodological deficiencies.[50]

Avoiding specific kinds of inequity

According to the WHO, '*Equity* is the absence of avoidable or remediable differences among groups of people, whether those groups are defined socially, economically, demographically, or geographically. *Health inequities* therefore involve more than inequality with respect to health determinants, access to the resources needed to improve and maintain health or health outcomes. They also entail a failure to avoid or overcome inequalities that infringe on fairness and human rights norms' (see http://www.who.int/healthsystems/topics/equity/en/ (accessed on 14 May 2013)).

There are different ways in which concerns about avoiding inequities can arise in genetics. For example, the expected benefit and cost-effectiveness of genetic tests may depend on characteristics of the target group, for example, prevalence of the mutation (which depends on ethnic or family background) or gender (if disease expression differs according to sex as in the case of hereditary haemochromatosis).[4,51] Stratifying patients by these criteria may raise both ethical and legal concerns – for example, if genetic testing for hereditary haemochromatosis in Northern European countries were to be limited to male subjects or to people of European ancestry because cost-effectiveness is better in these groups. As a consequence, limits may be defined for using the risk of disease as a criterion in decision making.

The role of equity in decision making

If resources are not sufficient to provide all genetic tests, equity as prioritization criterion can be implemented in terms of lotteries or first come, first serve.[21] Both of these principles are blind to additionally relevant factors, and particularly, the latter in practice favours those who are wealthier, more powerful and better connected.[21] Although no use of lottery could be found in the genetics literature, the criterion 'length of waiting time' and thus 'first come, first served' was mentioned in the survey of Adair and co-workers.[7]

Equity is likely to have a much larger role in the form of applying certain criteria (such as scientific evidence of health benefit) and procedures (such as rules for stakeholder participation) consistently across decisions. Furthermore, the concern about specific kinds of inequity appears to have a role in decision practice, which is illustrated by a range of legal regulations to avoid genetic discrimination.[52]

Promotion and reward of social usefulness. The fourth category of principles for allocating scarce resources mentioned by Persad *et al*[21] is 'promoting and rewarding social usefulness'. Promoting social usefulness implies prioritizing services to specific individuals to facilitate future usefulness, for example, by giving priority to the health-care staff in the allocation of scarce influenza vaccine. This may include allocation to individuals who use fewer resources, for example, by agreeing to improve their health. Rewarding social usefulness implies prioritizing specific individuals who have promoted important values or have undergone specific sacrifice in the past. This may include allocation to individuals who reduced their need for health-care resources due to healthy lifestyle choices.[21]

Denying care based on the principle of rewarding and promoting social usefulness involves a range of ethical concerns. For example, use of this principle might require intrusive and humiliating enquiries about whether or not an individual adhered to a healthy lifestyle.[21] Currently, the principle of promoting and rewarding social usefulness does not seem to have an important role in prioritization decisions in general nor to be of high relevance for prioritizing the use of genetic tests.

Table 1 provides an overview of prioritization criteria. The criterion related to social usefulness is omitted because it is unlikely to be relevant for prioritizing genetic tests.

DISCUSSION

This study provided an overview of normative criteria for prioritizing genetic tests. All of these are faced with particular strengths and limitations: prioritization based on claims, which accrue from medical need appears to have the strongest ethical basis and practical acceptability. However, there is limited available methodology and practical experience with frameworks of measuring and balancing health and intervention needs and costs. Frameworks establishing intervention need such as the EGAPP framework are too resource consuming to be applied to all genetic tests, which are existing and applied in decision practice; also, they are currently unable to prioritize across different intervention needs.

Benefit maximization is the standard criterion applied by health economic frameworks and well-developed evaluation methods are available and applied by institutions such as NICE. However, they can be even more resource consuming than frameworks for evidence assessment. Also, different economic schools of thought exist with different associated outcome measures and practical implications so that an ethically grounded choice of the most appropriate framework needs to be made. Finally, there is ethical criticism to benefit maximizing frameworks because individual claims rather than overall benefit could be considered an appropriate normative basis for prioritizing health-care resources.

Also, treating people equally is a frequently stated principle of fair resource allocation. However, it is a challenging task because it requires a specification of which dimensions of equality are considered normatively relevant. Rather than a principle on its own, it is likely to be relevant in terms of applying other criteria with a firm normative basis equitably to all patients requesting a genetic test.

