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The Economics of Prenatal Screening

by

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DISCUSSION PAPER 42

UNIVERSITY OF YORK

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THE ECONOMICS OF PRENATAL SCREENING

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Acknowledgements

I am grateful to Ken Wright and Alan Maynard for arranging funding for the study reported in this paper, and for their comments on earlier drafts.

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Abstract

Ethical issues must take first place in the evaluation of a prenatal screening programme. The economic issues are less weighty but will be important in the decision as to whether a particular programme is introduced. This paper presents a critique of the published economic studies of screening and seeks to establish the proper place for economic arguments, in an overall appraisal. It criticises the calculation of a summary "economie benefit" and proposes other methods for the presentation of economic information.

An appendix gives separate critical summaries of twelve published studies.

THE ECONOMICS OF PRENATAL SCREENING

1. Introduction

Prenatal screening programmes have become well established within the last fifteen years with the consolidation of amniocentesis and foetal blood sampling as well-tried procedures and the development of diagnostic tests applicable to the foetus for Down's Syndrome, neural tube defects and haemoglobinopathies. They are likely to be extended in the future as new techniques are proven (the sampling technique of chorion villus biopsy, for example, and the diagnostic technique for cystic fibrosis) and as awareness is spread through the population (Wald (1984), Brambati et al (1986), Chervenak et al (1986)).

The aim of prenatal screening programmes is to reduce the incidence of disability. This is done by testing during pregnancy to detect the defective foetus. At the choice of the mother the pregnancy may then be terminated, averting the birth of a disabled child.

In deciding whether it is right to introduce a particular prenatal screening programme there are many questions, of different kinds, to be answered. Is it right that the foetus should not live? If the answer in some circumstances is "yes" then what are these circumstances? Is it right to test many pregnancies, and in testing accidentally injure or kill non-defective foetuses in order to diagnose those that are defective? Is it right to spend money on prenatal screening, or could that money be better spent on assistance to handicapped children, or on kidney dialysis treatment, or on some other beneficial programme?

Economics has a contribution to make to the assessment of prenatal screening because a full answer to the last question will require estimation of the costs of screening and other programmes and of savings which may result from a reduced incidence of disability. Production of such estimates is the particular expertise of economists. Of course, a full answer to the last question also requires that the benefits of screening and any alternative programme should be compared and neither here nor in the debate on the other questions above can the economist claim any special professional competence. But to recognise this limitation is not to detract from the importance of the contribution which the economist can make.

This paper offers a critique of the economic studies of prenatal screening which have been published. The need for such a critique arises because the published studies in general employ a type of thinking which is engrained in economists but which is not appropriate to this particular topic. It is commonplace to say that to evaluate a screening programme the economic benefit must be considered along with medical, social and intangible considerations. The difficulty appears to be however that combining the different pieces of economic information into a single estimate of benefit actually makes it harder to set economic effects against other considerations. "Economic benefit" remains in unhappy isolation.

This paper therefore begins by setting out the consequences of a prenatal screening programme in order that the economic aspects can be seen in context. The next section then sets out the approach taken by economic studies. The following three sections, addressed primarily to economists, discuss the economic aspects and suggest how economic information should be compiled and presented. In the final section a framework for evaluating

prenatal screening is sketched and the place of economic appraisal within that framework is indicated.

2. The Consequences of Prenatal Screening

A prenatal screening programme has costs. To run it requires health care resources, including staff (laboratory personnel, radiographers, obstetricians and so on) and equipment, clinic and hospital space, and reagents. In addition the time of mothers and perhaps of members of their household will be taken up by attendance for screening, and they will also incur transport costs. The termination of affected pregnancies after diagnosis will make further call on health care resources and on mothers' own time.

The effects of the programme on pregnancy and its outcomes are presented systematically for the neural-tube defect screening programme by Chamberlain (1978) and Hibbard et al (1985). They are noted here, separating pregnancies which suffer tangible effects from the rest. Tangible effects include the intended consequences of the programme, that defective pregnancies are detected and, in many cases, terminated at the choice of the mother. They also include the unintended consequences of screening; the termination of normal pregnancies wrongly diagnosed defective, and side effects of the methods of foetal diagnosis, namely an increase in foetal loss through spontaneous abortion and some increase in morbidity among live born infants.

What the effect of screening on other pregnancies might be is largely a matter for speculation, as to how the mothers concerned learn about and experience the programme. Depending on the quality of information and

counselling supplied, one might expect that the majority, the true negatives, may experience a heightened initial concern about the risk of disability offset by the reassurance of a negative test. Small numbers of mothers falsely tested as negative may experience the programme as worse than useless. For the minority diagnosed as positive who eventually choose not to terminate pregnancy, the outcome is not affected by screening. Whether they gain or lose from knowing the diagnosis and making their difficult choice is not self-evident.

It should be noted that some women, knowing themselves at high risk of defective pregnancy, may refrain from reproducing in the absence of screening but choose to do so when foetal diagnosis becomes available (Modell et al. (1980), Modell (1986)). Freedom from fear for these mothers, and the subsequent birth of healthy offspring, are important benefits of foetal diagnosis.

The last heading under which to examine the consequences of screening programmes is that which describes the aim of these programmes: avoiding the birth of disabled infants. The elaboration of these consequences by economists is typically negative in form. Thus, so many births are averted, hence the resource costs of delivery at term are not incurred. Of the averted births so many would have been still births or neonatal deaths, leaving so many individuals to survive sometimes into adult life. As a consequence mothers and their households do not have the task of caring for these individuals, a task which can be a heavy emotional and financial burden. The state does not have to contribute to that task through medical, educational and institutional services, nor through financial support.

There are other consequences of averting these births. Although chosen by mothers, termination can nonetheless be followed by a period of acute grief (Donnai et. al. (1981), Lloyd and Laurence (1985)). More happily, it is held that, after termination of pregnancy, without the task of caring for the handicapped child, mothers may well conceive and bear further children who would not have been born in the absence of the screening programme.

3. The Economics of Prenatal Screening

The approach taken by economists will be illustrated from the papers by Mikkelsen et al. (1978) and Layde et al. (1979). (Further particulars of these papers are given in the appendix, summary 6 and summary 8.)

The first study is a simple study of the prevention of Down's syndrome. It selects from the consequences of screening two outcomes to which money values can readily be attached, namely that health service resources are used in the screening programme (for diagnosis and for terminations) and that the reduction in affected births reduces the resources devoted to care of Down's syndrome individuals (whether in provision of education, or institutional care, or through cash benefits). The value of resources used in the programme is its cost and the value of resources not now spent on care is its economic benefit. If benefits exceed costs for a particular maternal age group then the existence of this net economic benefit gives an argument for screening that age group.

Layde et al., in their study of screening for neural tube defects in the USA, make a similar calculation of net economic benefit by setting the resource costs of the programme against the resources saved by screening and termination. This second example however is more complex than the

first on two counts. It applies a much broader definition of "resources saved", which recognises that, as well as saving actual expenditure on care, society has economic gains from the increased production of mothers (who would otherwise be unpaid carers for the disabled individuals). So the measure of gross economic benefit is the overall increase in resources made available to society as a result of screening. Secondly, the authors recognise that mothers may have further children once an affected pregnancy is terminated and that the birth of these children would itself have resource consequences. They therefore give two estimates of economic benefit, one with and one without "replacement" of the affected pregnancy.

