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Opportunity analysis of newborn screening programs

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Abstract

There exist congenital diseases that reduce newborns' potential opportunities. This reduction is sometimes alleviated if the congenital disease is early detected thanks to a newborn screening program. We propose an outcome measurement of newborn screening programs based on the opportunity gains they offer. We show that, under plausible assumptions, the ranking of the available screening programs for a particular disease, according to this new outcome measurement, do not depend on the metric of opportunity. We also apply our model to the current debate about choosing between a selective or a universal newborn hearing screening program to detect congenital hearing impairment.

Keywords: opportunities, potential success, screening programs

JEL classification: D61, D63, I12, I18

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1 Introduction

One of the most flourishing areas within health economics is the one that concerns equity in the delivery of health care (see Wagstaff and van Doorslaer, 2000; Williams and Cookson, 2000; and the literature cited therein). The word "equity" usually refers to the distributive justice in the allocation of a commodity ("health care" in this case). The underlying motivation in most of the above-mentioned literature is the so-called 'just compensation principle' by which health inequalities that are not attributable to an individual's responsibility should be compensated by society. An obvious instance of health inequalities that are not attributable to an individual's responsibility are those inequalities due to congenital impairments. The 'just compensation principle' implies that society should do its best to alleviate the consequences of congenital diseases in impaired infants.

There are some congenital impairments whose negative consequences could be alleviated by means of an early detection and a subsequent treatment. Typical examples are, for instance, congenital hearing impairment, hypothyroidism or phenylcetonuria. For these cases, the implementation of an early detection protocol seems to be sufficiently justified on the sole basis of the 'just compensation principle', even independently of its direct cost. Indeed, early detection protocols can be deemed as an efficient tool to avoid future compensations to individuals suffering from congenital impairments.

If the decision about the implementation of an early detection protocol appears to be unquestionable, the selection of a particular protocol among the alternatives to be implemented is, by no means, a straightforward decision. Usually, this decision is conducted by an economic evaluation of the existing alternatives. The economic evaluation of health care programs involves both technical and value judgements. This, and the special nature of the commodity being considered, is the germ of the complexity of this problem whose multiple angles leave room for new techniques to be developed with which we could face the problem. In this paper we present one of such new techniques to address the evaluation of early detection protocols, also known as *screening* programs.

Screening is traditionally defined as testing a population of asymptomatic individuals to identify precursors of a disease. The subjects who test positive are sent on for further evaluation in a subsequent diagnostic evaluation to determine whether they do, in fact, have the disease. An implicit assumption underlying the clinical interest of screening programs is that early detection, before the development of symptoms, will lead to a more favorable prognosis. This is so because, by means of a screening, it is possible to

treat the disease before it becomes clinically manifest, which is more effective than a later treatment. Usually, there are different screening strategies for a given disease. As mentioned above, we assume that the decision about implementing one of them is taken up exogenously and the issue is to select the best strategy to carry out, among the available ones.

The first problem that one has to face in order to run an economic evaluation of screening programs is the outcome measurement of the available strategies. This is a major issue in the discipline of health economics, since no measure has presented itself free of shortcomings and clearly superior to the other existing ones. In a companion paper, Herrero & Moreno-Ternero (2005a), we analyze the problem of selecting among screening programs, making use of QALYs, possibly the most frequently employed measure in health economics (e.g., Gold et al., 1996; Drummond et al., 1997; Dolan, 2000). It is a quite tractable measure and therefore easy to use. Nevertheless, it is both practically and conceptually dubious. The use of this measure is commonly associated with the assumption that health care resources should be allocated so as to achieve the maximal health gain as measured by additional QALYs. Many authors have raised concerns about the equity implications of this allocation rule (e.g., Wagstaff, 1991; Bleichrodt et al., 2004; Østerdal, 2005). It has also been argued that QALYs rely on very restrictive assumptions on individual preferences (e.g., Drummond et al., 1997; Dolan, 2000).

Here we propose evaluating newborn screening programs for a given congenital impairment by means of an *opportunity analysis*. By this we mean computing the opportunities a newborn screening program offers to a randomly given individual (i.e., the *average opportunity* the program offers). To formalize our model we make use of a metric of opportunity. That is, we associate with each individual a unique number on a zero-one scale, interpreting that number as the degree of the potential opportunities the individual enjoys. This numerical measurement renders the subsequent analysis very tractable analytically and, as we shall see later in the text, under some assumptions, the opportunity analysis is robust to the particular metric being considered.

For a given cohort of newborns susceptible of being impaired, we distinguish four reference groups after implementing a screening program: true positives; false positives; true negatives; and false negatives. We assume that individual opportunities do not vary within these four subgroups. The expected opportunities of an individual when a screening is implemented is the probability weighted sum of the opportunities in each of the four groups. Since these are program-specific probabilities, the expected oppor-

tunities will differ depending on the screening program being considered. If we adopt as the 'status quo' the absence of screening, we may identify as the outcome of a screening program the potential opportunity gains it offers, with respect to the status quo.