The principle of rewarding and promoting social usefulness has a long-standing tradition in the reflection over fair allocation, tracing back to Aristotelian ethics. However, applying this criterion in the prioritization of genetic tests is unlikely to be acceptable, for example, because it would require intrusive questions to patients.

Overall, claims based on health and intervention need appear to be the strongest normative basis for allocating scarce health-care resources to genetic tests; however, benefit maximization frameworks are much further developed for addressing problems of resource scarcity.

Limitations

There are many issues in the design and provision of genetics services that are ethically relevant but could not be addressed within this study, which focused on the prioritization of beneficial genetic tests only. These are, for example, the way how incidental findings should be addressed appropriately; ethical issues with preconception and prenatal screening; and privacy concerns related to information spill-overs of genetic tests.

Reasonable people may disagree about which of the four sets of criteria should guide the prioritization of genetic tests. Also, there may be conflicting analyses of the policy implications of common values, for example, because of different interpretations of the criteria or scientific evidence of benefits or economic and social costs.[53] In such situations of disagreement about substantive criteria, it has been claimed that decisions should meet the criteria of procedural fairness.[54] Therefore, prioritization activities should also account for principles of procedural justice such as the framework of accountability for reasonableness by Daniels and Sabin,[54] which could not be addressed in this study.

Clearly, the prioritization of health-care resources is in the remit of those whose resources are prioritized – thus, the payers of health insurance distributions or committees of third party payers on their behalf. Therefore, any recommendation developed on a European level can only serve as a complement national and state regulation.

Implications for further research

To arrive at a recommendation of prioritization criteria or even an algorithm to derive a standardized priority score in conjunction with information sources such as the EuroGentest clinical utility gene cards or the UKGTN gene dossiers, further research is necessary. This includes the following steps: first, a normative framework needs to be chosen to identify relevant prioritization criteria; second, the criteria following from the framework need to be operationalized to allow for empirical assessment; third, relative weights for the criteria need to be determined, for example, through discrete choice experiments.[55]

To improve the legitimacy of such weights, the empirical evidence should attempt to capture value judgments of key stakeholders such as clinicians and patient representatives. Furthermore, given that disagreement may persist, any prioritization guidance should additionally be developed in a decision process oriented at principles of procedural justice such as accountability for reasonableness.

Finally, prioritization of care is interrelated with but still separate from prioritization of applied research. Therefore, also research with stakeholder involvement on prioritization of research would be desirable.

Implications for decision makers

In the current economic climate, public health-care budgets are faced with particular resource constraints. This is likely to lead to situations where not all desirable tests can be funded and priorities have to be set which genetics services to provide – therefore, this activity is generally of high relevance for decision makers.

Fair and reasonable prioritization of genetic tests is a complex challenge for which no easy solutions exist. Therefore, efforts should be made to choose an appropriate framework for the explicit prioritization of genetic tests in a normatively and economically reflected manner. Otherwise, there is the risk that limits are set implicitly in an unfair manner. This overview of criteria can be used as a first orientation for reflecting whether current service provision corresponds with reasonable priorities. Such reflection can feed into the development of standard operating procedures, for example, regarding which patients receive higher priority and which are assigned to waiting lists.

Evolving science has led to multiple new genetic tests available; research and practical experience in health-care priority setting can help allocate these medical innovations in a reasonable and fair manner.

CONFLICT OF INTEREST

The authors declare no conflict of interest.

ACKNOWLEDGEMENTS

We acknowledge the comments from various colleagues, especially Per Carlsson, Jürgen John, Helena Kääriäinen, Alastair Kent, Michael Krawczak, Ulf Kristofferson and Irma Nippert. All errors of omission and commission remain our own. The study was partly supported by EuroGentest, an EU FP6 supported NoE Contract Number 512148 (EuroGentest Unit 3: Clinical genetics, community genetics and public health, Workpackage 3.2 (J Schmidtke)) and its successor EuroGentest2 (contract number 261469).

The findings and conclusions in this manuscript are those of the authors and do not necessarily represent the official position of the Centers for Disease Control and Prevention.

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