The methodologies of other studies differ in detail but they share the approach of the two examples described here. That approach in its general form, is to identify those consequences of screening which affect the availability of productive resources, to take the use of resources by the programme as the cost, an increase in resources made available by screening as the benefit, and the overall sum as the net economic benefit which if positive provides one argument for introducing the screening programme.

That the contribution of economics has been cast in this form creates two difficulties of interpretation. The first is that the economic consequences of screening have been identified and assessed in separation from other outcomes, and an overall assessment requires that all effects be taken into a single view. None of the authors reviewed would question this need, and they would place their contribution as being a partial assessment of the proposal to be taken into account together with non-monetisable consequences in an overall appraisal. It has already been stated and will be argued further in Section 5, however, that to present economic information as a single summary figure of net economic benefit makes it

difficult to place this information in a general evaluative scheme.

Before the presentation of economic information can be discussed however, the second, radical, difficulty must be faced. The economic studies bring forward as economic benefits the difference in the productive resources available to society between the situation arising from screening and termination of affected pregnancies and the situation, in the absence of screening, where disabled individuals are born and live out their lives. The difficulty lies in the definition of "society". In the absence of screening, resources are devoted to the welfare of disabled individuals. Following screening, these resources are available to contribute to the welfare of the rest of society. If "society" is taken to include the potential disabled individuals and "the welfare of society" is taken to include their welfare then screening saves no resources. It is only if the welfare of society can properly be taken to exclude the welfare of the potential disabled individuals that the "economic benefits" generally presented have any rationale. The next section considers whether the common definition of benefits can be defended.

4. The Welfare of Unborn Children

In the sometimes passionate argument which takes place over the ethics of abortion the welfare of the unborn child is raised as an issue, but whether it should be considered and how it can be weighted are themselves subjects of dispute.

An extreme formulation of a woman's right to choose requires that the childs welfare, if it is a consideration at all, is a consideration only to the mother. The very title of the most determined opponents of this view, the Society for the Protection of Unborn Children, shows their insistence

that the child's welfare should be the central one. In the more particular arguments on the ethics of termination for congenital or developmental disability, the question of the disabled individual's quality of life is sometimes addressed, but a stronger theme is the welfare of the mother and of society at large and the burden which the disabled child would create.

What can clearly be said from this diversity of argument is that there is no concensus which would a priori justify the economist exlcluding the child from calculations of aggregate welfare. The estimates of "economic benefit" which have been produced are therefore open to question and one must examine their implications to see what the figures mean, whether the estimation of benefits in this fashion produces useful knowledge.

The most useful example to take is the study by Henderson (Henderson 1982). The author was concerned, as were Layde et al. (1979), with the resource effects of "replacement", non-disabled children and therefore calculated the resource effect of such a birth on the same basis as had been employed for the birth of an individual suffering from spina bifida. (Layde et al. also perform this calculation, but their method had technical flaws which Henderson avoids and his results therefore are preferred.) The resource effect of a disabled birth was £1608, and that of a non-disabled birth was £1881.

In the study itself the resource effect of a non-disabled birth was used only to modify the total resource effect of screening, the "economic benefit" of screening being lowered if termination of some affected pregnancies is followed by birth of replacement children. However, it is the very fact that a non-disabled child was shown to have a positive resource cost which is of interest here. This result means that

termination of all pregnancies would show an "economic benefit", whose logical status is exactly the same as that of the "economic benefit" of screening. This appears a reductio ad absurdam. We do not accept "economic benefit" as providing any ground at all for universal termination. It therefore cannot be that the "economic benefit" of screening gives grounds to believe screening to be a good thing.

Thus the common economic methodology which excludes the unborn child from its consideration produces a "benefit" which in the ordinary usage of the work is no benefit at all. The obvious step, suggested by Culyer (1985) is to revise the methodology so that the welfare of the child is brought into consideration. When this is done, many of the elements which on the standard approach are considered to be costs to society of a disabled birth must now be reckoned to have corresponding benefits to the potential child. If, (following normal economic practices) monetary values are to be placed on these benefits they will be estimated as equal to cost, the normal practice in the absence of alternative valuation. The effect of allowing for the welfare of the child on the economic benefits of screening is quite radical, as can be seen from the table below.

Costs and benefits following birth of a disabled child

light face: costs and benefits when the child's welfare is excluded from consideration

heavy face: additional benefits when the child's welfare is included

Costs	Benefits
educational costs	**benefits of education**
institutional costs	**benefits of institutional care**
maternal production foregone	**benefits of mothers service**
	disabled individuals production
disabled individual's consumption	**benefits of disabled individual's consumption**

In light face are shown elements typically included in the net resource cost of a disabled child, and in the heavy face the additional elements to be included if the welfare of the child is considered. If in the absence of other data the additional benefits are valued at cost it will be seen that the only item which does not net to zero on the new methodology is the disabled individual's production. Thus, while the standard approach gives a net resource cost of a disabled birth and hence a resource gain following screening, the new methodology will show a simple resource loss following screening, equal to the loss of output which the disabled individual would have produced.

Both this new methodology and that of the standard approach are attempts to summarise the resource effects which follow the birth of a new person into a single measure of change in the economic welfare of society. Neither can be judged a success. The standard approach appears to support a policy of universal termination. The new methodology brings forward productive capacity as the single measure of benefit brought by the new individual. The search for a useable summary measure might be continued. For example, the scope of economic valuation could be extended to consequences of birth not so far monetised: this is the approach of Henderson, who seeks to value the "psychic benefit" to parents. Methods of estimating the benefits to the child other than by setting them equal to cost could be developed, so that the essentially trivial elimination of balanced costs and benefits which occurs on the inclusive methodology would occur no longer. The technical details of the calculation, particularly the discount rate, could be reappraised so as to find a measure which showed a non-disabled child as a net benefit and so avoided the paradox of universal termination.

The search for a useable summary measure may however be put to one side as it does not promise easy results. Instead, the economics of screening may retain as its content those resource effects included in the published studies (educational costs, lost maternal earnings and so on). However, it must be explicit that its results are not estimates of a single "economic benefit to society", since the notion of society is itself ambiguous, but show instead the effect on resources available on the one hand to the mother and her household and on the other to the state.

To make this explicit will defuse the paradox of universal termination. To say that the birth of children results in net costs to the mother and her household and to the state is a quite mundane statement, and no inference is to be drawn that it provides an economic argument for universal termination. Whether comparable statements about the costs of disabled children provide an argument for screening will be considered in the next section.

5. The meaning of economic benefit

It has been argued that the economic benefit to society of prenatal screening is not a straightforward concept, and that it is necessary to be explicit as to who benefits economically from screening. The mother and her household, and the state, benefit economically because they save the resource costs of caring for the disabled individual. The argument now to be developed is that the benefits to these different recipients, the mother and her household on the one hand, and the state on the other, should always be separately stated because the economic information summarised under the two different heads differs in its implications for a general evaluation of screening.