We formalize this model in the paper and show that the opportunity analysis just described can be considerably simplified under some assumptions. More precisely, we show that under three assumptions; namely, (i) opportunities do not decrease 'per se' by being referred to a screening program, (ii) no differences in opportunities between healthy individuals with different test results, and (iii) opportunities of true positives are strictly larger than those of false negatives (with a constant difference across screening programs), the outcome of a program can be seen as its level of *sensitivity*, i.e., the probability of finding by the screening procedure a disability when it is actually there. This implies that the conclusions of an opportunity analysis of newborn screening programs, for a given disease, would not depend on the particular metric of opportunity chosen, as long as this metric obeys the three assumptions described above. This reinforces the interest of our result.

The economic literature on the measurement of opportunity can be seen as part of a more general attempt to establish non-welfarist foundations of social choice (e.g., Peragine, 1999). Welfarism is the demand that the evaluation of any social state be based on the utilities generated in that state (e.g., Sen, 1991). Much of the traditional social choice theory is welfarist in the sense that the goodness of any social state is taken to depend, ultimately, only on the individual utilities or welfares of the people in that state.[1] The literature on the measurement of opportunity is an attempt to give a more central place to the freedoms and liberties of individuals in the determination of social welfare. Measuring the opportunities available to an individual amounts to measuring an individual's freedom of choice. Hence the relevance of this literature from the egalitarian point of view, as opposed to the concerns regarding the equity implications of QALY-based analysis of health care programs, mentioned above.

In all the economic literature on the measurement of opportunity, however, different measures are characterized by formal axioms usually justified by casual appeals to intuition, without reference to any clear theoretical position about what freedom of choice is and why it matters (e.g., Sugden, 1998). The, perhaps, most controversial question investigated in this litera-

[1] On the rationale of the welfarist approach, see Arrow (1951), Harsanyi (1955) and Mirrlees (1982) among others.

ture is that of linking a quantity-based measure of opportunity (what we call a metric of opportunity) with some notion of the value of the available alternatives. Our main result in this paper shows that, under mild assumptions, the opportunity analysis of newborn screening programs is immune to this controversial question, as its conclusions are fairly robust to the particular metric of opportunity being employed for the analysis.

We conclude the paper applying our model to the current debate on the implementation of newborn hearing screening programs in some states of the US and in some European countries. We show that, according to an opportunity analysis, universal programs are preferred to selective programs, in which only newborns with a risk factor are screened. This conclusion agrees with the more recent pediatric recommendations that have been published (e.g., Joint Committee on Infant Hearing, 2000).

The rest of the paper is organized as follows. Section 2 contains the basic model of the opportunity analysis. The results for this model are presented in Section 3. An application to the case of congenital hearing impairment is taken up in Section 4. Section 5 concludes.

2 The model

We consider a particular congenital disease or impairment for which there exist newborn screening programs that permit its early detection. We assume that the early detection of the impairment, followed by an adequate treatment, might reduce considerably its negative consequences. Let $\mathcal{N} = \{1, ..., n\}$ be the corresponding cohort of newborns susceptible of being impaired. The status of a newborn with respect to the disease is either $d = 0$ (if the infant is healthy) or $d = 1$ (if the infant is impaired). We denote by $\rho \in [0, 1]$ the *prevalence* of the disease in the cohort, i.e., the fraction of impaired newborns in the cohort.[2] We denote by G_0 the set of newborns with negative disease status and by G_1 the set of newborns with positive disease status. By construction, $\mathcal{N} = G_0 \cup G_1$ and the number of newborns in each of the subgroups is $|G_0| = (1 - \rho) \cdot n$ and $|G_1| = \rho \cdot n$, respectively.

Newborns can be partitioned into four groups, according to whether they do or do not have the disease and whether their screening tests are positive or negative. Thus, there are four groups of newborns: *true positives*, newborns whom the screening correctly indicates to have the disease; *false positives*, those who do not have the disease but who have a positive screening test;

[2] If i denotes the number of impaired newborns in $\mathcal{N}$ then $\rho = \frac{i}{n}$. We interpret this number as the probability of a newborn in the cohort being impaired.

false negatives, those who have the disease but are mistakenly cleared by the screening; and *true negatives*, those who do not have the disease and are correctly identified as such by the screening.[3] We can compute how likely an individual would belong to each of the four groups by using characteristics of the population (prevalence) and of the detection ability of the screening test (sensitivity and specificity). The *sensitivity* of the screening test (π_1) is the conditional probability that an individual with the disease is positively detected by the test. The *specificity* of the test (π_2) is the conditional probability of an individual without the disease being correctly detected as negative in the test. Using these definitions, the probability of an individual being a true negative is the probability that she does not have the disease $(1-\rho)$ times the probability that the screening correctly indicates that she does not have the disease (π_2). The probabilities of the individual to be a true positive ($\rho\pi_1$), a false positive ($(1-\rho)(1-\pi_2)$) and a false negative ($\rho(1-\pi_1)$) can be similarly expressed. The advantage of this way of writing the screening probabilities is that it makes easier to assess the implications of variations in the parameters ρ, π_1 or π_2 separately.

Let $\mathcal{S} = \left\{s^1, ..., s^m\right\}$ denote the set of available screening programs for the early detection of the disease. Let s^0 denote the 'status quo', i.e., the scenario without any screening program. For all $j = 0, 1, ..., m$, each screening program s^j is defined as $s^j = s^j(\pi_1^j, \pi_2^j, c^j)$, where π_1^j and π_2^j denote the sensitivity and the specificity of s^j respectively, and c^j denotes the incremental costs of the screening program with respect to the status quo.[4] By costs of a screening program, we mean the costs incurred by the test, i.e., technology and wages of the specialists who supervise it, and the costs of the final diagnostic evaluation to which every positive infant is referred after the screening test. It is worth noting that we assume all impaired individuals receive diagnostic evaluation, regardless of whether their impairment is detected early or not. Consequently, the incremental health-care cost of implementing a screening program s^j is

$$c^j = c_s^j + r \cdot (1-\rho) \cdot (1-\pi_2^j) \cdot c_d, \tag{1}$$

where c_s^j is the cost of the screening itself, r is the return rate and c_d the cost of the diagnostic evaluation.[5]

For ease of exposition, denote an infant's test result in the screening program s^j as $t^j = 0$ if it is negative, and as $t^j = 1$ if it is positive. Then, for

[3] If there is no screening program being implemented then the group of false negatives is G_1, whilst G_0 is the group of true negatives.

[4] From here onwards, unless otherwise stated, it is assumed that all costs are per capita.

[5] By return rate we mean the percentage of infants returned for follow-up testing.

each $s^j \in \mathcal{S}$, $d \in \{0,1\}$ and $t^j \in \{0,1\}$ denote by $G^j_{(d,t)}$ the group of infants sharing disease status d and test result t^j, after implementing s^j. Thus, $\mathcal{N}$ is expressed as follows:

$$\mathcal{N} = G^j_{(1,1)} \cup G^j_{(0,1)} \cup G^j_{(1,0)} \cup G^j_{(0,0)}. \tag{2}$$

According to the notation introduced above, it is straightforward to see that the probabilities of being in each of the groups are given by:

$$\begin{aligned}
\rho^j_{(1,1)} &= \rho \cdot \pi^j_1 \\
\rho^j_{(0,1)} &= (1-\rho) \cdot (1-\pi^j_2) \\
\rho^j_{(1,0)} &= \rho \cdot (1-\pi^j_1) \\
\rho^j_{(0,0)} &= (1-\rho) \cdot \pi^j_2
\end{aligned}$$

Now, we compute the potential opportunities of each newborn. To do so, we need a *metric of opportunity*. A metric of opportunity is a mapping associating with each individual a unique number on a zero-one scale representing the degree of potential opportunities, and interpreting 0 (1) as the lowest (highest) possible degree of potential opportunities an individual might face. Instances of metric of opportunity could be the probability of attaining a minimum level of income, or a certain level of education, or even a certain life expectancy above some level of good health. Formally, a metric of opportunity is a function

$$\Omega : \{0,1\} \times \{0,1\} \times \mathcal{S} \cup \{s^0\} \mapsto [0,1],$$

where $\Omega(d,t,s^j) \in [0,1]$ denotes the degree of potential opportunities of an individual with disease status d and a test result t^j after implementing s^j.

We define the *degree of opportunity* associated with each screening procedure $s^j \in \mathcal{S}$ as

$$\Omega^j = \sum_{d,t=0,1} \rho^j_{(d,t)} \cdot \Omega(d,t,s^j), \tag{3}$$

where $\rho^j_{(d,t)}$ is the probability of being in $G^j_{(d,t)}$. In other words, Ω^j is the sum of the degrees of potential opportunities associated with each group (true positives, false positives, false positives and true negatives) multiplied by the probability of an individual being in the group. In this respect, Ω^j can be interpreted as the expected opportunities of an infant after implementing s^j.

In particular, the degree of opportunity associated with the 'status quo' comes determined by

$$\Omega^0 = \rho \cdot \Omega(1, 0, s^0) + (1 - \rho) \cdot \Omega(0, 0, s^0). \tag{4}$$

Consequently, the *degree of opportunity gained*, associated with a screening $s^j \in \mathcal{S}$, is $\mathcal{O}^j_\Omega = \Omega^j - \Omega^0$.

3 The result

In this section we provide additional assumptions under which the opportunity analysis described in Section 2 is independent of the metric of opportunity.