This proposition will be approached by way of a brief exposition of some of the standard ideas of economic cost-benefit analysis (Drummond (1980), Akehurst and Holtermann (1979)). The simplest situation for such an analysis exists when the application of certain productive resources causes production of things which are of value to society. The valuable outcomes, moreover, are of such a kind that a monetary statement of their value to society can be made - typically, they will be commodities and can be valued by their market price. We also know of other possible uses to which the resources might be put, and can quote monetary statements of the value to society of what would be produced by these alternative uses. In such a situation the result of cost-benefit analysis will be a statement of the monetary value of the benefit of the proposed use on the one hand and on the other a statement of its opportunity cost. "Opportunity cost" is the benefit foregone in implementing the proposal, the monetary value of what could be produced by the necessary resources in their best alternative employment. In practice, economists often take a methodological short cut. They do not specify alternative resource uses or estimate their particular benefits but instead price the resources which could be devoted to other use and take that sum as an estimate of the benefit they would produce. However, the principle remains that a cost-benefit analysis takes account both of the valuable outcomes of the proposal under discussion and the potential valuable outcomes of other proposals which would be precluded by its implementation.

The assessment of prenatal screening will necessarily differ from this model. It is not the case that the value of the outcomes of prenatal screening, beneficial or harmful, can readily and uncontroversially be stated in financial terms and the summary of cost and benefits must therefore include non-economic as well as economic items. Secondly,

screening programmes will, if implemented, cause resource savings and these savings may well exceed the programmes' resource costs. Thus, rather than preclude the implementation of alternative proposals, the implementation of screening makes possible the production of additional valuable outcomes through the redeployment of resources.

The financial costs of a disabled child to mothers and their households have been documented by Baldwin (1985). They include a reduction in mothers' earning capacity, and to a lesser extent that of fathers, from the demands of caring for the child. The household has to fund the child's own consumption and additional household consumption, such as house adaptation. The effects vary with family circumstances, with the severity and type of disability and the child's life expectation. They are in many cases substantial and are not in general recompensed by the transfer payments made available. Averting the birth of the disabled child through screening therefore is of economic benefit to mothers and their households. However, it is inappropriate to view the economic benefit in isolation. There is a risk of reifying a single effect into separate economic and non-economic benefits and perhaps believing that the economic benefit can convey the <u>value</u> to the household of averting the birth. Rather, quantification of economic effects can contribute to a description of the single benefit of screening, the relief to the household from the burden of caring for the disabled child.

While economic effects on the household are to be viewed as a part of the direct good achieved by the screening programme, the economic impact on the state by contrast is an impact on society's ability to achieve <u>other</u> goods. The difference to the state between the situation following screening and the situation without screening, when disabled individuals require care, is purely a financial difference. To attach evaluative

meaning to that difference requires that we consider what it means for achieving other goods, whether through the health service or through other state action.

The principles to be followed in the calculation and presentation of the resource effects on the state are therefore those used by government in assessing public sector proposals in general.* However, a different method of presentation will be appropriate to show the economic effects on mothers and their households. A single figure which sums the cost over the expected lifetime of the average disabled individual and discounts to present value will not be very meaningful. A presentation which showed the cost per week to representative households would be of more use in letting policy makers form a view of the benefit of screening in relieving the burden of care upon the household. To allow proper interpretation such figures should be accompanied by comparable estimates of the cost of a non-disabled individual, since it is not only disabled children who impose costs on their parents and to omit a comparative figure might overstate the impression.

6. The birth of "replacement" children

There is clear evidence that the availability of foetal diagnosis increases the birth rate among mothers who know themselves to be at risk of conceiving a child with a recessive genetic disease (Modell (1980) and (1986)), and economists have hypothesised that some mothers who terminate a pregnancy diagnosed defective, whether affected by such a disease, by a neural tube defect, or by Down's syndrome, will then conceive and bear a

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* These principles are well established among economists. They require future costs and savings to be discounted at the ruling public sector rate and corrected to factor cost. Account must be taken of transfer payments.

normal child, a "replacement" for the disabled child whose birth was averted. Since these normal children will impose resource costs on households and on the state, and since it is hypothesised that their births are a consequence of screening, it is necessary to consider how they should be reckoned in an overall assessment.

To show a reduction in the "economic benefits" of screening equal to the cost to the mother and her household of rearing the replacement child, the treatment adopted by a number of authors, is wrong. It is not after all, suggested that the additional children are an unwanted by-product of screening reluctantly supported by their mothers, and a methodology which implies that they are misleading. It is better to treat the birth of additional children as being generally a benefit to mothers, but one which cannot be valued in monetary terms. However, there remains the economic impact upon the state: the cost of education and other services, pensions and other transfers, net of taxes contributed by the individual. This amount should be noted, for it shows the extent to which some of the resources which were released by screening have been preemptively claimed by the birth of additional children and are not available for other uses.

The treatment of "replacement" proposed here is quite different from that adopted in many of the economic studies. In several studies the "replacement situation" and the "non-replacement" situation have an equal methodological status. In one study, only the "replacement situation" is considered (Hagard, Carter and Milne (1976)) and in two others the "replacement situation" is methodologically prior (Hagard and Carter (1976), Conley and Milunsky (1973): see notes in the appendix). In contrast, "replacement" features here as a minor and possibly negligible gloss on the economic argument.

Underlying these different treatments are different conceptions of what a prenatal screening programme is and how therefore it may be evaluated. If it is seen as a means by which a woman may bear an unimpaired rather than an impaired child - essentially equivalent say to not smoking during pregnancy, but with the capacity to prevent much greater impairment - then it is entirely appropriate to start from a "replacement" model. This paper begins from the belief that there are important evaluative differences between prenatal screening and not smoking as methods for avoiding impairment, and that the useability of economic assessments has been limited by these differences being ignored.

7. The costs and benefits of screening

The following tabulation brings together most of the components for the evaluation of a prenatal screening programme. The terms "cost" and "benefit" have been used very broadly.

Costs and benefits of prenatal screening

Costs	Benefits
A. THE STATE	
Programme costs.	
Termination costs.	Averted costs of delivery at term.
Costs of additional non-disabled children.	Averted costs of care for disabled children.
B. MOTHERS AND THEIR HOUSEHOLDS	
1. TRUE NEGATIVE DIAGNOSIS	
Time and travel costs of diagnosis.	Reassurance.
Foetal loss after diagnosis.	
2. FALSE DIAGNOSES	
False positive terminations.	
Adverse psychological effects of false negative diagnosis.	
3. TERMINATIONS OF DEFECTIVE PREGNANCY	
Grief.	Averted burden of care for the disabled.
	Additional non-disabled children.

A "cost" is a bad thing, either a direct bad effect on individual welfare or a call on the resources of the state (which reduces society's ability to achieve other goods). "Benefit" is used in the opposite sense.*

Three points of detail may be mentioned before the table is used to recapitulate the role of economic assessment. Firstly, a benefit to the state is shown equal to the averted costs of delivery at term of pregnancies which are terminated following screening. Unaccountably a number of studies which have included the cost of termination have overlooked this element, and while both it and termination costs have typically been small when set against programme costs if one is judged large enough to include them both must be included. Secondly, a number of logically possible outcomes have been excluded as being numerically negligible so that, for example, foetal loss after diagnosis is shown only against true negative diagnoses. Thirdly, private time and travel costs have been shown only for true negatives whereas such costs will in fact apply to all mothers. If true negatives do not comprise the overwhelming majority of diagnoses (as might be the case with prenatal diagnosis of recessive genetic conditions where both parents were carriers) then the treatment will need revision.