The first assumption says, roughly, that opportunities do not decrease 'per se' by being referred to a screening program. In other words, the degree of potential opportunities of a true (false) negative individual after implementing a screening program coincides with the degree of potential opportunities of a healthy (impaired) individual in the status quo. Formally:

Assumption 1: *For all* $s^j \in \mathcal{S}$ *we have*

$$\Omega(0, 0, s^j) = \Omega(0, 0, s^0), \text{ and } \Omega(1, 0, s^j) = \Omega(1, 0, s^0).$$

The second assumption says that there are no differences in potential opportunities between healthy individuals with different test results, i.e., between a false positive and a true negative individual. Formally:

Assumption 2: *For all* $s^j \in \mathcal{S}$ *we have*

$$\Omega(0, 1, s^j) = \Omega(0, 0, s^j).$$

The third assumption says that early detection of the disease is advantageous at an individual level, and that this individual improvement is independent of the screening program chosen. That is, the degree of opportunity gained by an impaired infant after being detected by a screening program is strictly positive and constant (although depending on the metric of opportunity) for each program. Formally:

Assumption 3: *For all* $s^j \in \mathcal{S}$ *we have*

$$\Omega(1, 1, s^j) - \Omega(1, 0, s^0) = \lambda(\Omega) > 0.$$

We have the following result:

Theorem 1 *If the metric of opportunity satisfies assumptions 1, 2 and 3, then the degree of opportunity gained that a screening program offers, according to this metric, is its sensitivity, up to a (multiplicative) constant factor.*

Proof. Let Ω be a metric of opportunity that satisfies assumptions 1, 2 and 3. By (4),

$$\Omega^0 = \rho \cdot \Omega(1, 0, s^0) + (1 - \rho) \cdot \Omega(0, 0, s^0).$$

Similarly, by (3), given a screening procedure $s^j \in \mathcal{S}$, its degree of opportunity according to Ω is

$$\Omega^j = \sum_{d,t=0,1} \rho^j_{(d,t)} \cdot \Omega(d, t, s^j).$$

Since for every $d \in \{0, 1\}$ and $s^j \in \mathcal{S}$, $G^j_{(d,0)}$ and $G^j_{(d,1)}$ are disjoint sets, then $\rho = \rho^j_{(1,0)} + \rho^j_{(1,1)}$ and $1 - \rho = \rho^j_{(0,0)} + \rho^j_{(0,1)}$. Thus,

$$\Omega^0 = (\rho^j_{(1,0)} + \rho^j_{(1,1)}) \cdot \Omega(1, 0, s^0) + (\rho^j_{(0,0)} + \rho^j_{(0,1)}) \cdot \Omega(0, 0, s^0).$$

Consequently,

$$\begin{aligned}
\mathcal{O}^j_\Omega &= \Omega^j - \Omega^0 \\
&= \rho^j_{(0,0)} \cdot (\Omega(0, 0, s^j) - \Omega(0, 0, s^0)) + \rho^j_{(1,0)} \cdot (\Omega(1, 0, s^j) - \Omega(1, 0, s^0)) + \\
&\quad \rho^j_{(0,1)} \cdot (\Omega(0, 1, s^j) - \Omega(0, 0, s^0)) + \rho^j_{(1,1)} \cdot (\Omega(1, 1, s^j) - \Omega(1, 0, s^0))
\end{aligned}$$

By Assumptions 1 and 2, the first three terms are zero, and therefore we have:

$$\mathcal{O}^j_\Omega = \rho^j_{(1,1)} \cdot (\Omega(1, 1, s^j) - \Omega(1, 0, s^0)) = \rho \cdot \pi^j_1 \cdot (\Omega(1, 1, s^j) - \Omega(1, 0, s^0)).$$

By Assumption 3, $\lambda(\Omega) = \Omega(1, 1, s^j) - \Omega(1, 0, s^0) > 0$, for all $s^j \in \mathcal{S}$. Then,

$$\mathcal{O}^j_\Omega = k \cdot \pi^j_1,$$

where

$$k = k(\rho, \Omega) = \rho \cdot \lambda(\Omega) > 0.$$

Note that k depends on the prevalence of the disease and the metric of opportunity. It is not, however, screening program-specific. ■

It can be inferred from the proof of the theorem that both the degrees of opportunity gained and the sensitivity levels of the programs yield the same ranking of the available programs. The main relevance of Theorem 1 lies therein; the ranking of the available newborn screening programs for a given disease, according to the opportunities they offer, does not depend on the metric of opportunity that we decide to use, provided this metric obeys the three assumptions introduced above. It is worth noting, nonetheless, that the cardinal information of this ranking is captured by the constant (k) that appears in the proof, which depends on the prevalence of the disease and the metric of opportunity. Hence, we acknowledge that our result is only informative when the analysis refers to screening programs for the same disease. It cannot be used, however, to compare screening programs of different diseases.

To conclude with this section, one might argue that the model described is extremely simple and somehow unrealistic. In particular, it only computes the influence of suffering a congenital disease on the potential opportunities of a newborn and rules out any other key possible *circumstances*, such as gender, race, parental socioeconomic status, level of formal education attained by parents, etc., that also influence individual potential opportunities. The model presented in this paper, however, can be easily enriched to account for additional individual circumstances and their effect on the potential opportunities of a newborn. It can be shown that an analogous result to Theorem 1 would also be obtained in this generalized model, provided we generalize accordingly the above three assumptions to this setting (i.e., referring to individuals with the same circumstances) as well as we include an additional assumption saying that being identified as a true positive does not depend on the remaining circumstances (e.g., Herrero and Moreno-Ternero, 2005c).

4 Application: the case of congenital hearing impairment

We now apply our model to the particular case of congenital hearing impairment. This is an impairment that satisfies all the medical requirements to impose a prevention program, based on a newborn screening protocol. First of all, it is a serious impairment, for which a lack of early diagnosis will cause problems in language acquisition. Significant hearing loss interferes with the development of speech perception abilities needed for later language learning. These impairments in communication skills can lead to learning disabilities and ultimately, to limitations in career opportunities.

Moreover, it is more frequent than other impairments for which newborn screening programs are in use in developed countries. Finally, there are reliable screening methods, with high levels of sensitivity and specificity, and there is also an effective treatment available. As a consequence, there is a broad agreement to impose a newborn hearing screening program (e.g., Joint Committee on Infant Hearing, 2000).

Having reached this consensus, the debate moved to select between a universal and a selective alternative. In a *Universal Newborn Hearing Screening* (*"UNHS"* hereafter) every newborn is tested, whereas in a *Selective Newborn Hearing Screening* ("*SNHS*" hereafter) only those who were born with a risk factor, such as being in the neonatal intensive care unit or having a family history of hearing impairment, are tested. A UNHS is more expensive but also more effective, since only 50% of newborns with a hearing impairment belong to a group at risk. It is currently mandated in 32 states of the United States. The SNHS, however, was and continues to be practiced throughout the United States and the rest of the world (e.g., Keren et al., 2002).

There is ample literature on choosing between UNHS and SNHS, especially from the medical viewpoint (see Joint Committee on Infant Hearing, 2000; Thompson et al., 2001; and the literature cited therein), but also from an economic viewpoint (e.g., Kemper and Downs, 2000; Kezirian et al., 2001; Keren et al., 2002; Herrero and Moreno-Ternero, 2005b). The aim of this section is to apply our model to provide an additional viewpoint to this current debate about choosing between the two alternatives. We think the opportunity analysis we shall present next represents an achievement with respect to the previous literature, as this literature fails to capture ethical aspects when it comes to measuring benefits associated with the implementation of screening programs.

4.1 Protocols

According to the recommendations of the Joint Committee on Infant Hearing, every neonate should be tested by *Otoacoustic Emissions* (*"OAE"* hereafter), a less efficient and expensive test that is thought to represent a reflection of sound waves when sounds are presented to normal ears, and they are not detected in ears affected by the large majority of types of hearing loss in newborns. Those who fail this test should be referred to a subsequent test (*Auditory Brainstem Responses*, *"ABR"* hereafter), a more efficient and expensive test that presents sounds to the ear and detects nervous system activity in specific locations of the hearing pathway (Joint Committee on

Infant Hearing, 2000).

This is, however, somewhat vague and imprecise as there exist different versions of both tests that are currently in practice in hospitals. For instance, there are two alternative options based on OAE: transient evoked otoacoustic emissions (TEOAE) and distortion product otoacoustic emissions (DPOAE). Similarly, although complete automated ABR testing remains the gold standard for determination of hearing loss, there is a shorter screening version (S-ABR) being used, that is less expensive and quicker.

Consequently, we evaluate two slightly different versions (combining some of the alternatives mentioned above) of the universal 2-stage screening protocol recommended by the Joint Committee on Infant Hearing, for which the medical literature has provided enough data to perform our analysis. In the first version (protocol $U1$), we consider automated TEOAE and ABR as described by Kemper and Downs (2000). In the second version (protocol $U2$) we consider OAE tests as a single entity involving both TEOAE and DPOAE, because, as argued by Kezirian et al. (2001), their cost and validity have been similar to date. As a second stage, the ABR testing refers to the shorter screening version (S-ABR) to obtain the precise protocol described by Kezirian et al. (2001). Finally, there is a different UNHS currently in practice in a Spanish region (Navarra). In this case, the protocol has three stages. The first stage consists on an OAE test to every newborn at the third day of life, before leaving the nursery. For those who failed it, there will be a second OAE at the fifteenth day of life. Finally, the third stage involves a new OAE test for those neonates who failed the second stage and return at the third month. We will refer to this protocol as $U3$.

A selective screening includes a previous stage with a high-risk criterion (HRC), and then applies the protocol for infants at risk for congenital hearing loss. We therefore have three alternative selective screening procedures, which will be called $S1$, $S2$ and $S3$. Each protocol (selective or universal) concludes with a diagnostic evaluation for those who failed after the last stage.