The main contribution of economic assessment is seen to be to Section A, the effect of screening upon the state, which can be reduced to a matter

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* To include costs averted as a result of screening among the "benefits" of screening has been questioned by various economists (Drummond (1980). A danger is seen that, once averted costs are named among benefits, their perceived importance will swell like a cuckoo in the nest until the benefit of screening is reduced to the money that is saved. A technical objection is that to treat averted costs as economic benefits rather than negative costs distorts the ranking of projects by benefit-cost ratios. The tabulation above will not be misused in either of these ways, and the naive useage of "cost" and "benefit" for bad and good consequences may be retained.

of financial costs and savings. Economic analysis can identify and sum these effects and give a statement of the extent to which a prenatal screening programme will increase or decrease the resources available to society to pursue other goods. In Section B, which lists direct effects on individual welfare, the contribution of economics is limited to the enumeration of benefits and costs as few aspects lend themselves to monetary valuation. Those that do are the time and travel costs of diagnosis met by individuals and the financial aspects of caring for children whether disabled or non-disabled. The requirement to view these economic effects within a general evaluative framework has not always been observed, and mistaken interpretation has resulted. Thus, when authors have included time and travel costs as programme costs they have not noted that these are in general freely incurred and thus the benefit of reassurance which applies to the majority, the true negatives, has not been noted as offsetting that cost. More importantly, the financial cost of a disabled child has been viewed in isolation with the implication, not always resisted, that it is in avoiding this cost that the benefit of screening lies. A logical corollary of this mistaken emphasis has been that economists often appear to describe the birth of additional non-disabled child as a cost and by implication a bad thing, instead of a good about which economics can say little.

Nowhere in the table is there mention of costs or benefits to the individuals most critically affected by screening, to whom screening is literally a matter of life and death. Since this tabulation is a tabulation of good things and bad things in a very broad sense it could be completed by including "living" as a benefit for additional children born as a result of screening and "not living" as a cost to those whose birth is prevented. The reason for not completing the table in this way is a belief

that "living" and "not living" are different in kind from any of the entries shown, and that it is more appropriate to remember independently that the major effect of screening is that it discourages the birth of some babies but encourages the conception of others.

Indeed, a recapitulation of the costs and benefits of screeening might distinguish three classes of effect. These matters of life and death would stand first. A second class would comprise the effects on the welfare of mothers and their households. Effects on the state would come third. The moral seriousness of the effects decreases from the first class to the third and the use of economics in the evaluation of effects increases. Economics has a subservient role and it is important that the form in which economic information is presented fits the form of the overall argument. Economists can calculate the effects of a programme on the state. They can contribute to the discussion of effects on mothers and their households. They should abandon the notion of a single economic benefit of screening lest moral, medical and economic assessments proceed in parallel without ever being able to meet.

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12. Andreano and McCollum (1983) 'A benefit-cost analysis of amniocentesis.'

1. Conley, R. and Milunsky, A. 'The Economics of prenatal genetic diagnosis.'

in Milunsky, A. (ed.) The Prevention of Genetic Disease and Mental Retardation, Philadelphia, W.B. Saunders Co. (1975)

Object To illustrate the costs and benefits of genetic analysis, with particular reference to Down's Syndrome, using the experience of Milunsky's (American) laboratory in 1970 to 1973.

Costs Costs of amniocentesis and karotyping procedures are taken from the laboratory's experience. An estimate of abortion costs is also taken into subsequent calculations of cost per birth averted (although it is a minor component of the total).

Experience The laboratory carried out 486 chromosome analyses in the period, the bulk on pregnancies indicated by maternal age or family history to be at risk for Down's syndrome. Six serious abnormalities were detected, and four pregnancies (all Down's syndrome) were terminated.

Benefits Figures are calculated for a "replacement" case (where termination of a defective pregnancy is followed by conception and birth of a non-disabled child) and for a "non-replacement" case. In the non-replacement case the only benefit estimated is the averted cost of institutional or medical care. For the replacement case, an estimate of the lifetime excess production of a non-disabled over a

disabled child is added.

Institutional care is assumed to be for all Down's syndrome children (and also for those affected by Hunters' syndrome or trisomy 18) for twenty years between ages 10 and 30. The real cost of care is assumed to grow at 2.5% per annum. When the replacement case is being evaluated an amount is netted off to represent the education and consumption of the non-disabled child. For children with Tay-Sachs disease medical, not institutional, costs are quoted.

Excess production of a non-disabled child is estimated as gross income from employment plus unmarketed output, net of the disabled childs' production. (For males unmarketed output is valued at 25% of gross income from employment; for females not employed it is estimated equal to the net income from employment of employed females. For females in employment it is half the amount of those not in employment). Production of disabled persons is reckoned to be 20% of the non-disabled figure for Down's syndrome or trisomy 18, and zero for Hunters' syndrome or Tay-Sachs disease. Production is assumed to grow at 2.5% per annum.

Cost base & discounting 1972 prices are used. Amounts for future years are discounted at 7%.

Results Costs per Down's syndrome birth averted are obtained by attributing screening costs of all 486 pregnancies to the four Down's terminations. (The resulting figure is quoted

as applying to trisomy 18 as well.) Benefit-cost ratios for Down's are two in the non-replacement case and three in the replacement case. Costs per birth averted of the unilocal genetic disorders (Tay-Sachs, Hunter's) are quoted on the assumption that foetal diagnosis will be applied where both parents are known to be carriers, so that cost per averted case is the sum of four diagnoses and one termination. Cost per case being lower by construction benefit-cost ratios are much higher, between 20 and 80.

Comment The authors state that it is impossible to estimate all the care costs of a disabled child (for example, schooling and day-care for those in the community, and medical and dental treatment). However, items other than the two or three which they have included could have been estimated to the same degree of accuracy and the set which they have chosen cannot be held to give a good estimate of the cost of either a disabled or a non-disabled child. Nevertheless it does include the major items for which the disabled child's costs are different from those of the non-disabled and thus it provides a reasonable estimate of the net cost of a disabled child in the replacement case. The authors' methodological focus upon replacement accords with their feeling that for a life of disability prevention, and replacement, is equivalent to cure.

2. Glass, N. 'Economic aspects of the prevention of Down's syndrome (mongolism).'

in Bailey, N.T.J. and Thompson, M. (eds.) Systems Aspects of Health Planning. Amsterdam, North-Holland Publishing Company (1975).

Object To assess economic costs of, and costs averted by, a proposed screening programme for Down's syndrome pregnancies among older mothers in the United Kingdom.

Costs There are to be 700,000 pregnancies per year, of which 1.5% are of women 40 or over and 6% women 35-39. Estimates from unspecified NHS sources of amniocentesis costs and karotyping costs are given, for different assumed repeat rates and work-rates. Karotyping cost is typically six times amniocentesis cost. No estimate of costs to mothers is included.

Outcome U.K. 1970 incidence rates, 100% coverage, and 100% accurate diagnosis and termination of affected pregnancies are assumed. 170 live births of affected children to mothers aged 40 or over would be avoided. Slightly more would be prevented for mothers aged 35 to 39.