To summarize, we focus our attention on six alternative early detection programs. Table 1 shows the mean estimates of the general and specific data from each procedure (SQ refers to the status quo in which no screening procedure exists). Additional information about such data, like their confidence intervals, can be obtained in Kemper & Downs (2000), Kezirian et al. (2001) and Keren et al. (2002). Formally, following the notation of Section 2, let s^0 denote the absence of , s_1 (s_2) [s_3] the first (second) [third] UNHS procedure, and s_4 (s_5) [s_6] the first (second) [third] SNHS, based on high

risk factors.

Table 1
Data of the screening procedures

	Screening						
Parameters	SQ	$U1$	$U2$	$U3$	$S1$	$S2$	$S3$
Sensitivity (π_1^j)	0	.784	.902	.840	.463	.532	.496
Specificity (π_2^j)	1	.996	.950	.995	.999	.998	.999
Direct cost (c^j)	0	10.05	13.91	11.68	1.59	1.65	1.57
Prevalence (ρ)				.0011			

4.2 Opportunity Analysis

We now move to the opportunity analysis based on our model presented above. The assumptions of Section 3 are sound in the framework of newborn hearing screening. Hence, the opportunity analysis can be reduced to the study of the sensitivity of each program. Consequently, programs can be ordered according to the degree of opportunity gained they offer in the following way:

$$U2 \succsim U3 \succsim U1 \succsim S2 \succsim S3 \succsim S1,$$

where $s^i \succsim s^j$ is to be read "program s^i offers a higher degree of opportunity gained than program s^j".

For the sake of completeness, and to get additional information about the cardinality of preferences over the set of alternatives, we provide an opportunity analysis for a given metric of opportunity. We might think of several metrics to deal with this task. Here, we consider the notion of *degree of potential success*, introduced by Mariotti (2002), that particularly fits this example.

The basic idea provided by Mariotti is the definition of success by means of different variables, reaching some minimal values. Success could be, for instance, the attainment of a minimum level of income, or a certain level of education, or even a certain life expectancy above some level of good health. Our particular definition of success in this example will be "living above the poverty line". The metric of opportunity Ω is to be interpreted as the probability for an individual of reaching success, i.e., the probability for an individual of living above the poverty line.

To the best of our knowledge, no satisfactory database including information about income and poverty within the population of hearing impaired

exists. Here, in order to obtain a proxy for these data, we rely upon the *Online Resource for U.S. Disability Statistics*. This service provides statistics calculated by the Cornell University Rehabilitation Research and Training Center on Disability Demographics and Statistics (StatsRRTC) using data from the Current Population Survey (CPS), which is conducted by the Census Bureau and the Bureau of Labor Statistics. In the CPS, persons with a disability are those who have a "health problem or disability which prevents them from working or which limits the kind or amount of work they can do." In the year 2004, an estimated 25.5 (30.8) percent of civilian non-institutionalized, men (women) with a work limitation, aged 18-64 in the United States lived in families with incomes below the poverty line (cf. Houtenville, 2005). In the same year, an estimated 7.8 (10.8) percent of civilian non-institutionalized, men (women) without a work limitation, aged 18-64 in the United States lived in families with incomes below the poverty line (cf. Houtenville, 2005). We shall use these numbers as a proxy for the definition of the metric of opportunity Ω.

We observe from the above data that *gender* is a key (or at least, relevant) factor to determine the probability for an individual of living above the poverty line. Thus, we use the generalization of our model, described at the end of Section 3, to capture this additional circumstance. Formally, let $\Omega(g, d, t, s^j)$ denote the degree of potential opportunities of an individual with gender g, impairment status d, and a test result t, after implementing s^j. Then, if m (f) means 'male' ('female'),

$$\Omega(m, 0, 1, s^j) = \Omega(m, 0, 0, s^j) = \Omega(m, 0, 0, s^0) = 0.922,$$

and

$$\Omega(f, 0, 1, s^j) = \Omega(f, 0, 0, s^j) = \Omega(f, 0, 0, s^0) = 0.892,$$

for all $s^j \in S$. Similarly,

$$\Omega(m, 1, 0, s^j) = \Omega(m, 1, 0, s^0) = 0.745,$$

and

$$\Omega(f, 1, 0, s^j) = \Omega(f, 1, 0, s^0) = 0.692,$$

for all $s^j \in S$.[6] Finally, we assume that for those impaired individuals who were detected by a screening program, probabilities are slightly higher. There is some uncertainty about the precise magnitudes of the reductions

[6] Note that we are assuming here that the above numbers are independent of the screening program to which an individual was referred to (if she was referred at all). This is precisely the content of Assumptions 1 and 2 introduced above in Section 3.

in morbidity arising from earlier detection of congenital hearing impairment although the benefits are certainly positive (e.g., Thompson et al., 2001). We model this fact by saying that the probability of living above the poverty line for an impaired individual who was detected by a screening program is the average of the corresponding probabilities for a disabled and a non-disabled individual. Formally:

$$\Omega(m,1,1,s^j) = \frac{0.745 + 0.922}{2} = 0.834,$$

and

$$\Omega(f,1,1,s^j) = \frac{0.692 + 0.892}{2} = 0.792,$$

for all $s^j \in S$.

Now, by definition,

$$\begin{aligned}
\mathcal{O}^j_\Omega &= \Omega^j - \Omega^0 \\
&= \rho^j_{(f,0,0)} \cdot \Omega(f,0,0,s^j) + \rho^j_{(f,0,1)} \cdot \Omega(f,0,1,s^j) - \rho_{(f,0)} \cdot \Omega(f,0,0,s^0) + \\
&\quad \rho^j_{(f,1,0)} \cdot \Omega(f,1,0,s^j) + \rho^j_{(f,1,1)} \cdot \Omega(f,1,1,s^j) - \rho_{(f,1)} \cdot \Omega(f,1,0,s^0) + \\
&\quad \rho^j_{(m,0,0)} \cdot \Omega(m,0,0,s^j) + \rho^j_{(m,0,1)} \cdot \Omega(m,0,1,s^j) - \rho_{(m,0)} \cdot \Omega(m,0,0,s^0) + \\
&\quad \rho^j_{(m,1,0)} \cdot \Omega(m,0,0,s^j) + \rho^j_{(m,1,1)} \cdot \Omega(m,0,1,s^j) - \rho_{(m,1)} \cdot \Omega(m,0,0,s^0),
\end{aligned}$$

where $\rho^j_{(g,d,t)}$ denotes the probability of being in the group of infants sharing gender g, impairment status d and test result t, after implementing s^j, whereas $\rho_{(g,d)}$ denotes the probability of being in the group of infants sharing gender g and impairment status d after implementing s^0. Then, the above data imply that

$$\mathcal{O}^j_\Omega = \rho^j_{(f,1,1)} \cdot \lambda(\Omega, f) + \rho^j_{(m,1,1)} \cdot \lambda(\Omega, m),$$

where

$$\lambda(\Omega, m) = \Omega(m,1,1,s^j) - \Omega(m,1,0,s^0) = 0.089,$$

and

$$\lambda(\Omega, f) = \Omega(f,1,1,s^j) - \Omega(f,1,0,s^0) = 0.1,$$

Finally, we assume that 51.1% of the newborns are females.[7] Let then $\rho_m = 0.489$ and $\rho_f = 0.511$. Since there is no reported evidence showing

[7] This is indeed the proportion of females in the US population in 2004 (e.g., U.S. Census Bureau).

that being identified as a true positive depends on the individual gender, we assume that, for $g = m, f$, $\rho^j_{(g,1,1)} = \rho_g \cdot \rho \cdot \pi^j_1$, where ρ is the prevalence of congenital hearing loss. Thus, the degree of opportunity gained each program offers is

$$\mathcal{O}^j_\Omega = 0.011 \cdot (0.489 \cdot 0.089 + 0.511 \cdot 0.1) \cdot \pi^j_1,$$

Table 2 shows the degree of opportunity gained each program offers.

Table 2

Degree of opportunity gained						
	Screening					
Opportunity	s^1	s^2	s^3	s^4	s^5	s^6
$(\mathcal{O}^j_\Omega)$	0.000078	**0.000090**	0.000084	0.000046	0.000053	0.000050

5 Discussion

We have presented a new technique to select the best newborn screening program, for a particular congenital impairment, out of a set of mutually exclusive alternatives. Such a technique consists of evaluating screening programs by means of the potential opportunity gains they offer. In other words, we use a "utilitarian/harsanyian" social evaluation criterion with the important modification that "utilities" are interpreted as "opportunities". In doing so, we show that we can use the *sensitivity* as a proxy for the social desirability of a screening programme, as the three assumptions on which our theorem relies are more plausible under this interpretation.[8]

If financing is not an issue, we would advocate for implementing the program that provides the highest potential opportunities. Now, in health care, as in other areas of social policy, decisions have to be made concerning the allocation of scarce resources. Usually, the program that yields the highest degree of opportunity gained (i.e., the most effective program) is also one of the most expensive programs (as it is indeed the case in our application regarding congenital hearing impairment). If so, we recommend

[8] The reason for using a "utilitarian/harsanyian" social evaluation function is that this criterion has the unique virtue of being at the same time based on rationality at the social level and respecting the Pareto principle (e.g., Harsanyi, 1955). A well known drawback, nonetheless, of the utilitarian criterion is its indifference to ex-ante and ex-post inequalities. Several alternatives could have been considered to face this problem, such as taking a weighted sum of ex-ante and ex-post egalitarian criteria or introducing a measure of ex-ante fairness in the measurement of individual opportunities, (e.g., Bleichrodt, 1997; Fleurbaey, 2007).

the following algorithm, inspired by the discipline of health economics (e.g., Johannesson and Weinstein, 1993; Birch and Gafni, 1993; Garber, 2000), to address the trade-off between costs and effectiveness of these programs and decide accordingly the program that has to be implemented.