Savings Estimates depend on assumed survival and extent of institutionalisation. 15 or 25% are assumed to die in the first year. 10 or 20% to be in institutions for life. Of the remainder, half are institutionalised at 15 and the rest at 25. Life expectancy after age 1 is not discussed. Estimates are then presented of institutional costs, of

educational costs and of family expenditure for those at home (equal to the difference in expenditures of one (non-disabled) child and two child families in the U.K.). The first two items are estimated both at a 5% real growth rate and as constant in real terms: the last at a real growth rate of 3%. The three components are of the same order of magnitude. The first component is the largest and the last is smallest.

Cost base & discounting Amounts are quoted in 1974 prices. A discount rate of 10% is used.

Results Savings exceed costs on all assumptions for mothers aged 40 or more, but are less than costs for those aged 35 to 39.

Comment Glass gives a literate account for the place of economic analysis in decision making. He is careful throughout to refer to averted cots as "savings" rather than "benefits". The set of items costed as savings is a hybrid, two being public costs, one private. Absence of suitable data is given as the reason for excluding consideration of maternal earnings foregone.

3. Hagard, S. and Carter, F.A. 'Preventing the birth of infants with Down's syndrome: a cost-benefit analysis.'

British Medical Journal 1976 i, 753-756 (1976).

Object To estimate the net economic benefits of a Down's syndrome screening programme for older women in the West of Scotland.

Costs The costs of running a programme over 20 years for women aged 40 and over and for women aged 35 to 39 are estimated, based on practice in the West of Scotland. Private costs of transport and lost working time represent 15% of the total. Laboratory costs are two thirds of the total. Other elements are publicity, ultrasound, genetic counselling and amniocentesis. Prevalence is based on Swedish data. There are 550 pregnancies to women over 40 and some 2000 to women aged 35 to 39.

Outcome 90% participation is assumed, and 99% sensitivity. On average 8.1 Down's syndrome pregnancies are detected and terminated each year among women aged 40 and over, as well as one case of spina bifida. Outcomes for women aged 35 to 39 are not reported.

Benefits Benefits of averting the birth of 20 successive annual cohorts are calculated and discounted to a total present value. Benefits are the averted costs of care. They are calculated first for the "replacement case", where termination of pregnancy is followed by the conception and birth of a non-disabled child, by summing across the

different cost elements the excess of a disabled child's costs over the average cost of a non-disabled child. An estimate of benefit for the "non-replacement case" is then obtained by adding to averted excess costs an estimate of the lifetime consumption of a non-disabled child.

Excess costs averted for the spina bifida birth are taken from the companion paper (Hagard, Carter and Milne (1976)).

Assumptions on life expectancy of Down's syndrome infants are presented. Degree of handicap expected is based on data from North East Scotland. The extent of institutional care is derived from West of Scotland data: the proportion of survivors in care is 25% by age 75 and increases by 25% every ten years till all are in care from age 45. Excess educational costs for those not in care are calculated. Maternal production forgone is estimated as one half of total production by mothers with a non-disabled child of similar age. Disabled individuals themselves have an average lower output than non-disabled. Only those with IQ greater than 50 are assumed to work, with half non-disabled productivity.

Cost base and discounting

Figures are quoted at July 1974 price levels. A 10% discount rate is used.

Results

Cost benefit ratios for women over 40 are 1.13 for replacement and 2.58 for no replacement. For all women over 35 they are 0.63 and 1.25 respectively. It is argued that the true ratio for a programme aimed at women over 35 may

well exceed unity because of non-replacement, because of greater participation by older women, and because the calculated ratios have excluded costs associated with the high morbidity of Down's syndrome individuals.

Comments

Since a general afp screening programme has been established in U.K. is it doubtful that any cases of spina bifida will be detected as a by-product of Down's syndrome screening.

It would seem better to present separate benefit-cost results for women aged 35 to 39 rather than results for all those over 35 since it is the incremental results of extending the programme's coverage which need to be assessed.

The methodology reverses the natural order in computing the costs of care, first calculating "excess costs" of a disabled over a non-disabled child (the "replacement case") and then adding back costs of a non-disabled child (to estimate costs of the "non-replacement" case). The paper does not argue that "replacement" should have priority because it is statistically the norm. The logic of the chosen methodology is perhaps that the non-disabled child is held to be an evaluative norm. The "replacement case" is calculated first because it brings out the comparison with this "normal" level. The "non-replacement" correction, showing the resource costs of the norm itself, is then a tail-piece.

Reference

Hagard, S., Carter, F. and Milne, R.G. (1976) See Summary 4 for full citation.

4. Hagard, S., Carter, F. and Milne, R.G. 'Screening for spina bifida cystica: a cost-benefit analysis.'

British Journal of Preventive and Social Medicine, 30, 40-53 (1976).

Object To estimate the benefit-cost ratio of a proposed afp screening programme for spina bifida in the West of Scotland.

Costs Public and private costs are estimated for a programme covering 43,000 pregnancies a year with a prevalence (open and closed spina bifida) of about 3 per 1000. Public costs estimated from West of Scotland practice include publicity to raise the participation rate, clinic and laboratory costs, ultrasound for dating and diagnosis, amniocentesis and genetic counselling costs. Private costs are lost earnings and travel costs. Rather than annuitise capital costs the authors calculate the present value of the costs of the programme over 20 years.

Outcome 90% participation is assumed. All except those of known high risk have serum afp assayed. 20% require ultrasound to confirm dates, and 10% a retest. 300 serum-positive and 300 high risk women receive diagnostic ultrasound, genetic counselling and amniocentesis. 75 cases of open spina bifida (and 88 of anencephaly) are detected and terminated.

Benefits Survival to five years and degree of handicap of spina bifida infants are estimated from Stark and Drummond (1973) and other studies. Moderately handicapped survivors are

assumed to have a normal life expectancy: others die by age 50.

Excess costs, above the average level for a non-disabled child are calculated. Education and medical costs are estimated from West of Scotland data. Institutional care is assumed for 5% of survivors aged 5, 15% of those aged 30 or more. Additional childhood consumption and social service costs are assumed. The estimate of lost production assumes some 70% of survivors work. It is corrected by an estimate of lower consumption because individuals with spina bifida on average, die younger than others. Lost maternal production is calculated on the basis that 70% of mothers who might otherwise work will not do so.

The benfits of averting the birth of 20 successive annual cohorts are calculated and discounted to give a total present value.

Cost base and discounting

Costs are quoted at July 1974 prices. Discount rates of 5, 10 and 15% are used.

Results

Private costs represent about 20% of the cost of the screening programme. Hospital costs are the largest averted cost item, 20-40% of the total (depending on the discount rate). The overall benefit-cost ratio (at 10% discount) is 1.86. This might fall to 1.44 at a 50% participation rate, or rise to 2.29 if ultrasound were regarded as a free input. There is extensive discussion of how the ratio falls with a lower assumed sensitivity and lower incidence of spina

bifida.

Comments The choice of excess costs as a measure of benefit is not discussed. It may be held that women will in general conceive and bear a non-disabled child after termination of an affected pregnancy, and excess costs are then the net costs averted by screening. On the other hand it may be held that the cost of a non-disabled child is the "normal" level of cost, and that the benefit of screening is properly measured by the extent to which screening averts excess costs, costs above the normal level. (See also "Comments" in Summary 3.)

It would be better to exclude from the benefits of the screening programme the consequences of averting the births of spina bifida children to high risk mothers, since these mothers will be offered amniocentesis even in the absence of general screening.

References

Stark, G.D. and Drummond, E.D. (1973) 'Results of selective early operation in myelomeningocele.' Archives of Diseases in Childhood, 48, 676-83.

Hagard, S. and Carter, F.A. (1976) See Summary 3 for full citation.

5. Mikkelsen, M., Nielsen, G. and Rasmussen, E. 'Cost-effectiveness of antenatal screening for chromosome abnormalities.'

in Scrimgeour, J.B. (ed) Towards the Prevention of Fetal Malformation, Edinburgh, Edinburgh University Press (1978).

Object To estimate the net economic benefit of screening for Down's syndrome in Denmark.

Costs Unit costs, based on the experience of Danish centres, are estimated for amniocentesis and karotyping. The latter is three times the former. Abortion costs are also included, though these are a small element in the total. Total programme costs for Denmark are presented for four age groups - up to 29, 30 to 34, 35 to 39, 40 and over - making use of the prevalence of Down's syndrome in Copenhagen and the maternal age distribution in Denmark.

Outcomes Complete coverage and diagnostic accuracy are assumed. The number of births of Down's syndrome individuals averted in the four age groups would then be 44, 19, 13 and 8.

Benefits Benefits are taken to be the averted costs of care. Local data for survival and the extent of institutionalisation are used. Costs comprise intitutional costs, education and sheltered workshop costs, and cash benefits. The total is therefore the public sector cost averted by averting births.

Cost base and discounting Costs are quoted at 1974-75 prices. A discount rate of 5% is used.

Results

The results show benefits three times cost for women aged 40 and over, and slightly exceeding cost for those aged 35 to 39. However, benefits are only 40% of costs for those 30 to 34, and 15% of costs for younger mothers.

6. Glass, N.J. and Cove, A.R. 'Cost-effectiveness of screening for neural tube defects.'

in Scrimgeour, J.B. (ed) Towards the Prevention of Fetal Malformation, Edinburgh, Edinburgh University Press (1978).

Object To compare the financial savings to the public sector with the costs of a possible alpha-feto protein screening programme in the United Kingdom.

Costs There are assumed to be 650,000 births annually. The proportion of women who attend clinics by the 16th, 18th and 20th week are calculated, to derive the total numbers who would be tested by programmes which cut off testing at these different points. Total programme costs without elemental breakdown are quoted for the different cut-off dates and three different patterns of laboratory provision.

Outcome A national prevalence of open spina bifida of two per 1000 is assumed. 10% of affected pregnancies are assumed to be to "high risk" mothers. These would be offered amniocentesis in the absence of serum screening, and averting these affected births is therefore excluded from calculation. On the assumption that all pregnancies diagnosed defective are terminated, and that diagnostic sensitivity may be 45% or may be 75%, the numbers of births averted by the three possible programmes are calculated. The 18-week estimates show 400 births averted at 45% sensitivity and 700 at 75% sensitivity: 30% and 45% respectively of the total number of spina bifida infants who

would otherwise have been born.

Savings Survival of spina bifida children is taken from Edinburgh data (Stark and Drummond (1973)). Savings to the public sector from births averted are calculated, to include hospital costs, education costs (including residential schools), and cash benefits. Savings are calculated and summed over the 10 years after screening.

Cost base and discount rate These are not given.

Results Public sector savings over 10 years are more than twice programme cost at 45% sensitivity, and more than four times cost at 75% sensitivity. There appear to be economies of scale in testing so that the later the cut off date and the larger the programme the higher the saving/cost ratio.

The paper also quotes costs per case detected. For the main screening programme it is about £1000; for high risk mothers, about one half this level. Adding to the diagnostic procedure an ultrasound examination for women uncertain of dates would cost about £80,000 per additional cost detected. "Assuming no interaction between raised maternal serum AFD and chromosomal disorders" (a wise caveat, since an inverse relation has now been shown) the cost of karotyping all amniotic fluid samples for Down's syndrome would be about £35,000 per case discovered.

Comment The paper gives few details of the methods of calculation.

7. Layde, D.M., van Allmen, S.B. and Oakley, G.P. 'Maternal serum alpha-fetoprotein screening: a cost-benefit analysis.'

American Journal of Public Health, 69, 6, 566-573 (1979).

Object To estimate the monetary costs and benefits of a possible afp screening programme in the United States.

Costs 0.2% of pregnancies are to "high-risk" women. They will be offered ultrasound for dating and for detection of twins or anencephalics if serum afp is greater than the median, followed as necessary by amniocentesis and the offer of termination. "Low risk" women will be offered ultrasound, and amniocentesis and termination as necessary, if serum afp is above 2.5 times the median level. Borderline serum afp will be retested. Costs of the diagnostic procedures, genetic counselling and termination are estimated from out-turn costs of a programme in Maine together with estimates of the numbers, in a national programme, subject to the different procedures. These estimates in turn depend on figures for sensitivity and specificity of the procedures, taken from British and American experience, and on a prevalence of 0.75 per 1000 for anencephaly, 0.9 per 1000 for spina bifida.

Outcomes If prevalence in the screened population is typical of that in the population at large, and if all pregnancies diagnosed defective are terminated, then per 100,000 screened, 59 spina bifida pregnancies will be terminated (of which four are "high-risk") and 35 spina bifida infants will be born.

69 anencephalic pregnancies will be terminated (4 "high-risk") and 10 anenaphalics born. 19 false positive terminations will be carried out.

Benefits

Benefits are estimated as averted costs of care of spina bifida individuals. Survival is estimated from British Data, as are the proportions of survivors requiring institutional and special educational care (see Hagard, Carter and Milne (1976)). Estimates are presented for medical costs, (from American data), institutional and educational costs. The estimate of maternal production foregone assumes that mothers' production is at half that of mothers with non-disabled children. Offset against these savings is the production of spina bifida children, assumed to be 30% of average production. All these elements are assumed to grow by 2.5% annually in real terms. The final averted cost is that of general support at home ("food, clothing, lodging, etc.", set at the everage level of non-disabled individuals). In order to quote net benefits in the "replacement" case costs and production of non-disabled children are also estimated. Costs comprise medical care, education and general support. Absent from the costs of non-disabled children is any element of maternal production foregone.

Discounting and cost base

Costs are quoted at 1977 prices. A discount rate of 7.5% is used.

Results

The gross benefit of averting the birth of a spina bifida infant is $68,000, to which may be added a further resource

benefit of $13,000 if termination is followed by conception and birth of a non-disabled child. The benefit-cost ratio of the programme is 1.95 for "non-replacement" and 2.35 for "replacement".

Comment

It appears that the costs of both disabled and non-disabled children to the rest of society may be understated. The individuals general consumption does not appear as a resource cost; what appears is merely "general support (food, clothing, lodging, etc)". Thus consumption that is not supported in this way be the family but supported from the individual's own production has not been netted off. The value of production, which is taken into account as being a benefit to the rest of society from the existence of the child, thus includes not only production which accrues to society but also an element consumed by the individual - a contribution to the individual's welfare, not to the welfare of the rest of society. It is the principle of the methodology adopted by the study to exclude the welfare of the disabled or replacement child from consideration, and it therefore appears that the methodology has been inconsistently applied. The cost to society of a disabled or of a replacement child has been understated.

This will in part account for the study's finding that non-disabled children are a net resource benefit to the rest of society, while other studies have found them to be a resource cost. Another partial explanation for the difference is that there is no element in the cost of a non-disabled child for lost maternal production.