(i) As a first step, programs that are *strictly dominated*, i.e., those for which there exists another available program more effective and less expensive, should be excluded.

(ii) In the case of mutually exclusive programs with a dedicated budget, programs should be ranked according to effectiveness and then calculate the incremental cost-effectiveness ratio for each successively more effective program.

(iii) If any of these incremental ratios turns out to be less than the previous one in the sequence of increasingly effective mutually exclusive programs, then the less effective one is ruled out by *extended dominance*, and it should never be implemented irrespective of the amount of resources available.

This algorithm results in a sequence of programs with increasing incremental cost-effectiveness ratios. The optimal decision rule is then to move up the list of incremental ratios and implement successively more effective (and expensive) programs until the resources are exhausted.[9]

The algorithm works as follows, for the application of congenital hearing impairment considered in Section 4:

(i) Protocol s^4 is strictly dominated by protocol s^6 and therefore should be excluded from the set of alternative options.[10]

(ii) The ranking of the remaining alternatives, according to their effectiveness, is $s^2 \succsim s^3 \succsim s^1 \succsim s^5 \succsim s^6$. Thanks to Theorem 1, it is straightforward to show that the incremental cost-effectiveness ratio of two given programs s^i and s^j is given by

$$R_{i,j} = k \cdot \frac{c_i - c_j}{\pi_i^1 - \pi_j^1},$$

where k is the constant factor appearing in the proof of Theorem 1. From here, it is straightforward to see that $R_{5,1} > R_{1,3}$. Thus, since s^3 is more effective than s^1, we exclude s^1 by extended dominance.

(iii) We therefore have four programs (s^6, s^5, s^3, s^2) with increasing incremental cost-effectiveness ratios $(R_{6,5} < R_{5,3} < R_{3,2})$. Thus, the optimal

[9] This decision rule applies under the assumption of divisibility of programs with constant returns to scale (e.g., Birch and Gafni, 1993).

[10] Note that, thanks to Theorem 1, the relative effectiveness of the two programs can be addressed just by looking at their sensitivity levels.

choice would be to move up the list of incremental ratios and implement successively more effective (and expensive) programs until the resources are exhausted

Could we say, however, something more about our particular problem of newborn hearing screening programs, without knowing the available budget of resources? The answer is positive, as explained in what follows.

The economic evaluation of health care programs may be carried out from the hospital's perspective (including only health-care costs and ignoring all others) or from the social perspective (including not only health-care costs but also the costs arising from resources consumed in other sectors). The decision about the perspective to be used is a matter of debate (e.g., Gold et al., 1996; Drummond et al., 1997; Johannesson and Meltzer, 1998). An opportunity analysis as the one we present in this paper seems to be only justified from a societal perspective. Thus, it is worth mentioning that there are some (crucial) indirect costs associated to newborn hearing screening programs that have not been addressed in our previous analysis (e.g., special education or disability allowances). It turns out that, if we adopt the societal perspective, and compute these indirect costs, then all the programs being considered are cost-saving programs (an aspect that on its own could solely justify the implementation of a newborn hearing screening program, as opposed to the status quo). Now, if we rank programs according to the whole stream of costs (actually savings) associated with them, then we obtain the following:

$$U2 \succsim U3 \succsim U1 \succsim S2 \succsim S3 \succsim S1,$$

which is precisely the ranking we offered in the opportunity analysis of Section 4.[11]

To summarize, if we run an opportunity analysis (that discards any information regarding costs) or a cost-opportunity analysis from the societal perspective (in which we consider not only direct, but also indirect costs), then we have a complete ranking of the set of available newborn hearing screening programs. It is worth noting that, in particular, this ranking says that universal programs are preferred to selective programs, as recommended by the Joint Committee on Infant Hearing Statement. If, instead, we run a cost-opportunity analysis from the hospital's perspective (in which only direct costs are considered) then the mechanism presented above tells us the program we have to implement, as a function of the available resources

[11] See Herrero and Moreno-Ternero (2005b) for further details on the (indirect) costs associated to these programs.

we have. In any case, all the possible analyses are considerably simplified thanks to our result, that makes the opportunity measurement robust to changes in the metric of opportunity being considered.

To conclude, it is worth noting that, even though in this paper we addressed opportunities by means of a metric of opportunity, opportunities can also be interpreted in terms of *capability sets* (e.g., Sen, 1985). In a health care context, the capability set of a certain person is to be understood as the set of health profiles achievable by this person. It is not her health outcome, but rather, the set of her plausible health outcomes. The opportunities of a person increase when her capability set becomes higher. This is precisely what would happen for an impaired newborn whose impairment is detected by means of a newborn screening program. The opportunity analysis presented here could have been easily adapted and framed in terms of capability sets.

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