Reference

Hagard, S., Carter, F. and Milne, R.G. (1976). See Summary 4 for full citation.

8. Hook, E.B. 'Genetic counselling and prenatal cytogenetic services: policy implications and detailed cost-benefit analysis of programs for the prevention of Down's syndrome.'

in Porter, I.H. and Hook, E.B. (eds) Service and Education in Medical Genetics. New York Academic Press (1979).

Object To help make the case for subsidising screening programmes for Down's syndrome locally (New York State) and nationally by considering how far they may be extended to younger mothers while remaining of net economic benefit to society.

Method Programme costs per woman screened are quoted from experience in New York State. Benefits (i.e., costs averted by avoiding the birth of a Down's syndrome child) are taken from Conley and Milunsky (1975) and inflated to 1978 price levels. Combining unit costs with age-specific prevalence of Down's syndrome pregnancy the author estimates a "marginal break-even age". The cost of averting one affected birth to women of this age is equal to the benefit (of care cost averted). A "programme break-even age" is also calculated such that a programme offered to all women of, or older than, this age will have benefit equal to cost. (This measure depends on the maternal-age distribution of pregnancies and on the proportions of women of different ages who choose antenatal diagnosis, as well as on maternal-age specific prevalence of Down's syndrome.) The effect on these two measures of varying assumptions of cost, benefit, and other factors is examined.

Results The plausible marginal break-even age is around 36 years: to limit diagnosis to women above that age will maximise economic benefit. On the most plausible assumptions the programme break-even age is below 30.

Comment It would assist the case for public subsidy if the authors having calculated net economic benefits then considered to whom the benefits accrue, since benefits to the public sector provide a stronger argument for subsidy than do private benefits.

Reference

Conley, R. and Milunsky, A. (1975). See Summary 1 for full citation.

9. Sadovnick, A.D. and Baird, P.A. 'A cost-benefit analysis of prenatal detection of Down's syndrome and neural tube defects in older mothers.'

American Journal of Medical Genetics, 10, 367-378 (1981).

Object To estimate benefit-cost ratios for amniocentesis offered to mothers aged 30 or over in British Columbia (B.C.).

Costs The data presented are the maternal-age specific costs by single year age groups of detecting one affected foetus. ("Affected" means affected by Down's syndrome, spina bifida or anencephaly.) These costs are drawn from Sadovnick and Baird (1982b). That paper calculates the weighted average of the cost of detecting anencephaly by ultrasound (when the main cost is that of genetic counselling) and the cost of detecting spina bifida or Down's syndrome by amniocentesis (when the main costs are karotyping (50% of the total) and genetic counselling (20%)). All these costs are borne by the public sector.

Benefits All estimates are made from B.C. data. An obstetric benefit is taken into account, of the amount by which the cost of normal delivery at term exceeds the cost of a therapeutic abortion. The benefit of avoiding the care costs of spina bifida individuals is quoted from Sadovnick and Baird (1982 a). The benefit of avoiding the care costs of Down's syndrome individuals is calculated as the sums of educational costs, residential care costs and medical costs, the last element comprising cardiac care and repair,

hospitalisation, and physician usage. All Down's syndrome care costs are calculated as the excess cost of a Down's syndrome individual over the average cost of a non-disabled individual. (In the case of residential care costs, the average non-disabled individual is taken to mean the average non-disabled individual in foster care.)

The total benefit of averting the birth of an "affected" individual to a mother of a particular age is then calculated as the sum of the obstetric benefit, plus the averted cost of care for a spina bifida individual times the proportion of "affected" pregnancies affected by spina bifida, plus the averted cost of care for a Down's syndrome individual times the proportion of "affected" pregnancies affected by Down's syndrome.

All benefits are benefits to the public sector: no benefits to mothers and their households are included.

Cost base and discounting

Figures are quotes at 1980 prices. A discount rate of 14% is used.

Results

Obstetric benefits are never as much as 1% of the total. Averted care costs of spina bifida individuals contribute 33% of total benefit from screening mothers aged 30, but only 15% at age 35 and 4% at age 40.

Benefits per affected pregnancy detected exceed costs for all maternal cohorts aged 35 or over. If the benefits of averted costs of Down's syndrome individuals only are considered these exceed costs for maternal cohorts aged 36

or over.

It is pointed out that total benefits of screening will exceed programme costs even if screening is extended to ages lower than these, the defecit of benefits minus costs for younger cohorts being offset by the surplus for those older.

Comment

It is not clear why the "excess" costs of care, over and above the costs of the non-disabled child, are taken as the measure of benefits, rather than full costs. It is not argued that the disabled child will be "replaced".

Whether it is appropriate to include the benefits of averted care for spina bifida individuals depends on the situation being addressed. If there is a separate screening programme for neural tube defects it is inappropriate for them to be included, while it is appropriate to include them if such a programme is not contemplated.

References

Sadovnick, A.D. and Baird, P.A. (1982a). 'A cost-benefit analysis of prenatal diagnosis for neural tube defects selectively offered to relatives of index cases.'

American Journal of Medical Genetics, 12, 63-73.

Sadovnick, A.D. and Baird, P.A. (1982b). Maternal age-specific costs of detecting Down's syndrome and neural tube defects.

Canadian Journal of Public Health, 73, 248-250.

10. Henderson, J.B. 'An economic appraisal of the benefits of screening for open spina bifida.'

Social Science and Medicine, 16, 545-560 (1982).

Object To estimate the economic benefits of averting the birth of 100 children with open spina bifida through the United Kingdom alpha-fetoprotein screening programme.

Resource benefits The benefits are taken to be the increase in resources available to the rest of society due to the disabled child not being born. Initially, these consist of:

(a) The direct care costs avoided.

(b) The increased maternal output available.

(c) The excess of the childs consumption over its production, which represents additional resources available to the rest of society after screening.

U.K. data are used to set out survival of spina bifida individuals. The proportions of individuals receiving different forms of care are taken from Hagard, Carter and Milne (1976) and other sources. Direct costs per 100 liveborn individuals comprise institutional care, medical costs and education. It is assumed that maternal production is affected to the extent that 70% of mothers who might otherwise have worked do not do so following birth of a disabled child. The excess of the disabled individuals consumption over production assumes (a) that consumption is that of a non-disabled child plus £13 per week plus the occasional wheelchair; (b) 50% of males and 60% of females are economically active, and their work patterns and production are similar to those not disabled.

Replacement This initial estimate of the resource effect is then supplemented by considering the resource effects of non-disabled "replacement" children who may be conceived and born after, and as a consequence of, the termination of an affected pregnancy. "Replacement ratios" from 0 to 200% are considered. The resource effect of a non-disabled child has the same elements as that of a disabled child (except institutional care). Despite their higher production they too are a net resource cost. Their net resource effects are appropriately discounted to allow for them being born at some time after screening and termination.

Psychic The overall estimates, including replacement effects, are **benefits** then further compounded by introduction of monetised "psychic benefits" of children. It is argued that non-disabled children bring a "psychic benefit" to parents of value at least equal to a monetary benefit sufficient to offset the monetary costs to the parents (namely, mothers' net lost earnings and the childs consumption to age 17). These psychic benefits directly modify the overall resource effect of replacement children. It is further argued that the psychic benefit of a disabled child may be equal to that of a non-disabled child, or half that figure, or zero: the overall resource benefits of screening are modified accordingly.

Cost base and discounting Costs are quoted at 1979 prices. Discount rates of 4, 7 and 10% are used.

Results Benefits of averting the birth of 100 infants with open spina bifida range from £0.2 m to £2 m: the lower the discount rate and the lower the replacement rate the higher the benefit.

The benefits are compared with costs of averting 100 births of approximately £0.25 m taken from an official study (DHSS (1979)). It is held that on the most plausible assumption, of 100% replacement and 7% discount, benefits exceed costs by £1 m and by a further £0.25 m if psychic benefits are brought into account.

Comments While technically meticulous in estimating the resource effects of screening, the study appears over-refined. When the resource effect has been complicated by alternative replacement effects and psychic benefit, it is not intuitively clear that it is a proper measure of the good done by screening, nor that it can properly be set against simple programme costs.

The programme cost estimate quoted is wrong by a factor of two since the figure of £0.25 m refers to the cost of averting 100 births of infants with neural tube defects, one half of who are anencephalic and die at or very soon after birth.

References

Hagard, S., Carter, F. and Milne, R.G. (1976) See Summary 4 for full citation.

DHSS (1979) Report by the Working Group on Screening for Neural Tube Defects, London, Department of Health and Social Security, (mimeo).

Note

A shorter paper which reports the same study is:

Henderson, J.B. 'Measuring the benefits of screening for open neural tube defects.'

Journal of Epidemiology and Community Health, 36, 214-219 (1982).

11. Sadovnick, A.D. and Baird, P.A. 'A cost benefit analysis of a population screening programme for neural tube defects.'

Prenatal Diagnosis, 3, 117-126, (1983).

Object To estimate the costs and benefits to the public sector of a maternal serum alpha-fetoprotein screening programme in British Columbia (B.C.).

Costs Total costs of a programme to cover 31,000 births annually are estimated, assuming 80% of mothers participate. The main cost elements are test and laboratory costs (75% of the total), administration and publicity (10%) and genetic counselling (13%). The option of karotyping all samples of amniotic fluid is costed as an addition.

Outcome B.C. incidence for anencephaly is 0.68 per 1000, for spina bifida 0.87 per 1000. Assuming 100% detection of anencephalics and 75% for spina bifida the programme would prevent the birth of 21 anencephalics and 20 spina bifida infants (of whom 18 would have been liveborn). If karotyping were carried out, 2 cases of chromosome abnormality would be identified.

Benefits All estimates are made from B.C. data. An obstetric benefit is attached to all averted births, the amount by which the cost of normal delivery at term exceeds the cost of a therapeutic abortion. The benefit of avoiding the care costs of spina bifida individuals is taken from Sadounick and Baird (1982 a). That paper estimates care costs using

B.C. data for survival and the elements of care. These include education, residential care, and medical care. The paper reports the excess cost for these elements, that is the excess cost of the average spina bifida individual above the average cost of a non-disabled child. No estimate is made of the benefit attaching to detection of chromosome abnormalities.

Cost base and discounting 1980 prices and a discount rate of 14% are used.

Results The ratio of benefits to costs was found to be 1.8 to 1, reducing to 1.6 to 1 when the cost of chromosome analysis was included.

Comment No rationale is given for using the "excess" care costs of a spina bifida individual, rather than the full costs, as a measure of benefit.

Reference

Sadovnick, A.D. and Baird, P.A. (1982 a) 'A cost-benefit analysis of prenatal diagnosis for neural tube defects selectively offered to relatives of index cases.'
American Journal of Medical Genetics, 12, 63-73.

12. Andreano, R.L. and McCollum, D.W. 'A benefit-cost analysis of amniocentesis.'

Social Biology, 30, 4, 347-373 (1983).

Object To estimate the net economic benefit of averting the birth of spina bifida and Down's syndrome infants by offering amniocentesis to women of different ages in Wisconsin and in the U.S.A. as a whole.

Costs Programme costs per person for administration-publicity, amniocentesis and diagnosis and terminations are estimated from experience in Wisconsin.

Outcomes Prevalence of the two conditions is based on mid-1970s figures for the U.S.A. (approximately 0.35 per 1000 live births for Down's syndrome and 0.40 per 1000 for spina bifida). The central estimates of the paper assume complete participation, complete diagnostic accuracy, and that all pregnancies diagnosed affected are terminated. Separate calculations are done by 5 year maternal age bands from 25 years old to over 40. A screening programme for women aged 35 or over would cover 3000 pregnancies annually in Wisconsin and 150,000 nationwide, and would avert the births of 360 Down's infants nationally (7 locally) and of 75 spina bifida infants nationally (2 locally).

Benefits Assumptions on survival, employability and the pattern of institutional care follow British sources (Hagard and Carter (1976), Glass (1975), Chamberlain (1978)).

Benefits are the averted costs of care. They include educational, medical and institutional care costs of disabled children, taken from a variety of local (Wisconsin) and national (American) sources. They also include maternal earnings foregone, assuming participation rates for mothers of disabled children one half those of other mothers. In addition, averted consumption of the disabled person is included as a benefit, against which is offset estimated production of the disabled assuming that all those not institutions are in work (those with spina bifida at average productivity and Down's syndrome individuals with lower productivity).

Cost base and discounting 1980 prices are used. The discount rate applied is $3^1/_2$%.

Results On the central assumptions the monetised benefits of screening are found to outweigh costs for women in each single-year age-range from age 36 upwards, or for all women aged 32 or over considered as a group.

The effect of different assumptions is examined by consideration of:

(a) A range of cost assumptions for amniocentesis, administration and termination.

(b) A proportion of women choosing not to terminate affected pregnancies.

(c) Different qualities and costs of institutional care of Down's syndrome individuals.

(d) The employment rate of non-institutionalised individuals being 50 rather than 100%.

The conclusions are quite robust. The least "beneficial" assumptions, high programme costs, low care costs, and low employment, still show a net benefit to women aged 37 or over considered as a group.

Comments

It seems inappropriate to include costs of termination in programme costs since these pregnancies would otherwise proceed to term and delivery costs would be incurred, which have not been presented as an averted cost.

The study uses a discount rate of 3 1/2% which is lower than those of other authors. A higher rate would reduce the calculated value of benefits and increase the maternal age for which benefits outweighed costs.

The authors do not consider the "replacement" case and hold that birth of a non-disabled child would result in a net resource gain to society. Thus their calculations would understate the resource gain from screening if in fact terminations are followed by the conception and birth of a non-disabled child. Whether the resource effect of a non-disabled child is computed as a gain or a loss depends upon the discount rate applied to the child's future production. It is possible that at the level chosen the calculation would show a gain but others authors have used higher rates and have shown a resource loss. If a higher rate is appropriate exclusion of "replacement" children may overstate not understate the resource gain from screening.

The paper includes extensive discussion of the non-monetary costs and benefits of screening and concludes that the major

such effect is "relief from anxiety and reduction in uncertainty". Thus in the author's view the monetary calculation understates the net benefit of screening.

References

Hagard, S. and Carter, F.A. (1976) See Summary 3 for full citation.

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Chamberlain, J. (1978) 'Human benefits and costs of a national screening programme for neural tube defects'. Lancet, 1978 (ii), pp. 1293-1296.